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Case Report

Exophiala jeanselmei as a Rare Cause of Chromoblastomycosis in India: A Case Report

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ABSTRACT

Exophiala jeanselmei, a dematiaceous hyphomycete commonly found in soil, decaying vegetation, and rotting wood is one of the lesser common organisms to be associated with Chromoblastomycosis (CBM). It is more commonly associated with subcutaneous infection such as Mycetoma in patients who are engaged in agricultural activities and in phaeohyphomycosis mostly in patients who have undergone organ transplant. Here, we report a rare case of CBM caused by *Exophiala jeanselmei* in an elderly patient with apparently no predisposing disease condition. *Exophiala* spp. as an etiological agent of CBM is rare esp. in India.

Key words: Chromoblastomycosis, CBM, *Exophiala jeanselmei*, Dematiaceous fungi, Sclerotic body.

Key Messages: *Exophiala jeanselmei* is a common etiological agent associated with subcutaneous infection like Mycetoma and phaeohyphomycosis. However, it is not as commonly associated with Chromoblastomycosis wherein the presence of sclerotic body in direct KOH mount is pathognomic of the disease. Here, we report a rare case of CBM caused by *Exophiala jeanselmei* in an elderly patient with apparently no predisposing disease condition.

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INTRODUCTION

Chromoblastomycosis (CBM) is a chronic cutaneous and subcutaneous fungal infection caused by certain dematiaceous fungi (usually *Fonsecaea, Rhinocladiella, Phialophora*, or *Cladophialophora)*. Histologically, CBM is characterized by the presence of medlar bodies (also known as sclerotic body). Here, we report a case of CBM caused by *Exophiala jeanselmei* in an elderly patient with apparent no predisposing disease condition. *Exophiala jeanselmei*, a dematiaceous hyphomycete commonly found in soil, decaying vegetation and rotting wood is one of the lesser common organisms to be associated with CBM. On reviewing 169 cases published in English literature from India since 1957 until May 2016 by Aggarwal *etal*. only two cases were found to be caused due to *Exophiala* spp. while in majority of the cases *Fonsecaea* spp. (66.1%) was the etiological agent followed by *Cladophialophora* spp. (25.1%) and *Phialophora* spp.(3.9%).[1] Progressive, cutaneous and subcutaneous fungal infection following the traumatic implantation of certain dematiaceous fungi. The disease has worldwide prevalence with predominant cases reported from humid tropical and subtropical regions of America, Asia, and Africa. Diagnosis is often delayed or misdirected either due to poor degree of clinical suspicions or clinical simulation of dermatological conditions. The infection is not uncommon in India and several case reports from the sub-Himalayan belt and western and eastern coasts of India have been published; however, very few have reviewed the cases. We reviewed 169 cases published in English literature from India during 1957 through May 2016, including 2 recent cases from our institute. A tremendous increase in the number of reported cases was noticed since 2012, since which, more than 50% of the cases had been published. A majority of the patients (74.1% Traumatic inoculation of *E. jeanselmei* may be the most common mode of acquisition leading to a variety of subcutaneous infections, including mycetoma, chromoblastomycosis, or phaeohyphomycosis. In immunosuppressed organ transplant recipients, *E. jeanselmei* is the most common dematiaceous fungus associated with skininfections.[2]

Case History

A 70 year old male patient, farmer by occupation, non-diabetic, presented to Dermatology outpatient department with a history of swelling over the left ankle and lower leg region which was accompanied with pain and difficulty to walk for the past 10 years. There was a papular lesion which developed at the antero-medial aspect of left ankle. The lesion was papular to begin with which then progressed into multiple nodules. Finally after few years the lesion developed into a verrucous, cauliflower–like mass. The patient had a past history of myocardial infarction for which he underwent treatment 7 years back. No other significant past medical and surgical history was divulged. There was no history of local trauma which the patient recalled off. Based on

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the clinical presentation, the most probable clinical diagnosis contemplated was chromoblastomycosis, wart, lupus vulgaris, fixed cutaneous sporotrichosis, cutaneous basidiobolomycosis and cutaneous coccidioidomycosis.

A Biopsy sample was taken by Dermatology Department and sent to the Microbiology Department for investigation. Modified Ziehl Neelson staining with the use of 1% and 25 % concentrated sulphuric acid as decolourizer was negative**.** The results of routine bacteria land mycobacterial cultures were also negative. In the mycology section, the sample was directly observed in 40% KOH (Potassium hydroxide) mount preparation which revealed the presence of sclerotic bodies (Figure 1).

The sample was simultaneously inoculated in Sabouraud dextrose agar (SDA), Sabouraud chloramphenicol cycloheximide agar (SCCA) and Brain heart infusion agar (BHIA) for fungal culture. SDA and BHIA were incubated at 37°C while SCCA were incubated at room temperature (25°C-30°C). After 2 weeks of incubation, mould like colonies was observed which matured in 4weeks. Macroscopically, the obverse and reverse appearance of the colonies on SDA is depicted in Figure 2.

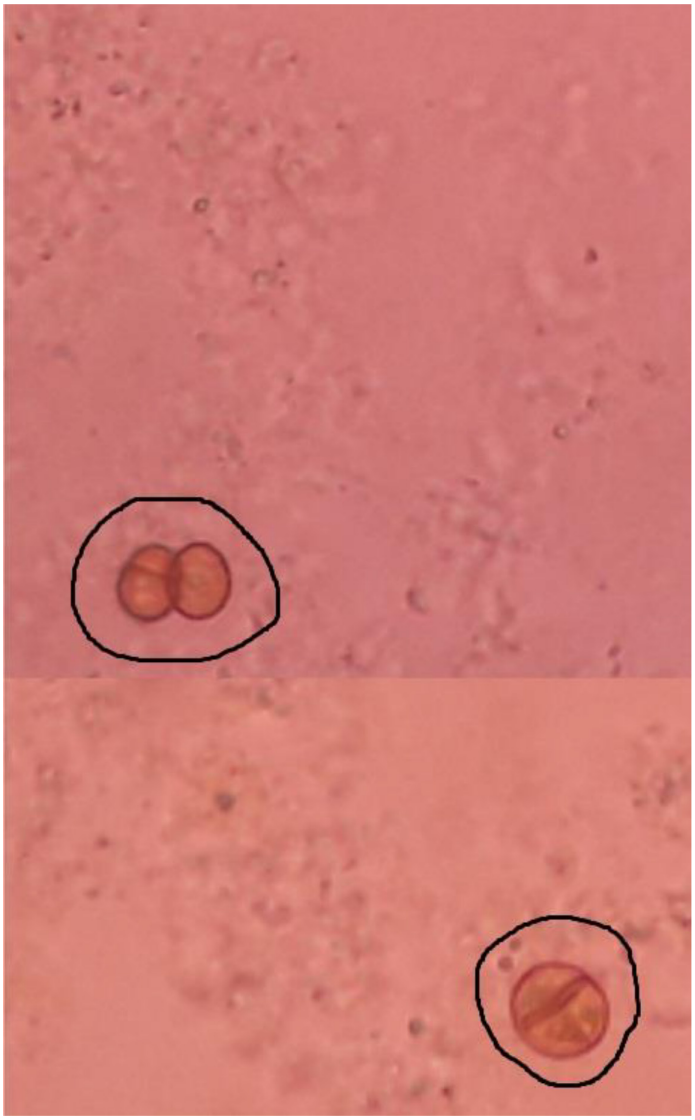


Figure 1: Sclerotic body (Marked within black circle).
Direct microscopy in KOH mount showing Sclerotic Body [marked within black circles] which is seen as dark brown, subglobose, multi-cellular, thick-walled round cells, 6-9µm in size that are seen to be dividing in septal plane by transverse septa).

Figure 2: Macroscopic view - Obverse (Left) and Reverse (Right).
Gross Morphology: Macroscopically, colonies on obverse [left] appeared as black which was velvety in texture with aerial mycelia. On the reverse [right], the colonies were black in colour with no obvious diffusible pigment seen).

Microscopically, on lactophenol cotton blue staining (LPCB), conidiophores with numerous oval shaped conidia clustering at the tip were seen which was consistent with microscopic pictures of *Exophiala jeanselmei* (Figure 3).

The presence of sclerotic body on direct KOH mount and LPCB finding ruled out other differential diagnosis considered earlier.

The patient was started on Itraconazole 200mg twice daily. Hyperthermic therapy with application of towel soaked in warm water was also done. The patient reviewed in the dermatology OPD after a period of two months. There was remarkable improvement noted with considerable decrease in the size of the lesion (Figure 4).

DISCUSSION

Subcutaneous phaeohyphomycosis is a rare infection. However, the number of cases appears to be increasing in recent years as the numbers of immunocompromised patients also have increased. It is more common in tropical and subtropical climates. However, our case showed that immunocompromised state is not a necessary condition for phaeohyphomycosis. Our patient also showed that there was no underlying disease predisposing to chromoblastomycosis which was in

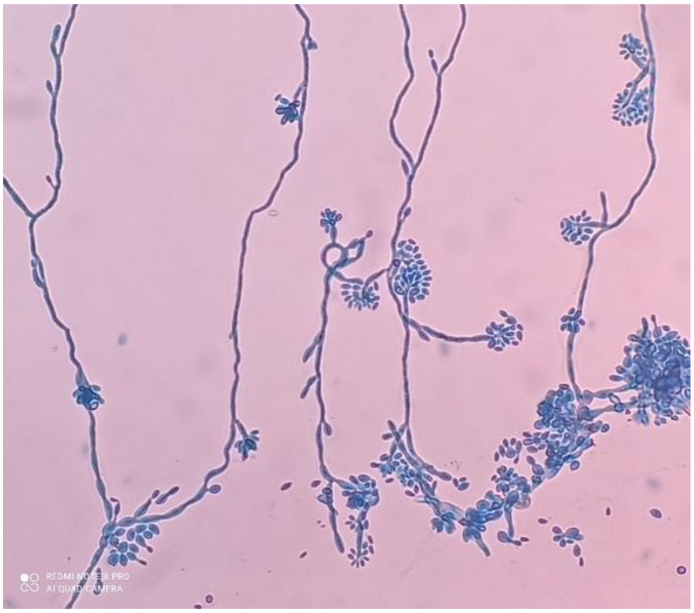
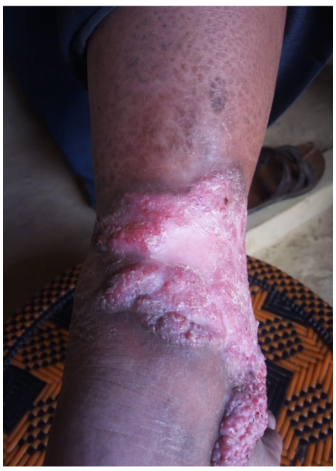


Figure 3: LPCB mount.
LPCB of Exophiala jeanselmei showing brownish septate hyphae forms with numerous conidiogenous cells. The conidia are oval and gathered in clusters and were predominantly present at the end and sides of conidiophores.



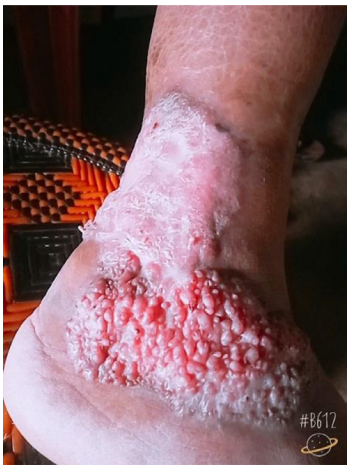


Figure 4(a): Lesion- Anterior view. **Figure 4(b):** Lesion - Medial view.
Figure 4: Chromoblastomycosis patient affected with Exophiala jeanselmei complex after 2 months of treatment with Itraconazole. In anterior view [Figure 4a], the lesion is seen to have regressed considerably. In medial view [Figure 4b], some degree of residual lesion is seen. The size is regressing insidiously with treatment.

concordance with a case reported Chintagunta *et al.*[3] Infection usually occurs through traumatic inoculation of the skin and subcutaneous tissue with contaminated matter, with majority of lesions occurring on the feet and legs of outdoor workers as observed in this patient.[4] The age of patients ranges from 3 to 60 years. However, in our case report the patient was 70 years of age as was seen in one case reported by Neumeister *et al.* where the patient was 73 years of age and was found to have idiopathic CD4 + T-cell lymphocytopenia.[5] The increased age of the patient may be the single most important factor for *Exophiala jeanselmei* manifestation in the absence of any other immunocompromised state. Males are more commonly affected because of their outdoor occupation as was the case in our report where the patient was a farmer. The majority of patient presenting with *Exophiala* spp. infection gave history of localised trauma.[6] However, no such history was divulged in our case as was seen in a case report reported by Chintagunta *et al.*[3]

The common site of involvement is upper and lower limbs over the fingers,[3] toenails,[7] or ankles,[4] and less frequently on the face, neck, and scalp. In this case report, patient presented with lesion over the antero-medial aspect of left ankle which was in concordance with other studies.[4,7] The lesions developed in our study were cauliflower-like. However, there were other studies which showed spectrum of lesions ranging from papulo-nodules to verrucous to ulcerative lesions sometimes presenting with sporotrichoid or dense dermal fibrotic lesions.[3,4,7,8]

It is of interest to note that *Exophiala* spp in general and *Exophiala jeanselmei* in particular, is rarely known to cause CBM.[1,9]progressive, cutaneous and subcutaneous fungal infection following the traumatic implantation of certain dematiaceous fungi. The disease has worldwide prevalence with predominant cases reported from humid tropical and subtropical regions of America, Asia, and Africa. Diagnosis is often delayed or misdirected either due to poor degree of clinical suspicions or clinical simulation of dermatological conditions. The infection is not uncommon in India and several case reports from the sub-Himalayan belt and western and eastern coasts of India have been published; however, very few have reviewed the cases. We reviewed 169 cases published in English literature from India during 1957 through May 2016, including 2 recent cases from our institute. A tremendous increase in the number of reported cases was noticed since 2012, since which, more than 50% of the cases had been published. A majority of the patients (74.1% The presence of sclerotic body in direct KOH examination in our case was pathognomic of Chromoblastomycosis.[10] In studies by Prashant *et al.*[8] Harris *et al.*[11] and Agger *et al.*[7] sclerotic body was not observed in direct KOH examination. Hence, they diagnosed the infection as phaeohyphomycosis. In all of these studies, fungal identification was based on culture and histopathological examination. Our culture examination showed mould-like growth which revealed *Exophiala jeanselmei* on LPCB mount preparation. The present case presented with multiple lesions. The patient was started on Intraconazole 200mg twice daily. There are several recommendations for thetreatment of subcutaneous *E. jeanselmei* infection.[12] The patient reviewed in the dermatology OPD after a period of two months. There was remarkable improvement noted with considerable decrease in the size of the lesion (Figure 4).

CONCLUSION

This case highlights the importance of firstly, considering even rare organism like *Exophiala* spp. asan etiological agent of CBM. Secondly, age may be the single most important predisposing factor in the absence of any immunocompromised condition. Thirdly, Itraconzole may still be a good drug for treatment of cases CBM due to *Exophiala* spp as was seen in our case.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

ABBREVIATIONS

CBM: Chromoblastomycosis; **KOH:** Potassium hydroxide; **SDA:** Sabouraud dextrose agar; **SCCA:** Sabouraud chloramphenicol cyclohex-

imide agar; **BHIA:** Brain heart infusion agar; **LPCB:** lactophenol cotton blue staining.

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