Exophiala jeaneselmei as a rare cause of Chromoblastomycosis in India: A Case Report

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ABSTRACT
Exophiala jeaneselmei, 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 jeaneselmei 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 jeaneselmei, Dematiaceous fungi, Sclerotic body.

Key Messages: Exophiala jeaneselmei 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 jeaneselmei in an elderly patient with apparently no predisposing disease condition.

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 jeaneselmei in an elderly patient with apparent no predisposing disease condition. Exophiala jeaneselmei, 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 et al., 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. (9.3%). A 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. jeaneselmei 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. jeaneselmei is the most common dematiaceous fungus associated with skin infections.

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 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 4 weeks. Macroscopically, the obverse and reverse appearance of the colonies on SDA is depicted in Figure 2. 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 concor-
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. However, no such history was divulged in our case as was seen in a case report reported by Chintagunta et al. The common site of involvement is upper and lower limbs over the fingers, toenails, or ankles, 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. 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. It is of interest to note that Exophiala spp in general and Exophiala jeanselmei in particular, is rarely known to cause CBM. 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. In studies by Prashant et al. and Agger et al. 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 the treatment of subcutaneous 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. In studies by Prashant et al. and Agger et al. 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 the treatment of subcutaneous infection. 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. as an etiological agent of CBM. Secondly, age may be the single most important predisposing factor in the absence of any immunocompromised condition. Thirdly, Itraconazole may still be a good drug for treatment of cases CBM due to Exophiala jeanselmei infection. 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).

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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 cycloheximide agar; BHA: Brain heart infusion agar; LPCB: lactophenol cotton blue staining.

REFERENCES


