CASE REPORT

Chromoblastomycosis: A case report

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ABSTRACT

Chromoblastomycosis is a chronic progressive cutaneous and subcutaneous fungal infection caused by dematiaceous pigmented fungi in tropical and subtropical climates.[1] The most common causative organisms are *Fonsecaea pedrosoi*, *Phialophora verrucosa*, *Fonsecaea compacta* and *Cladophialophora carrionii*. The fungi 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 it usually presents as nodular verrucous lesions.[2] Our patient presented with scaly reddish brownish plaque on dorsum of left hand.

CASE REPORT

A 40-year-old male Pakistani farmer presented with a solitary mass on dorsum of left hand of one-year duration. The condition started with gradual onset and followed slowly progressive course. The lesion was painless and non tender. There was no family history or personal history of the same condition. The patient did not have diabetes or hypertension. Laboratory investigations such as CBC, hepatic and renal profile were within normal range. Markers for hepatitis B & C and HIV were also negative. Skin examination revealed solitary, scaly, crusted reddish brown plaque on dorsum of right hand (Fig. 1). 5 mm punch biopsy was taken from the centre of the lesion under local anesthesia. Histopathological examination showed pseudo- epithelial hyperplasia with horn cyst and follicular neutrophilic suppuration. The dermis showed granulomatous dermatitis formed of lymphohistiocytic cells admixed with plasma cells, giant cells, neutrophils and eosinophils. Dermal focal neutrophilic suppuration were also observed and pigmented fungal sclerotic bodies (medlar bodeis or copper bodies) were detected within dermal neutrophilic suppuration (Fig. 2). Zeihl Neelsen stain for acid fast bacilli was negative. Detection of the pigmented copper penny sclerotic bodies was specific for a diagnosis of chromoblastomycosis. The patient was treated with systemic itraconazole (200 mg/day) and cryotherapy for two months and showed com-

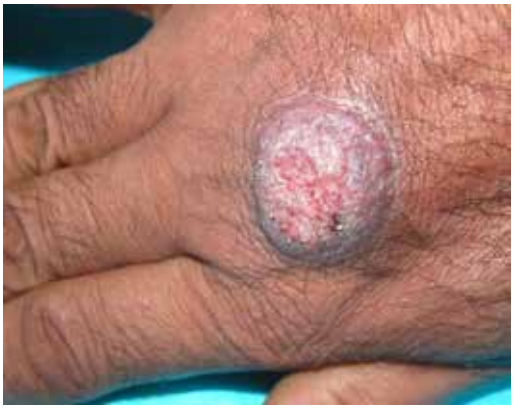

Fig. 1 Reddish brown scaly crusted plaque on dorsum of the right hand.

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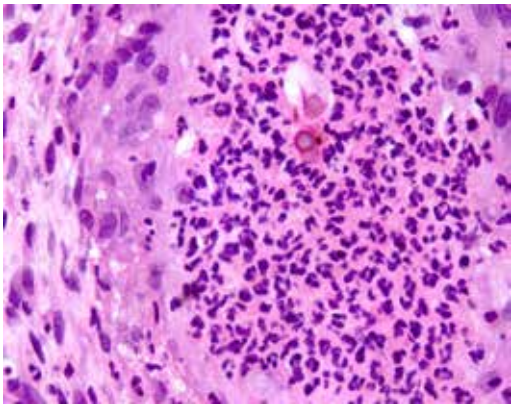

Fig. 2 Pigmented sclerotic/ medlar bodies with copper penny appearance lying freely within neutrophilic microabscess.

plete cure. The patient was followed up for 6 months with no recurrence.

DISCCUSSION

Chromoblastomycosis (CBM) is a chronic, granulomatous mycosis of the skin and subcutaneous tissue produced by the traumatic inoculation of various dematiaceous fungi of the order Chaetothyriales and family Herpotrichiellaceae present in soil, plants, and decomposing wood, prevalent in tropical and subtropical regions of the world.[1,2]

Pedroso and Gomes observed the first cases of CBM in 1911, but it was not until 1920 that the authors published the four cases that reported as having been caused by *Phialophora verrucosa*.[3] However, Brumpt[4] contended that the fungus belonged to a different species, which he named *Hormodendrum pedrosoi*, later renamed *Fonsecaea pedrosoi* by Negroni.[5] According to Castro and Castro.[6-9] the first author to publish was a German physician Max Rudolph,[10] who lived in Brazil, and who in 1914 published six cases of CBM observed in the town of Estrela do Sul, Minas Gerais State. Rudolph emphasized the disease's clinical characteristics, and in four of the six cases he cultured and isolated a brownish-black fungus which he inoculated in animals. There is no record of a histopathology report. In 1915, Medlar[11] and Lane[12] described the first cases of CBM in the United States. Thaxter isolated and classified the fungus from these cases, calling it *Phialophora verrucosa*.[13]

In 1935, as the name "chromoblastomycosis" suggested that the etiological agents display budding yeasts in the tissue, Moore and Almeida (1935) proposed the term "chromomycosis" to replace "chromoblastomycosis".[14] More cases were reported in European countries.[15] The fungus *Acrotheca aquaspersa*, later *Rhinocladiella aquaspersa*, was described in 1972 by Borelli.[16]

The causative organism of CBM belong to the er Chaetothyriales, family Herpotrichiellaceae, and include *Fonsecaea pedrosoi, Fonsecaeamonophora, Cladophialophora carrionii, Fonsecaeanubica, Phialophoraverrucosa, Fonsecaea pugnacius, Rhinocladiella aquaspersa, Cladophialophora samoensis, Cyphellophoraludo viensis, Rhinocladiella tropicalis*, and *Rhinocladiella similis*.[17] Studies on the ribosomal DNA (rDNA) internal transcribed spacer showed that *Fonsecaea pedrosoi* and *Fonsecaea compacta* are identical species.[18]

The most prevalent species (90%) is *F. pedrosoi*.[19] Cases of CBM caused by *Exophiala jeanselmei* and *Exophiala spinifera* have been reported in the literature.[20] In Panama (2007), there is a report of CBM caused by *Chaetomium funicola*.[21]

Clinically, Lesions of chromoblastomycosis are usually polymorphic ranging from ulcer to papulonodular or verrucoid hence clinically mistaken for other lesions.[2-6] 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]

Multi-features of CBM lesions makes differential diagnosis mandatory with pathological processes of different etiologies, including: phaeohyphomycosis, paracoccidioidomycosis, sporotrichosis, lobomycosis (lacaziosis), coccidioidomycosis, North American blastomycosis, leishmaniasis, mycetoma, leprosy, cutaneous tuberculosis, non-TB mycobacterial infections, protothecosis, rhinosporidiosis, botryomycosis, tertiary syphilis, ecthyma, sarcoidosis, psoriasis, halogenoderma, and neoplasms, including squamous cell carcinoma, keratoacanthoma, and sarcoma.

Examination by direct microscopy using potassium hydroxide (KOH) 10-20% or KOH/DMSO reveals muriform (sclerotic) bodies, pathognomonic of CBM regardless of the causative species. Occasional dematiaceous hyphae may be associated with the muriform bodies in the material. The specimens with the highest likelihood of a positive result are those from lesions with the so-called "black dots" that are visible on the lesion's surface, representing transdermal elimination of the fungus. Miranda et al. (2005) used vinyl adhesive tape for the diagnosis of some deep mycoses, including CBM.[22] in our case, examination by direct microscopy using potassium hydroxide (KOH) was negative.

Identification of CBM by Fungal culture in Sabouraud agar is used to isolate and identify species, but the causative agents usually present very similar macromorphological characteristics. *F. pedrosoi* produces velvety, dark-brown, olive-green, or black colonies. *Phialophora verrucosa* produces slow-growing, velvety, moss-green, brown, or black colonies. *C. carrionii* displays colonies very similar to those of *F. pedrosoi*. *R. aquaspersa* colonies are velvety and moss-green to black.[23] In our case, Fungal culture in Sabouraud agar was not done.

Pathology of CBM shows an epidermis with hyperparakeratosis, pseudoepitheliomatous hyperplasia, intracorneal microabscesses, and transdermal elimination of fungi, either inside or outside the microabscesses. The dermis presents dense granulomatous inflammation with different degrees of fibrosis, consisting of mononuclear cells (histiocytes, lymphocytes, and plasma cells), epithelioid cells, giant cells (Langhans and foreign body types), and polymorphonuclear cells. Fungal cells with their characteristic micromorphology - round, dark-brown, thick-walled, 4-12 microns in diameter and with multiplanar reproduction, called muriform (sclerotic) bodies - are found in intraepidermal microabscesses in multinucleated Langhans and/or foreign body-type cells, in suppurative or tuberculoid granulomas, easily identified by hematoxylin-eosin stain.[24] All these histologic features were observed in our case.

Complete surgical excision was considered the treatment of choice for chromoblastomycosis before the advent of triazole antifungal agents. However, currently with the availability of potent antifungal agents, cryotherapy 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 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.[25] Many treatment modalities like cryotherapy, thermotherapy, laser therapy and surgical excision are available.[26,27] Our case was treated with cyrotherapy and itraconazole with complete cure within two months.

Chromoblastomycosis is rare cutaneous disease and should be considered in the differential diagnosis of chronic solitary skin lesion with long duration. Histopathlogical examination is mandatory to confirm the diagnosis so that an early appropriate therapy can be started. Chromoblastomycosis spreads very slowly, rarely fatal, and usually has good prognosis; but is a therapeutic challenge. In our case, a combination of cryotherapy and itraconazole proved to be effective management for solitary lesion of chromoblastomycosis.

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