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Auricular Chromoblastomycosis: A Case Report and Review of Published Literature

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Abstract Subcutaneous chromoblastomycosis is an infection commonly seen in tropical and subtropical climates, usually caused by trauma with vegetables and often affects the host's lower limbs. We report a case of auricular chromoblastomycosis in a 67-year-old man and discuss the rarity of this clinical manifestation of chromoblastomycosis in the medical literature. In the present case, the etiologic agent was *Fonsecaea pedrosoi*, the most common agent found in Brazil.

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Introduction

Chromoblastomycosis is a chronic fungal infection of the skin and the subcutaneous tissue caused by traumatic inoculation of a specific group of dematiaceous fungi such as *Fonsecaea pedrosoi, Cladophialophora carrionii, Phialophora verrucosa, Fonsecaea compacta, Rhinocladiella aquaspersa,* or *Exophiala dermatitidis* through the skin [1]. It is characterized by the development of warty lesions, usually on the foot and leg. The lesions develop over a period of years and usually remain localized; metastases are very rare. The agents often gain entry into the human body by contact with thorns or wood splinters.

The objective of this article is to present a case of auricular chromoblastomycosis caused by *Fonsecaea pedrosoi* in a 67-year-old man and discuss the clinical and therapeutic aspects of this rare manifestation of chromoblastomycosis in the literature.

Case Report

A 67-year-old man presented to consultation referring pruriginous papules on the left auricular surface. He

used to work in rural zone, since his 8 years old. These lesions appeared 3 years earlier this appointment, and the patient had seen another dermatologist at that time. He stated that a biopsy was performed and it was inconclusive. He was treated with topical corticosteroids, but he had a lower response.

Physical examination revealed an erythematous plaque with 30 × 17 mm and few nodules on this auricular surface (Fig. 1). The patient complained of intermittent pruritus and local sting (Fig. 2).

A new biopsy and a fungal culture were performed, and the material was send to the Mycological Laboratory of "Fundação Pele Saudável". The material was cultured on Sabouraud Dextrose Ágar (SDA) with chloramphenicol and Potato Dextrose Ágar (PDA), and after about 10 days, grayish cottony fungal colonies grew out in both media. The bacterial and mycobacterial cultures are negative. PDA, Malt Extract Ágar (MEA), and Oatmeal Ágar (OA) were used to perform microscopic examination.

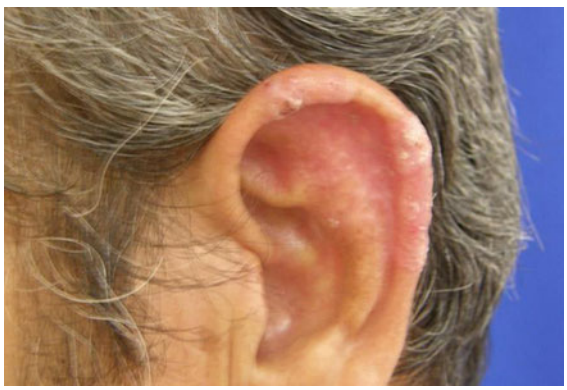

Fig. 1 Erythematous plaque with 30 × 17 mm and few nodules on auricular surface

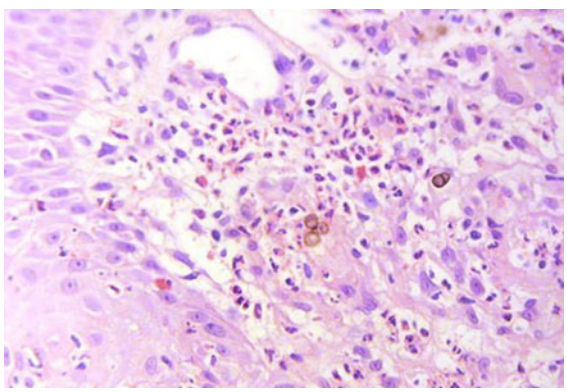

Fig. 2 Biopsy showing pseudo-epitheliomatous hyperplasia of the epidermis, dense infiltration of macrophages constituting granulomas, linfocytes, eosinophils, and neutrophils in dermis. Levedures with a brown pigment inside of the macrophages and giant cells

Abundant septate, dark brown hyphae, and suberect conidiophores that highly branch at apices were observed. The conidiophores were pale brown, erect, septate, and sympodial with conidiogenous zones confined to the upper portion. The conidia were brown and barrel shaped, arranged in short chain, characteristic of *Fonsecaea pedrosoi*, proving the diagnosis of chromoblastomycosis (Fig. 3). Although there is no standard method for in vitro susceptibility testing with *Fonsecaea pedrosoi*, we also performed a susceptibility antifungal test against itraconazole, voriconazole, posaconazole, and amphotericin B.

The susceptibility test procedures were performed according to the Clinical and Laboratory Standard Institute (CLSI) document M38-A2 published in 2008. The MIC value observed was 1,0 μg/ml for all the drugs.

The patient was treated with itraconazole 200 mg/day for 10 months and eight sessions of criotherapy. The patient referred an improvement in symptoms. A clinical remission of nodular lesions was observed. (Fig. 4). The treatment at this clinical location has low response when compared to other areas. The case reported marks improvement but not complete clinical and mycological cure.

Discussion

Chromoblastomycosis is most frequently observed in tropical zones, occurring generally in people engaged in forest or bush clearance. In subtropical regions, it can be observed in rural workers, especially those who do not routinely wear shoes, leading commonly to lower limb involvement [2].

In tropical humid regions, *Fonsecaea pedrosoi* is the principal etiologic agent encountered in chromoblastomycosis but in semiarid climate areas are observed several cases caused by *Cladophialophora carrionii* [3]. Less frequently, the disease is caused by *Phialophora verrucosa*, *Rhinocladiella aquaspersa,* or *Exophiala dermatitidis*. Recently *Exophiala jeanselmei* and *Exophiala spinifera* have also been observed forming muriform cells in lesions of

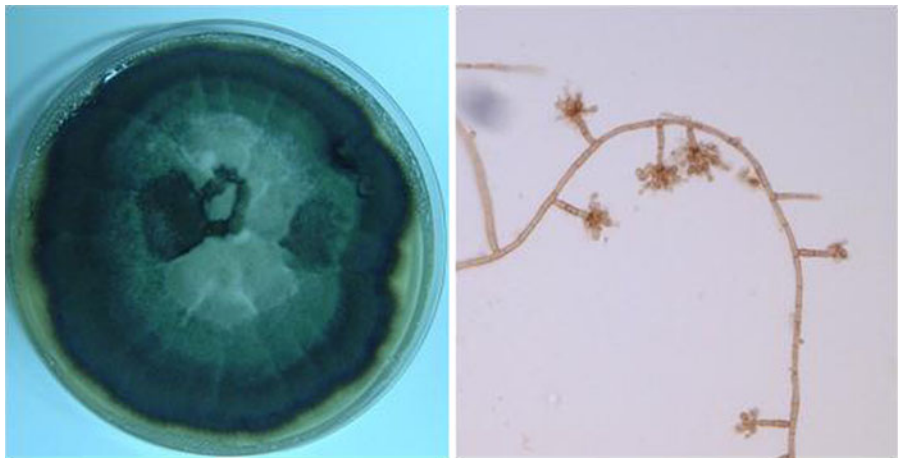

Fig. 3 Fungal culture isolating *Fonsecaea pedrosoi*

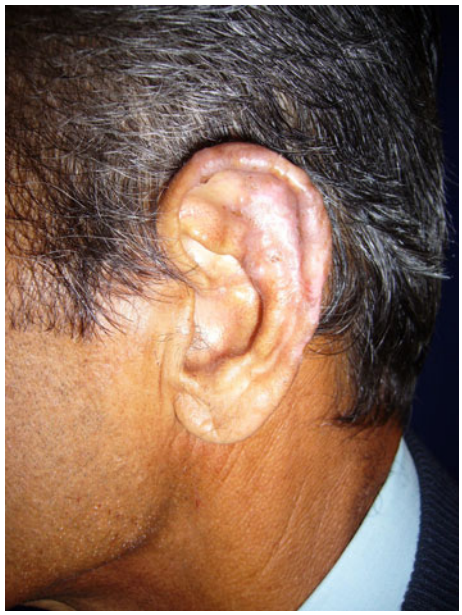

Fig. 4 Patient after 10 months of treatment with itraconazole and 8 sessions of criotherapy

chromoblastomycosis. *Fonsecaea compacta* is an uncommon agent [4].

Chromoblastomycosis should be differentiated from mycetoma, blastomycosis, leishmaniasis, tuberculosis, leprosy, and tertiary syphilis. However, the clinical and microscopic findings are diagnostic [5].

In this patient, the Montenegro's reaction was negative and the amastigote form of leishmania was not visualized, which differentiates from auricular leishmaniasis.

The treatment has been difficult, and a combination of various treatment modalities is sometimes needed to achieve the best result. There are three treatment modalities: physical, chemotherapy, and combination therapy. Their success is related to the causative agent, the clinical form, and also the severity of the lesions [6]. Surgical excision can be done but it requires the removal of a margin of uninfected tissue to prevent local dissemination. Criosurgery with liquid nitrogen is a low-cost, safe, and efficient method [7–10].

Most of the patients can be treated with itraconazole, terbinafine, or a combination of both. Itraconazole showed an excellent profile in the therapy of chromoblastomycosis. It is clinically and laboratorially safe, even when used at high doses and during a long period of time [11, 12]. There is no treatment of choice for this neglected mycosis. In general, it should be guided according to clinical, mycological, and histopathological criteria [6].

Auricular chromoblastomycosis constitutes a rarity in several cases already published. Our MEDLINE search revealed only four cases, all occurred in elderly men [13–16]. In all cases, the lesion was not suggestive and the diagnosis was casual. *Fonsecaea* spp., the most frequent agent isolated, was observed in two cases, and in the other cases, agents involved were *Rhinocladiella aquaspersa* and *Phialophora verrucosa* (Table 1). With the exception of the case published in Japan, we found the lack of information regarding the antifungal drugs used to treat these forms of chromoblastomycosis and also did not

Table 1 Summary of 04 cases of auricular chromoblastomycosis already published

Origin	Genera M/F[a]	Age	Isolated Agent	Duration in years	Treatment (drug/dosage)	Outcome	Reference
Colombia	M	60	*Rhinocladiella aquaspersa*	5	–[b]	–[b]	[13]
Brazil	M	66	*Phialophora verrucosa*	3	–[b]	Improvement of the lesion, but no cure	[14]
Cuba	M	45	*Fonsecaea pedrosoi*	15	–[b]	–[b]	[15]
Japan	M	71	*Fonsecaea pedrosoi*	0,3	Oral Flucytosine/ 7.5 g/day	Complete resolution	[16]

[a] M: Male/F:Female

[b] data not mentioned

obtain information about the success or failure of treatment of these cases.

In summary, our report addresses the clinical aspects of a rare clinical manifestation of chromoblastomycosis.

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