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Bilateral Verrucous Chromoblastomycosis in an Immunocompromised Patient

Lina Pichardo Di Vanna[1*], Mariel Isa Pimentel[1], Karen Reynoso[1], Manuel Cochón Aranda[1], Fernanda Nanita de Estévez[2], Roberto Arenas[3]

[1] Department of Dermatology and Venerology, Dominican Dermatology Institute "Dr. Huberto Bogaert Diaz"
[2] Pathology Section, Dominican Dermatology Institute "Dr. Huberto Bogaert Diaz"
[3] Mycology Section, "Dr. Manuel Gea Gonzalez" General Hospital, Mexico

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ABSTRACT

A traumatic inoculation with dematiaceous fungi results in a chronic granulomatous mycosis of the skin and subcutaneous tissue known as chromoblastomycosis. The majority of those affected are men who work in agriculture, whose primarily exhibit lesions on the lower limbs. We present a 75-year-old immunocompromised male farmer with a bilateral exuberant verrucous chromoblstomycosis caused by Fonsecaea pedrosoi, who did not follow the prescribed treatment at the onset of the clinical condition and returned five years later with excessive growth of the lesions. We report this case due to atypical clinical presentation, affecting both upper limbs.

Introduction

Chromoblastomycosis, a chronic and granulomatous fungal disease, is caused by dematiaceous (black) fungi through skin inoculation of fungal elements. The disease, mainly caused by *Fonsecaea pedrosoi* and *Cladophialophora carrionii,* is prevalent in tropical and subtropical regions [1]. It is one of the most common endemic subcutaneous mycoses, particularly frequent in the north of the Dominican Republic. In some endemic areas, this condition is considered an occupational disease, predominantly affecting workers exposed to contaminated plant materials or soil, such as farm workers and gardeners [2].

This entity predominantly affects the limbs, with the lower limbs being the most common. The disease slowly develops from a small nodule at the site of a past injury, often unnoticed by the patient, and evolves over months or years into large, wart-like, often unilateral plaques. Without early treatment, lesions progressively enlarge and can become nodular with scales and hemorrhagic/black dots, accompanied by pain or itching. Examination of superficial skin scrapings from lesions with a potassium hydroxide (KOH) can confirm the diagnosis. Other diagnostic tools include culture and histopathological study. Chroblastomomycosis is associated with high recurrence rates and low rates of cure [3]. We report a clinical case of an elderly farmer with inadequate metabolic control of his comorbidity.

Case Description

We present a case of a 75-year-old male farmer with poorly controlled diabetes that initially exhibited lesions on the right hand for the past year, characterized by erythematous plaques with adherent whitish scales and black dots on their surface (Fig. 1).

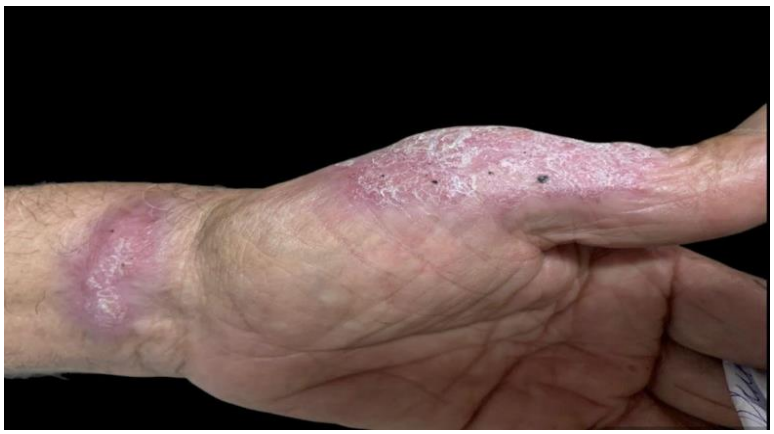

Figure 1

Direct examination with potassium hydroxide (KOH) revealed numerous fumagoid cells. The histopathology revealed hyperkeratotic epidermis, neutrophil collection, marked acanthosis, and irregular elongation of the rete ridges. In the superficial and mid dermis, fumagoid cells were present. A diagnosis of chromoblastomomycosis was made, and treatment with itraconazole 200 mg/day was initiated. The patient did not return for follow-up care until five years later. Currently, the patient returns due to a worsening clinical condition. On physical examination, the patient presents verrucous plaques with papillomatous projections, white and yellow scales and scattered black dots affecting both hands (Fig. 2 and 3).

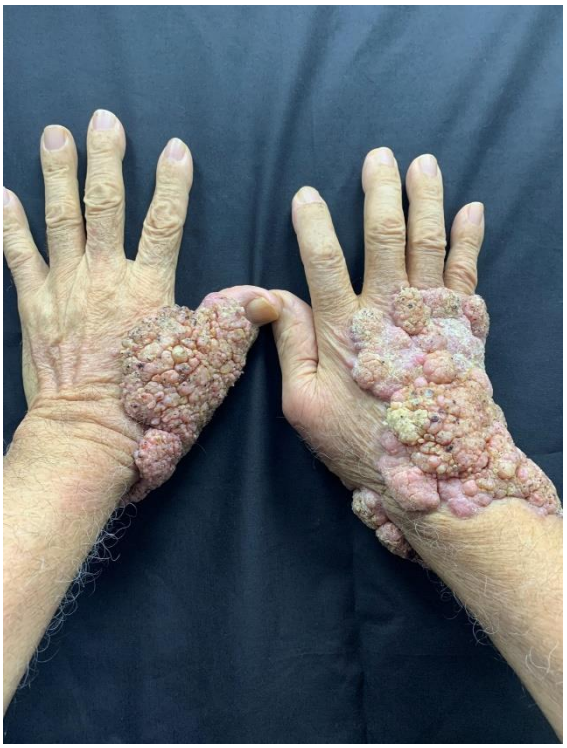

Figure 2

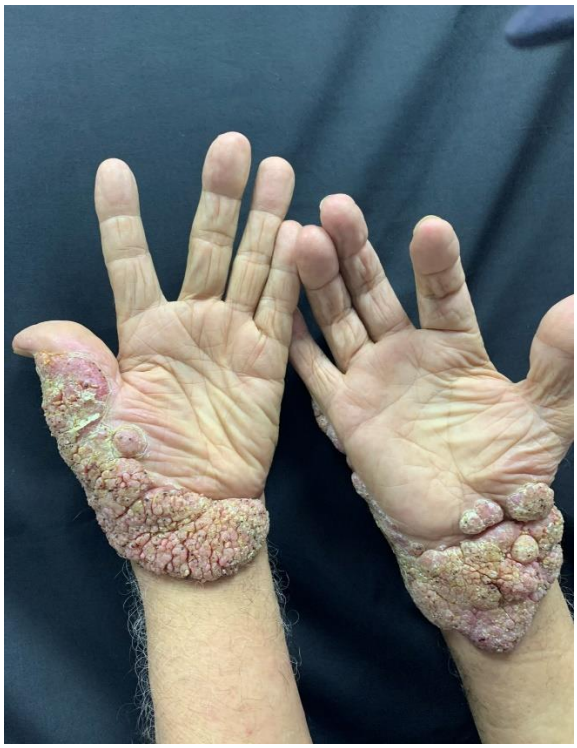

Figure 3

Dermoscopy revealed numerous yellow scales with marked hyperkeratosis, areas with black/hemorrhagic dots, and whitish fibrotic areas (Fig. 4).

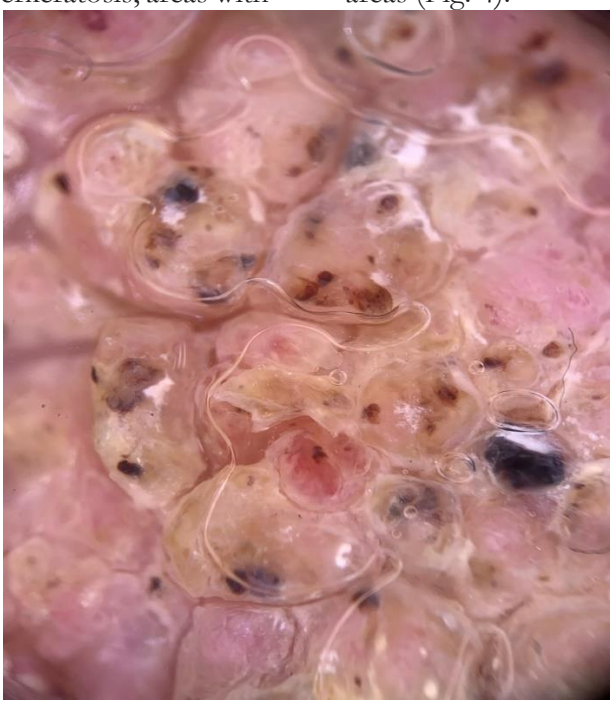

Figure 4

Molecular biology revealed that infection was caused by *Fonsecaea pedrosoi*. Due to the prolonged absence and poor metabolic control of his underlying condition, the patient exhibited excessive growth of plaques into exophytic lesions. Currently, he's being treated with 200 mg of itraconazole per day,

with partial improvement of the lesions (Fig. 5 and Fig. 6).

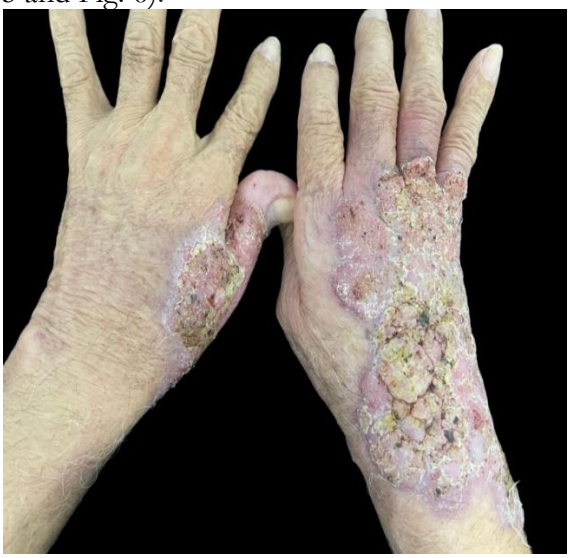

Figure 5

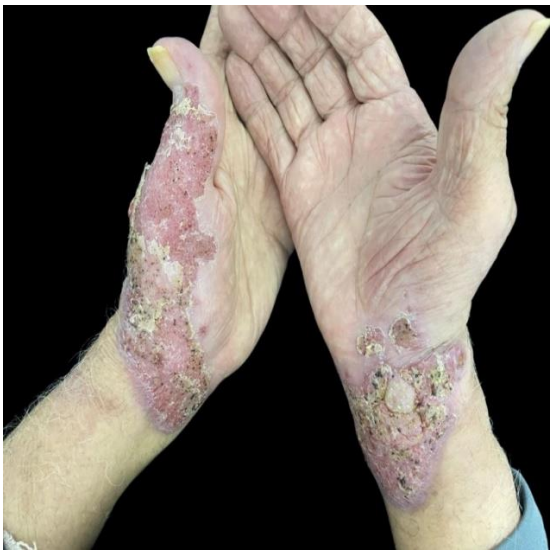

Figure 6

Discussion

Fonsecaea pedrosoi and *Cladophialophora carrionii* are the two principal etiological agents worldwide [1,5]. Chromoblastomycosis occurs in tropical and subtropical climates and is one of the most common endemic implantations (subcutaneous) mycoses, representing the first place in frequency among our Dominican population, followed by mycetoma and Phaeohyphomycosis. Madagascar is the country with the highest number of reported cases, followed by Brazil, Costa Rica, Dominican Republic, India, and Mexico. Its entry route is cutaneous by inoculation through a breach in the skin, mostly secondary to trauma [6].

This infection most commonly affects males. Chromoblastomycosis is considered an occupational disease in certain endemic locations, such as our country. There is a higher predisposition to affect men, with statistics ranging from a 4:1 to 17:1 male-to-female ratio. Besides the association of the affected sex with occupational exposure, it is believed that females may have a protective hormonal factor that could influence the adaptation of the fungus. Ethnic group does not confer a predisposition; however, it has been linked to heightened vulnerability to the human leukocyte antigen HLA-A29 [3,4].

The usual clinical topography of chromoblastomycosis is on the limbs (95%), most commonly on the lower limbs (75%). Involvement of trunk, eyelids, nose, or ears has also been reported. The disease develops over years. Following trauma, weeks to months later, the site of inoculation shows signs of infection. Patients may not remember the initial injury event. Based on its diverse appearance and clinical presentation, the condition is currently classified clinically into nodular, verrucous or vegetant, tumoral, cicatricial, and lymphangitic (sporotrichoid) variants. Mixed lesions are frequent. The initial lesion starts at the site of inoculation as a small nodule that grows slowly to form extensive wart-like plaques, which tend to be asymmetrical and unilateral [3]. There are limited cases of bilateral chromoblastomycosis documented in medical literature, and even more exceptional are the bilateral cases in the upper limbs, as in this patient case from Dominican Republic. If early diagnosis and treatment are not received, lesions show a tendency to grow over months and years,

progressing to large-sized plaques, evolving into nodular lesions covered with many whitish scales and hemorrhagic/black dots along their surface [7], similar to what happened to our patient, who reports that the initial manifestation began on the hands with discrete erythematous plaques with scaling and black dots on the surface, subsequently showing a tendency towards exaggerated growth over time into verrucous and hyperkeratotic lesions, as a consequence of not following the prescribed treatment at the initial consultation.

Conventional techniques for diagnosing chromoblastomycosis include direct, histopathological, and fungal isolation techniques. Observing muriform cells—which are pathognomonic for chromoblastomycosis —is the primary objective of both direct microscopy and histology. Histopathologically, pseudoepitheliomatous epidermal hyperplasia, hyper-parakeratosis, microabscesses, and irregular acanthosis alternate with areas of atrophy is seen in chromoblastomycosis. Usually, the dermis has dense, granulomatous inflammation with variable degrees of fibrosis. Mononuclear cells, such as giant cells, epithelioid cells, and polymorphonuclear cells, as well as lymphocytes, plasma cells, and histiocytes, are associated with this inflammation. The muriform cell, which can be observed alone or in clusters when routine hematoxylin and eosin staining is applied, is the hallmark histopathologic finding. In general, especially in long-term and severe cases, this entity is linked to high relapse rates and low cure rates [2,4].

Authors suggests that significant fibrosis occurs in the dermis and subcutaneous tissue because the fungi locally produce pyridinoline, a substance that induces the formation of cross-links in the collagen bundles of the tissues. It is important to emphasize that fibrosis is a process that hinders drug penetration and directly affects the low response to treatments. Due to the chronic nature of the condition, it tends to progress, leaving scars with achromic areas in the central region of the lesions [3].

Effective treatment requires a comprehensive evaluation of the causative fungus, the severity and spread of the infection, lesion location, and the overall health of the patient. Treatment typically involves prolonged antifungal therapy alongside physical interventions such as cryotherapy, laser, photodynamic therapy, and surgical removal. The latter is particularly effective for small, localized lesions, with excision recommended for all initial, small, and well-defined skin lesions. This can be augmented with antifungal medications like itraconazole or terbinafine [2,3].

Several antifungal drugs are available for treatment, including 5-fluorocytosine, 5-fluorouracil, thiabendazole, amphotericin B, ketoconazole, fluconazole, itraconazole, terbinafine, and posaconazole. The most effective treatments often involve itraconazole (200–400 mg/day) and terbinafine (500–1000 mg/day) for a duration of 6–12 months. Combining itraconazole or terbinafine with cryosurgery or local heat application often yields the best outcomes. For patients with resistant forms of the disease, a combination of itraconazole and terbinafine is commonly used [2-4].

Conclusion

A clinical case of a long-standing bilateral verrucous chromoblastomycosis in a Dominican farmer was presented. The causative agent, *F. pedrosoi*, was identified through molecular biology. One of the best therapies for this condition is itraconazole, which our patient has been receiving at a rate of 200 mg per day for the past few months, with considerable improvement. Ongoing monitoring over time is crucial to assess the patient's progress and evaluate the need to combine therapies in case of no response to the prescribed treatment regimen.

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