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Case Report

A Case Of Chromoblastomycosis - Clinical Management And Outcomes.

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Abstract

Chromoblastomycosis (CBM) is a rare, chronic fungal infection affecting the skin and subcutaneous tissues, predominantly encountered in tropical regions. This case report describes a 55-year-old male from Northern Europe who developed CBM following a rose thorn injury. Initially misdiagnosed as lichen simplex chronicus, the lesion progressed over two years, eventually requiring a biopsy that confirmed CBM through the presence of pathognomonic fumagoid cells. Treatment involved a combination of antifungal therapy with Itraconazole and surgical excision, followed by skin grafting. Despite initial graft failure due to a bacterial infection, a subsequent split-thickness skin graft healed successfully. This case underscores the importance of considering CBM in differential diagnoses of chronic verrucous lesions, even in non-endemic regions, and highlights the need for a multidisciplinary approach in management. The report also emphasizes the challenges in early diagnosis and the potential for recurrence, advocating for long-term follow-up and innovative treatment strategies.

INTRODUCTION

Chromoblastomycosis (CBM) is a rare, chronic fungal infection affecting the skin and subcutaneous tissues, predominantly encountered in tropical regions. Recognized by the World Health Organization (WHO) as a Neglected Tropical Disease, CBM is the second-most prevalent implantation mycosis globally with Sporotrichosis being the most prevalent (1,11). The most frequent causative organisms are melanized fungi, particularly Fonsecaea pedrosoi and Cladophialophora carrionii (2,3). CBM is often categorized as an occupational disease due to its prevalence among farmworkers, hunters, and miners. However, just walking barefoot is a risk factor. A higher incidence is observed among males (2,4). Diagnosis of CBM relies on a combination of clinical presentation, residence or exposure in endemic regions, and laboratory investigations including biopsy for culture. Microscopic evaluation often reveals pathognomonic fumagoid cells (alt. muriform cells, sclerotic bodies, or Medlar bodies) (5).

ETIOLOGY AND CLINICAL PRESENTATION

CBM was first described in Brazil in 1914 (6). The disease progresses slowly, initially manifesting as dermatophyte-like infections or small papules, which gradually evolve into plaques, nodules, verrucous, or exophytic lesions over time. If left untreated, these lesions can develop into tumorous, cauliflower-like masses. Malignant transformation into squamous cell carcinoma has rarely been documented. However, CBM generally remains confined to the superficial skin and subcutaneous layers, sparing underlying muscle and bone unless the patient is immunocompromised (8,9). The differential diagnosis for verrucous lesions includes leishmaniasis, tuberculosis, and verrucous carcinoma (7). Globally, a total of 7,740 cases of CBM were reported between 1914 and 2020. However, the incidence rate remains unclear, underscoring the need for local epidemiological studies to enhance understanding of its prevalence (10).

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CASE PRESENTATION

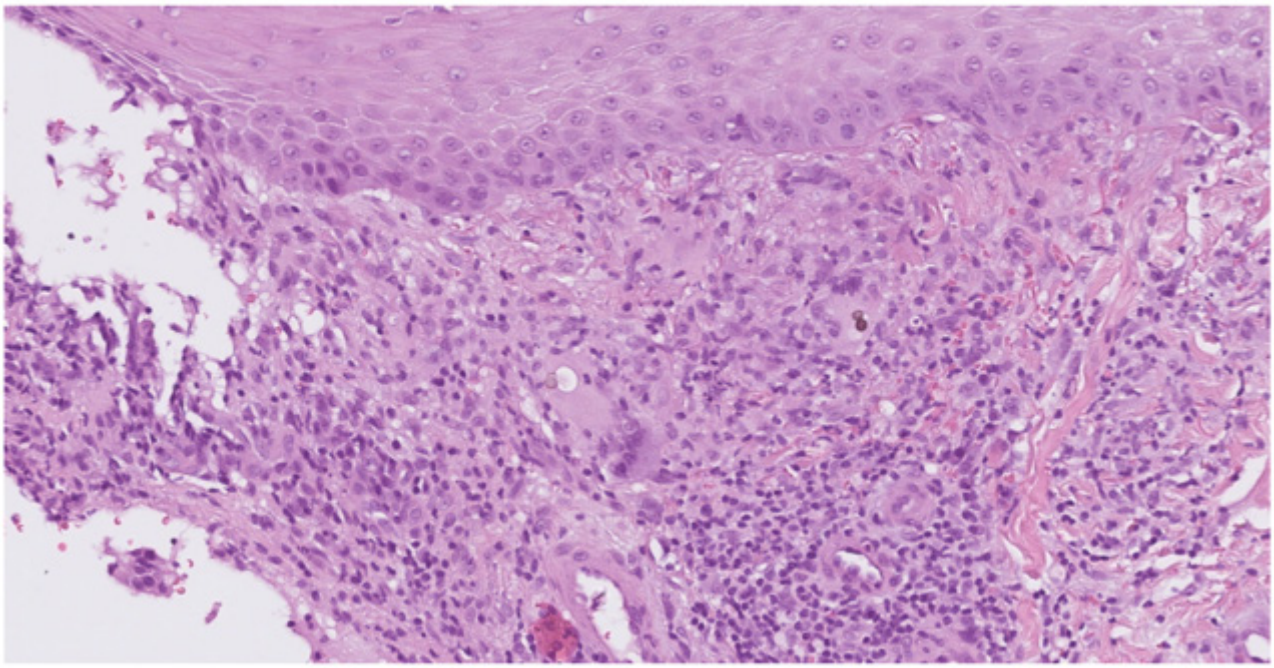
A 55-year-old male was referred to a dermatologist's office in January 2024 for a gradually increasing asymptomatic papule on the lower left leg (**fig. 1**). About 18 months prior, the patient recalls sustaining an injury to the area from a rose thorn while gardening. The dermatologist noted a lichenified red-purplish plaque that on exam was not suspicious for skin cancer, suggested the diagnosis of a solitary plaque of lichen simplex chronicus, and prescribed clobetasol ointment for four weeks. A follow-up with biopsy was recommended in case the lesion did not clear.

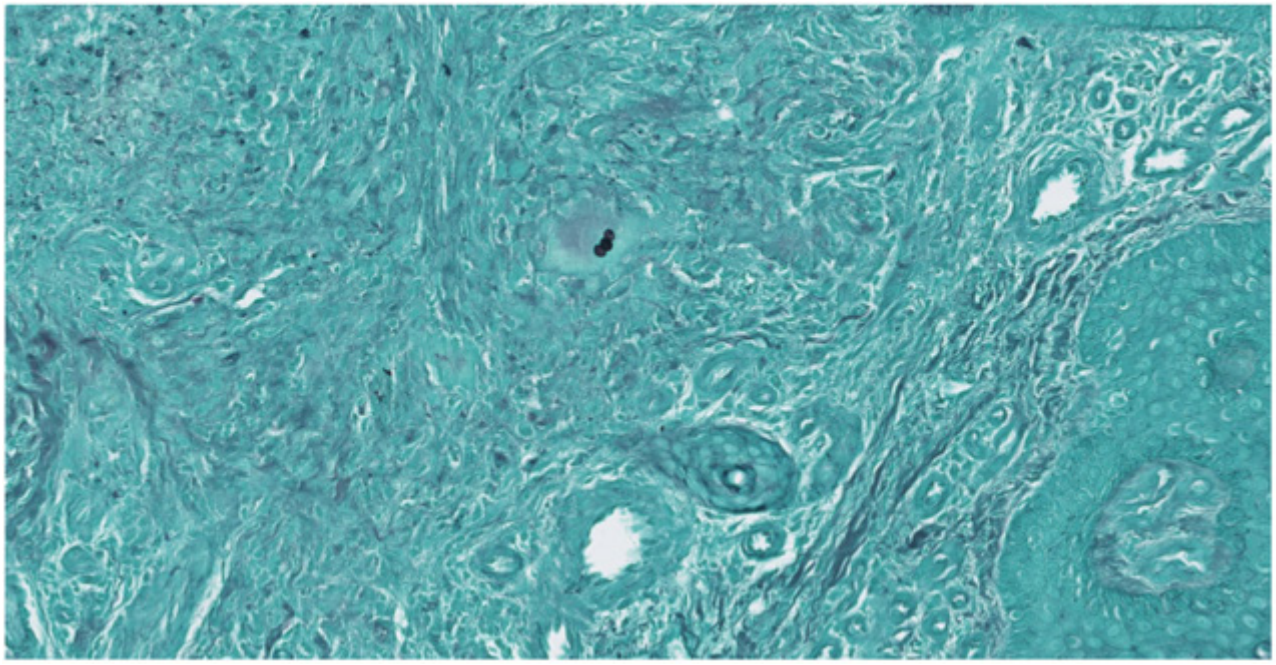
Figure 1. Clinical findings (photos by dept. of Plastic surgery, Aalborg Univeristy hospital).



Two months later - about two years after the initial injury - he attended follow-up and a 4 mm punch biopsy was performed. The biopsy revealed multinucleated giant cells containing pigmented sclerotic bodies, known fumagoid cells and pathognomonic for CBM (**Fig. 2**). Subsequent microbiological analysis confirmed the diagnosis of chromoblastomycosis (CBM).
To our knowledge, there is no established evidence-based consensus for management of CBM.

Figure 2. Microscopy, (by dept. of pathology, Aalborg University hospital).





Histopathologic examination, with hemotoxylin-eosin stain, revealed hemorrhagic and pustular crust and an epidermis with acanthosis, para-hyperkeratosis and microabscesses. In the dermis, numerous inflammatory cells were seen, among those a couple of giant cells. Extracellularly and within these giant cells, pigmented sclerotic bodies (Fumagoid cells), were identified, which was further highlighted on Grocott's methenamine silver stain.

Due to its rarity in the Northern hemisphere and in Europe in particular, the case was discussed with an expert from Brazil, who recommended a treatment of 200 mg Itraconazole twice daily for three months, followed by surgical excision. This treatment regimen aligns with the approach used by Valentin et al (1).

Excisional biopsy was performed with a 5 mm margin around the lesion limiting depth to just above the fascial layer. The excised specimen was divided in three equal parts for further laboratory analyses:

1. Formalin-fixed and paraffin embedded for standard H&E,
2. suspended in sterile 0,9% NaCl solution for culture and susceptibility testing
3. PCR analysis respectively for fungal species identification.

The excision resulted in a defect unsuitable for primary closure and required a full-thickness skin graft (FTSG) and adjunctive negative-pressure wound therapy. At the follow-up visit, the graft appeared viable and healthy. Unfortunately, a few days later, the patient presented with a greenish discoloration across the graft, which ultimately led to graft failure. Microbiological investigations revealed Staphylococcal infection, but no signs of fungal infection. A second surgery was performed using a split-thickness skin graft (STSG), which healed successfully. No recurrence at follow-up visits with dermatologist in October 2024.

DISCUSSION

No cases have been published out of Northern Europe, and only a few from France and Poland over the last 20 years (12,13). The clinical presentation, diagnostic process, and management were, however, consistent with what is commonly observed in CBM (1). Trauma, in this case from a rose thorn, is a well-documented mode of inoculation for the fungi responsible for the condition (1). The gradual progression from a papule to a larger verrucous plaque-like lesion aligns with the characteristic course of CBM (7). Early diagnosis is often challenging due to the non-specific nature of initial lesions, which can resemble other dermatologic conditions, such as dermatophytosis or warts. Histopathological examination remains crucial in confirming the diagnosis, particularly identification of the pathognomonic fumagoid cells (5).

The management of CBM typically involves a combination of antifungal therapy and surgical intervention, especially in larger lesions. Surgery remains a viable option for localized disease, as demonstrated in this case, where wide excision and subsequent skin grafting were ultimately successful (2,9). The initial failure of the FTSG, marked by the development of a green discoloration, which presumably was ascribed to Pseudonomas Auerginosa infection. Furthermore, the progression to the failure could have been impacted by the thickness of the FTSG or poor vascularization of the graft.

The recurrence of CBM following treatment is a well-recognized challenge. Despite surgical excision and grafting,

relapse rates remain high, and the disease often proves refractory to standard antifungal therapies (1,9). Long-term follow-up is therefore essential to monitor for recurrence, particularly given the recalcitrant nature of the infection. The successful healing of the STSG in our case without recurrence is a fortunate outcome, though continued surveillance is warranted. Follow-up was planned every three months for a year. By the 6-month follow up period, the patient had no remarks about the affected area on the limb.

This case underscores the critical importance of a multidisciplinary approach in managing CBM, particularly given the rarity and complexity of cases occurring outside tropical regions. Although CBM is commonly found in tropical areas, this case involving a man in Northern Europe without a travel history highlights the potential for the disease to emerge in non-endemic areas, possibly due to changing environmental or travel patterns. The collaboration between dermatologists, microbiologists, and plastic surgeons was pivotal in achieving a favorable outcome, demonstrating the value of integrated expertise for effective management. The insights gained from this case also emphasize the need for ongoing innovation in antifungal therapies and novel treatment strategies to reduce recurrence rates and improve long-term outcomes. These advancements are crucial not only for endemic regions but also for non-tropical areas where CBM may become increasingly prevalent as a public health concern.

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