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Case Report

From diagnosis to treatment- Navigating Chromoblastomycosis

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ABSTRACT

Chromoblastomycosis is a chronic fungal skin infection caused by dematiaceous fungi, characterized by nodular skin lesions and the presence of sclerotic bodies in affected tissues. Here, we report a case of chromoblastomycosis from Karnataka, India. A 22 years old female patient presented with a history of a vegetating ulcer with crusting. The diagnosis of chromoblastomycosis was made by demonstration of sclerotic bodies on histopathology of the biopsy specimen. Presence of pigment, multinucleate giant cells and eosinophils were the clues to search for the dematacious fungi on histopathology. High degree of suspicion by the clinician and the pathologist is important for definitive diagnosis.

1. Introduction

Chromoblastomycosis (CBM) is a dematiaceous fungal infection involving the deeper layers of the skin, responsible for a chronic and progressive lesion.[1] Dematiaceous fungi are peculiar in their ability to deposit melanin in their cell walls. Traumatic injury is frequently the cause and it commonly affects agricultural workers. Fonsecaea pedrosoi, Phialophora verrucosa, and Cladophialophora carrionii are the most common etiological agents. The lesions are polymorphous in nature, beginning as a small papule and progressing to hyperkeratotic plaques, verrucous nodules, or deep fungating ulcerations, making it a difficult clinical diagnosis. CBM is distinguished by "sclerotic bodies," which can be seen in a potassium hydroxide (KOH) mount and biopsy.[2] Diagnosis is frequently delayed due to clinical simulation with other dermatological disorders. Similarly, at histopathology, the diagnosis can be missed in the absence of high level of suspicion. Despite being known for almost a century and availability of several treatment modalities, CBM continues to represent a therapeutic challenge due to its resistive character and frequent recurrences.[3]

2. Case Report

A 22-year-old housewife, residing in Karnataka, presented to the Dermatology department with complaints of progressive papulonodular lesions on the right foot for 9 years. She gave history of thorn prick while walking barefoot while she was in Rajasthan 9 years back. Lesions developed few months after the thorn prick. Initially, there was a single erythematous to brownish raised lesion 5 x 5 mm in size on the instep of the right foot, which gradually increased in size and number. There was history of discharge of white grains on and off from the lesions. There was also repeated history of pus discharge

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which used to subside after a course of antibiotics, but the lesions remained static. There was increase in the size of swelling on prolonged exposure to water. On examination, multiple indurated hyperpigmented nodules and plaques with few erosions, present over the instep of right foot (Figure 1). Popliteal and inguinal lymph nodes were not palpable. Systemic examination was normal. Haematological and biochemical parameters were normal. The patient was immunocompetent and non-diabetic. Chest X-ray was normal. X-ray of the right foot showed soft tissue swelling. USG of the right foot showed a well-defined heterogenous predominantly hypoechoic lesions (Conglomerated) measuring 2.1×2.3cm involving plantar aspect of right foot taking significant vascularity on applying colour doppler. Punch biopsy was done from the lesion and histopathology revealed pseudoepitheliomatous hyperplasia, inflammatory infiltrates predominantly plasma cells, lymphocytes and few eosinophils seen in superficial and deep, interstitial, perivascular and periadnexal location (Figure 3A). Multinucleate giant cells were also noted. A brown colour pigment was seen extracellularly as well as within the multinucleate giant cells in the dermis (Figure 3C). Also seen were small 1 to 2 micron round brown-coloured bodies, singly and in aggregates, with peripheral dark rim and central pale area. These bodies were morphologically consistent with “copper penny” bodies of Chromoblastomycosis (Figure 3B). PAS stain highlighted the pigmented fungal cells with appropriately reactive controls, suggestive of a deep fungal infection.(Figure 3D) Fungal culture was positive. Bacterial culture revealed Methicillin Resistant Staphylococcus aureus. Based on history, clinical examination, Fungal culture, bacterial culture and histopathological examination, a diagnosis of Chromoblastomycosis with secondary bacterial infection by Methicillin Resistant Staphylococcus aureus was made. Patient was treated with Tab Itraconazole 200 mg twice daily, Tab Cotrimoxazole (Trimethoprim + Sulphamethoxazole) 160/800 mg twice daily, Fusidic acid cream, Luliconazole cream. On follow up after 4 weeks, the lesions reduced in size.(Figure 4) The patient is still under treatment and on regular follow up.

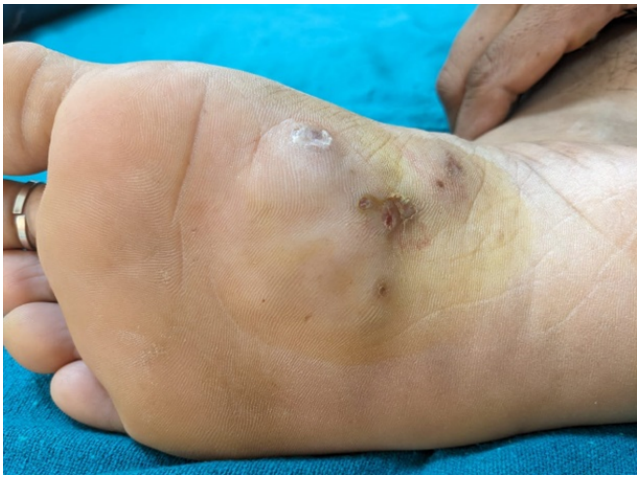

Figure 1: Initial presentation- Multiple indurated hyperpigmented plaques and nodules with few erosions and crusts over the instep of right foot

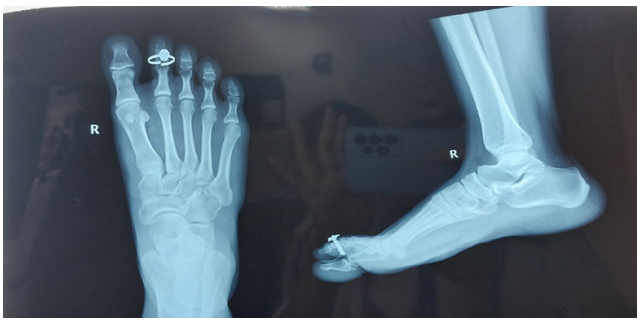

Figure 2: X-Ray of Right foot showing soft tissue swelling.

3. Discussion

Chromoblastomycosis is a chronic cutaneous and subcutaneous fungal infection arising as a result of traumatic implantation of dematiaceous fungi of skin.[4] It was first described by Lane and Medlar in 1915 in the United States. People who live in high levels of poverty are more likely to contract the disease. Although most patients do not remember an initiating trauma, there is a clear relationship between this condition and agricultural labourers who come into touch with colonised wood or soil. Most often, the lower legs are involved. Due to some species of pigmented fungi producing melanin that can be seen under a microscope in their cell walls, these organisms are also known as "dematiaceous" or "melanized" fungi.[2] The common aetiological agents are the following species: Fonsecaea pedrosoi, Cladophialophora carrionii and Phialophora verrucosa. Exophiala dermatitidis and Rhinocladiella aquaspersa are less frequently reported. Exophiala spinifera and Exophiala jeanselmei, which cause phaeohyphomycosis, frequently produce muriform cells. Other species that may cause CBM are: Fonsecaea nubica, Phialophora richardsiae and Fonsecaea monophora.[4]

Subcutaneous fungal deposition causes macrophages and Langerhans cells to engage in a series of granulomatous inflammatory reactions. According to D’Avila et al, verrucous lesions have an IL-4 and IL-10 predominance, which indicates a Th2 response. Additionally, they noticed a prevalence of TNF-alpha and IFN-gamma in atrophic lesions with well-formed granulomas, indicating a Th1 response.[5] It is prevalent in subtropical and tropical areas. Clinical variants of CBM include: nodular, verrucous, tumoral, plaque and cicatricial types.[6]

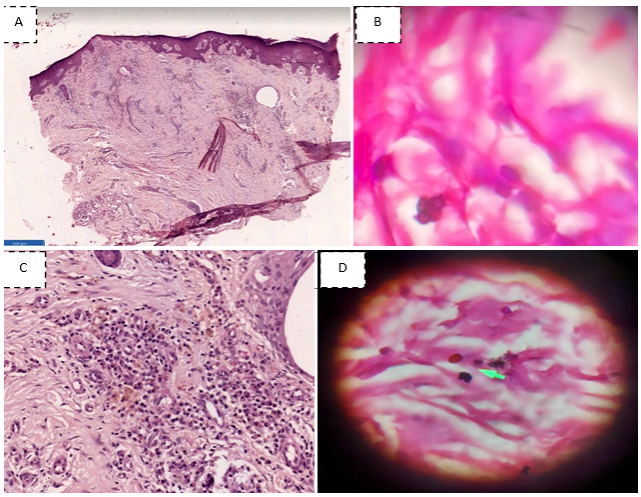

Figure 3: H&E staining of biopsy- **A:** Epidermis shows Pseudoepitheliomatous hyperplasia. Dermis shows inflammatory infiltrates predominantly plasma cells, lymphocytes and few eosinophils seen in superficial and deep, interstitial, perivascular and periadnexal location; **B:** Small 1 to 2 micron round brown-coloured bodies(Copper penny bodies), singly and in aggregates, with peripheral dark rim andcentral pale area; **C:** Brown colour pigment seen extracellularly; **D:** PAS stain highlighting the pigmented fungal cells

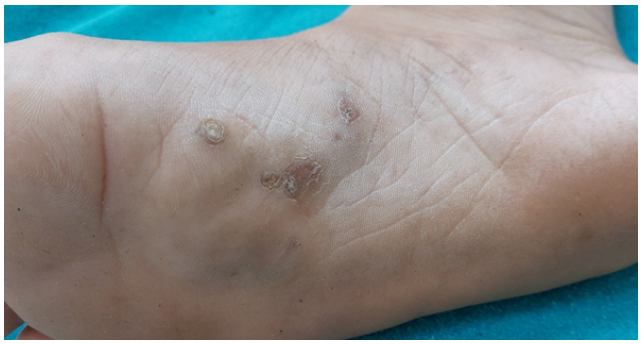

Figure 4: Follow up presentation after 4 weeks of treatment- Multiple indurated hyperpigmented plaques and nodules with scab and crusts, present over the instep of right foot.

Hyphal forms may be seen in the stratum corneum.[2] Direct KOH analysis of the lesions' crusts reveals numerous thick-walled, globose or ovoidal, dark-brown bodies that were singly or in groups.[7] Despite special fungal stains like Periodic acid schiff stain and Gomori methenamine stain being positive, haematoxylin and eosin stain is considered as gold standard for the diagnosis of CBM.[5]

Differential diagnosis of CBM includes fungal infections like blastomycosis, phaeohyphomycosis, paracoccidioidomycosis, coccidioidomycosis; bacterial infections like leprosy, cutaneous tuberculosis, nocardiosis; mycobacterial infections like Mycobacterium fortuitum, M. marinum; parasitic infestations like rhinosporidiosis and leishmaniasis; non-infectious disorders like psoriasis, squamous cell carcinoma, lupus erythematous and sarcoidosis.[4]

Various complications have been reported in literature including ulceration, secondary infections, ankylosis, lymphoedema and elephantiasis. Rarely, serious complications like squamous cell carcinoma may arise from long standing CBM. In immunocompromised patients, brain abscess has also been reported.[4]

Depending on the location and size of lesions, treatment may vary from medical management, surgery, chemotherapy, immunotherapy, and cryotherapy.[8] A single, primary lesion with clear margins may benefit the most from surgical removal. In mild to severe cases, systemic Itraconazole (200–400 mg daily) or pulse Itraconazole therapy (400 mg/day for 7 days/month for 3-4 months) or Terbinafine (500–1000 mg daily) for 6–12 months may be given. If the condition is severe, systemic therapy may be useful in shrinking the lesion so that it is amenable for surgical removal. Although Posaconazole and Voriconazole show promising results in implantation mycoses, they are still out of reach for many patients due to high costs.[2]

In our case, the patient developed complications like secondary bacterial infections repeatedly over several years. Bacterial tissue culture showed MRSA. The patient was treated with systemic and topical antibiotics for MRSA along with systemic and topical antifungals. Pain and discharge of grains from the lesions reduced in a few days following treatment. The size of the lesions also decreased gradually, which was evident on follow up at 4 weeks. The patient is still on treatment and is being followed up monthly.

4. Conclusion

Although CBM is known for many years, the diagnosis is difficult due to varied clinical presentations and therefore requires a high degree of clinical suspicion. Fungal tissue culture and histopathological examination are necessary for its diagnosis. Early identification and prompt treatment decreases the risk of further complications. Hence, this case is being reported to bring about an awareness among medical fraternity regarding this uncommon deep fungal infection.

5. Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's legal guardian has given the consent for images and other clinical information to be reported in the journal. The patient's legal guardian understands that name and initials will not be published, and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

6. Source of Funding

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7. Conflicts of Interest

There are no conflicts of interest.

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