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## EXTENSIVE LUPUS VULGARIS-LIKE CHROMOBLASTOMYCOSIS RESPONDING TO ITRACONAZOLE: CASE REPORT

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### SUMMARY

**We present a case report of a patient with extensive chromoblastomycosis that mimicks lupus vulgaris. This is a rare clinical presentation with atrophic patches on the trunk and crusted lesion on the lower extremities. The diagnosis was confirmed by histology, potassium hydroxide preparation and culture. Fonsecaea Pedrosoi was isolated on fungal culture. Patient was treated with itraconazole tablets 100mgs twice daily for three months with rapid improvement.**

**Sporadic cases of chromoblastomycosis occur in Eastern Africa. Patients present with disfiguring disease due to delay in treatment.**

### INTRODUCTION

Chromoblastomycosis is a fungal infection of the skin and subcutaneous tissue caused by dematiaceous fungi (1). A total of five fungi that inhabit soil, plants and wood have been identified as causing chromoblastomycosis. It is typically chronic in nature and presents as an expanding plaque on lower limbs and rarely on upper limbs. Chromoblastomycosis is found in the rural areas of tropical and subtropical climates (1).

### CASE REPORT

We present a 45-year-old married male patient from a nomadic pastoralist community. He presented to our

clinic with 20 years history of skin lesion which started on the left upper back. The initial lesions started as a small, non-itching plaque that progressively enlarged to cover the left upper back, left shoulder, left arm and left upper chest. After ten years similar skin lesions developed on left malar area and progressed in a similar pattern destroying the eyelids leading to inability to close the eye as shown on Figure 1A and C. Four years ago he developed itching verrucous skin lesions on the lower third of both legs which progressed distally and proximally to involve both lower limbs below the knee as shown on Figure 2A. None of the lesions has regressed or healed. No family member has similar skin lesions. He had been treated previously with gresiovulvin, topical steroids and topical anti-fungals without improvement.

**Figure 1A and C**

*Scaling depigmented atrophic lesion on the back and shoulder of a 46 year old male patient with destruction of the left lower eyelid, while figure 1B and D shows skin lesions after 3 months of treatment.*

**Figure 2A and B**

*Crusted and scaly verrucous plaques with areas discharging serous fluid and 2B is three months after treatment with fluconazole*



On examination, he was pale, wasted middle aged man with multiple skin lesions. The lesions on the trunk and the face were well demarcated erythematous and depigmented patches with central atrophy and scaling. Within the central atrophic areas are scattered tiny black spots. There was complete loss of the lower eyelid and inability to close the eye. The lower limbs below the knee were edematous and covered by verrucous nodules and plaques which were crusted and discharging serous fluid.

Baseline investigations done were essentially within normal. Rapid test for HIV infection was negative. Chest X-ray was normal and sputum for Acid Fast Bacilli was negative. Dark spots examined with 30% potassium hydroxide showed dark brown round fungal elements and septate hyphae as shown on Figure 3. Skin biopsy showed suppurative granulomas reaction with many giant cells. Round grouped, fungal elements with thick walls were seen free or inside the giant cells as shown on Figure 4. *Fonsecaea Pedrosoi* was isolated on fungal culture.

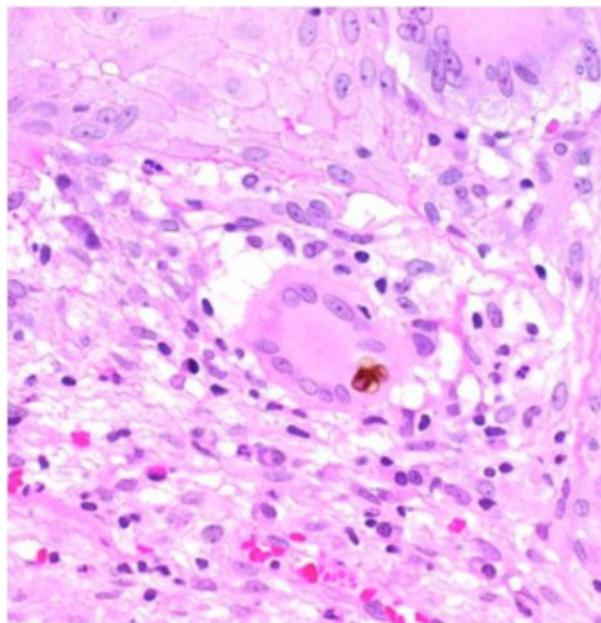
**Figure 3**

*Direct microscopy of 30% KOH preparation of the scales (x40) showing hyphae and sclerotic bodies.*



**Figure 5**

*Hematoxylin and Eosin histology showing suppurative granulomatous reaction with sclerotic bodies in cytoplasm of giant cells.*



The patient was treated oral itraconazole 100mg twice daily for three months with good response. Patient was monitored monthly and no adverse effects were reported.

At three months the skin lesions were healed with central depigmentation surrounded by areas of repigmentation and hypertrophic scars as shown on Figure 1B, 1C and 2B. Examination of skin scrapings with 30% potassium hydroxide was negative.

## DISCUSSION

Chromoblastomycosis is a rare chronic deep mycotic infection caused by dematiaceous (darkly pigmented) fungi, which include the genera *Fonsecaea*, *Phialophora*, and *Cladophialophora* (2). Infection is acquired via traumatic inoculation of the skin by contaminated thorns or wood splinters (2,3). Peasant farmers, miners and construction workers have a higher risk due to their occupational exposure. In general, men between the ages of 30 to 50 years have

a higher prevalence of the disease than the general population (4). This patient falls within the age group and comes from a nomadic pastoralist community. These people are frequently exposed to thorn pricks in the bush while herding animals. Though Tanzania is in the tropics with hot and humid climate the prevalence of chromoblastomycosis is relatively rare with few case reports from East Africa (5). Typically chromoblastomycosis is asymptomatic and progresses very slowly. Patients tend to seek medical care late when there is disfiguring or secondary bacterial infection and therefore treatment is delayed as demonstrated by this case.

In general chromoblastomycosis is a localised disease mostly affecting lower extremities which are prone to trauma. It seldom spreads through the lymph or blood vessels causing metastatic lesions away from the primary site (1). Despite extensive disease there was no nodal involvement in this patient. Clinically, chromoblastomycosis presents typically as a nodule or a plaque. Nodular type enlarges with verrucous surfaces while the plaque type expands with an active verrucous margins and central atrophic scaly skin. Scattered deposition of black dots on the lesion is an important diagnostic clue. Rarely, dissemination to brain can occur (2, 3). Apart from the verrucous presentation, there may be vegetative, fistulous, granulomatous, and squamous presentation (6). Chromoblastomycosis may be clinically confused with tubercular verrucosa cutis, sporotrichosis, lupus vulgaris, and even squamous cell carcinoma (2). This patient presented with chromoblastomycosis on unusual sites and morphologically the verrucous margins were absent on the lesions from the trunk, a presentation that is similar to lupus vulgaris. Chronic lymphedema and pruritus are common characteristics (1) which were also seen in this patient.

Diagnosis of chromoblastomycosis is by direct microscopy, histology and culture. Direct microscopy of 10% potassium hydroxide prepared scrapings taken from the lesion reveals round, brown, thick-walled, multiseptate sclerotic bodies that are pathognomonic of chromoblastomycosis. Specimens are more likely to yield a positive result if the scrapings are taken from black spots on the surface of the lesion. The black spots represent a transepidermal elimination of the fungal agents. Histopathologically, chromoblastomycosis generally presents as chronic granulomatous inflammation with pseudoepitheliomatous hyperplasia. The presences of large, spherical, thick-walled, brownish and septate sclerotic bodies are diagnostic. Mycological culture is used to characterize the morphological features of the fungal colonies and identify specific causative agents (7).

Successful treatment of chromoblastomycosis remains a clinical challenge (1). Several treatment options have been suggested however no randomized controlled trials have been done. Effectiveness of the treatment depends on the etiological agent, severity of the disease, host immunity and socio-economic status (8). Generally the cure rates are low and associated with high relapse rates (1). Small lesions in the early stages can be treated with surgical resection with curettage and desiccation (8, 9). Treatments with carbon dioxide laser, cryotherapy and topical heat also have been reported (8). Moderate and severe forms, with widespread lesions, usually require systemic treatment. While amphotericin B, thiabendazole, 5-Fluorouracil and ketoconazole are variably effective, itraconazole and Terbinafine have demonstrated the best results at high doses for 6-12 months (8, 10). This patient was treated with normal doses of itraconazole and responded well to treatment within three months. The optimum duration for treatment for a patient to be cured is unknown. This patient continued treatment for six months despite the negative potassium hydroxide results at three months. The main challenge in treatment is the supply of medications for longer duration in poor resource setting in Africa.

## CONCLUSION

We present a rare extensive morphological type of chromoblastomycosis which mimics lupus vulgaris clinically. Direct microscopy and histology is essential in making correct diagnosis. Poor health seeking behavior, unavailability and expensive medications contribute substantially to poor outcome in management of chromoblastomycosis.

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