

Lymphangitic chromoblastomycosis

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ABSTRACT

Chromoblastomycosis (CM), a chronic subcutaneous mycosis, is caused by several dematiaceous fungi, the most common being *Fonsecaea pedrosoi*. It usually occurs in the lower extremities following traumatic implantation of the organisms. We are reporting a case of chromoblastomycosis on the right lower limb in a sporotrichoid pattern caused by *F. pedrosoi*. The pattern was probably due to lymphatic spread that seems to be one of the rare presentations. The histopathology showed typical muriform or medlar bodies both intracellularly and extracellularly within the granuloma. Culture revealed sporulating organisms (*Cladosporium* and *Rhinocladiella* type) by a combination method, characteristic of *F. pedrosoi*. Our case responded well to itraconazole.

Key Words: *Fonsecaea pedrosoi*, Lymphangitic chromoblastomycosis

INTRODUCTION

Chromoblastomycosis (CM) is a chronic granulomatous mycotic infection of the skin and subcutaneous tissue caused by pigmented fungi, the most common being *F. pedrosoi*.^[1] It typically occurs on the exposed surfaces of the lower leg following traumatic implantation of the organisms.^[2-4] The lesions can involve other sites either by direct spread, autoinoculation^[5] or by hematogenous spread.^[6] The lymphatics may also play a role in disseminating the infection.^[1,7,8] We are reporting a case of CM caused by *F. pedrosoi* with an lymphatic spread.

CASE REPORT

A 40-year-old male agricultural worker of low socioeconomic status weighing 43 kg presented with a nonhealing ulcer on the right big toe and multiple

nodules over the anterior aspect of the right leg and foot [Figure 1], of one-year duration arranged in a linear fashion. He initially developed a nontender nodule on the dorsum of the right big toe which later got ulcerated. Subsequently, after three months, about five nodules started to appear in a linear fashion on the right foot and leg of size ranging from 1 to 2.5 cm in diameter with verrucous surface. He gave history of trauma at the site of initial lesion on the big toe. There was no regional lymphadenopathy.

His baseline blood and urine examination, renal and liver function tests were normal. Scrapings from the lesion and tissue smear did not demonstrate any organisms. Histopathology of the lesion showed pseudo-carcinomatous hyperplasia with neutrophilic micro abscesses in the epidermis. The subepidermal region showed granuloma composed of epithelioid cells and Langhan's giant cells. Muriform bodies were

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present intracellularly [Figure 2] as well as extracellularly. Fungal culture showed velvety dark gray to brown colonies with black pigment on the reverse [Figure 3]. Microscopic examination of the cultured organism showed sporulation by a combination method, with acrogenous conidia with short branching chains (*Cladosporium* type) and oval conidia at irregular positions on tips and sides of conidiospores (*Rhinocladiella* type), characteristic of *E. pedrosoi* [Figure 4]. This confirmed the diagnosis of chromoblastomycosis. We treated the patient with oral itraconazole 100 mg daily. Skin lesions had regressed by 80% after six months of therapy. However, the patient died due to ischemic heart disease.

DISCUSSION

CM is caused by several species of dematiaceous fungi and is usually confined to one of the lower extremities and affects only skin and subcutaneous tissue, though the draining lymph glands may participate in the pathological process.^[8] The CM lesion may be verrucous with central scarring (tuberculoid), severe scarring with a serpiginous border (syphiloid), scaly (psoriasiform) or indurated with fistula formation (mycetomatoid).^[9] Carrion described five morphologic types consisting of nodular, tumorous, verrucous, plaque and cicatricial lesions.^[9] The diagnosis of CM should be confirmed either by direct microscopy of the scrapings from the lesion in 20%



Figure 1: Nonhealing ulcer on the right big toe and multiple nodules over the anterior aspect of the right foot and leg



Figure 3: Fungal culture showing velvety dark gray to brown colonies with black reverse



Figure 2: Extracellular and intracellular muriform bodies (x400)



Figure 4: Microscopic examination (x100) of cultured organisms showing acrogenous conidia with short branching chains (*cladosporium* type) and oval conidia on tips and sides of conidiospores (*rhinocladiella* type)

KOH when thick-walled dark brown tissue forms of the fungus (fumagoid bodies/ muriform bodies/ copper penny bodies) are seen; by histological examination of a biopsy specimen with granulomatous reaction and spores; or by culture of scrapings or biopsy material.^[9]

Usually, histopathology reveals acanthosis and may demonstrate pseudo-carcinomatous hyperplasia. Neutrophils and giant cells may be seen infiltrating the epidermis with occasional formation of microabscesses. The dermis reveals a granulomatous tissue reaction with a mixed focal or diffuse inflammatory infiltrate consisting of lymphocytes, neutrophils, monocytes, plasma cells, eosinophils and giant cells of the foreign body and Langhan's type. The muriform bodies may be seen both intracellularly and extracellularly.

In our case both histopathological and mycological studies confirmed the diagnosis of CM caused by *F. pedrosoi*. However, the clinical presentation was atypical. The patient developed the lesion in a sporotrichoid pattern. Though CM can have a lymphatic mode of spread with satellite lesions, presentation in a sporotrichoid fashion is very rare. Carrion and Koppisch reported a case with well-documented involvement of the subcutaneous, lymphatic and muscular tissue.^[3] The involvement of lymphatic vessels (sporotrichosis-like) was observed by Fraga, Almada, Aleixo and Palominos.^[6] After this, no recent reports of sporotrichoid type of CM are available,^[3,4] except the description by Bopp et al.^[10] in which a vegetating plaque in the foot associated with inflammatory nodules on the leg was shown.

Our patient's response to itraconazole (ITZ) was good.

Hyle reported two cases treated successfully with 100 mg of daily ITZ therapy.^[11] Yu treated CM with 100 mg ITZ for 15 months with complete mycological and clinical recovery.^[12] Even though the current regimen of therapy is with 200-400 mg of ITZ alone or in combination with cryosurgery, considering the low body weight and socioeconomic status, we started the patient on 100 mg of ITZ with good response.

The present case is a good reminder that CM should be kept at the back of the mind in the differential diagnosis of sporotrichoid lesions.

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