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CONCISE COMMUNICATION

Case of chromoblastomycosis with pulmonary involvement

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ABSTRACT

Chromoblastomycosis is a slowly growing chronic cutaneous mycosis associated with a variety of cutaneous lesions. Extra-dermal involvement is rare. A 58-year-old man was admitted to the hospital with nausea, vomiting, weakness and a history of weight loss. On inspection, he had a large verrucous mass in the sacral region, and two large subcutaneous nodules in the anterior thoracic wall. He claimed the lesions were several years old. Biopsy and histological studies were positive for chromoblastomycosis. Routine chest radiography showed hilar enlargement, and a chest computed tomography was ordered. Pulmonary nodules were evident, and endoscopically acquired samples were also positive for chromoblastomycosis. Extra-dermal and systemic involvement in chromoblastomycosis is exceedingly rare and often associated with immunosuppression. There is only one other case of pulmonary chromoblastomycosis reported in the published work.

Key words: **chromoblastomycosis, cutaneous, infection, mycosis, pulmonary.**

INTRODUCTION

Chromoblastomycosis (CBM) is a slowly progressive cutaneous and subcutaneous mycosis. The pigmented fungi responsible for CBM are endemic of the tropics and countries such as India, Brazil and Mexico.[1,2] The most common etiological agents are filamentous fungi of the Dematiaceae family, of the genera *Fonsecaea*, *Phialophora* and *Cladophialophora*. *Fonsecaea pedrosoi* is the most commonly isolated fungi (70–90%) worldwide.[3] CBM presents first as papules in the skin (most commonly on lower extremities, due to inoculation by penetrating injury) that can progress to extensive dermal involvement and lymphatic spread.[1–3] Extra-dermal spread is rare.

CASE REPORT

A 58-year-old male farmer was admitted to the hospital with nausea, weakness and a history of weight loss. He revealed no relevant medical history. On his initial physical examination, a large, verrucous, tumor-like lesion was evident on his sacral region (Fig. 1a). The patient claimed it appeared over 15 years prior, but he did not seek prior medical attention. Two large, subcutaneous, mobile, non-painful masses had appeared on his anterior chest wall 3 months before his admission. He had no history of dyspnea, cough or fever. His body mass index was 18.2.

He was anemic, with a 7.15 g/dL hemoglobin level and no leukocytosis. Serum electrolytes and renal function were normal. Only hypoalbuminemia was found on his liver function tests (2.5 g/dL). Enzyme-linked immunosorbent assay for HIV was non-reactive. On routine chest radiography, a left hilar enlargement was evident. A chest computed tomography was ordered and showed a left parahilar mass that obstructed the bronchi (Fig. 1b). Tissue samples were obtained from cutaneous lesions and endoscopically from the hilar mass (Fig. 1c). All were positive for thick-walled, multiseptate, brown sclerotic cells after 10% potassium hydroxide application (Medlar bodies, Fig. 1d). There was no histopathological evidence of malignancy, or other fungal structures. A slowly growing, black colony covered with velvet-like silver mycelium grew in cultures. In slide-culture, brown, branching hyphae with dark elliptical conidia were evident (Fig. 2). Cultures were interpreted as positive for *F. pedrosoi*. A diagnosis was made of systemic CBM with pulmonary involvement.

There was no evidence of overt bleeding or occult blood loss in stool, and anemia was attributed to chronic disease and iron-deficiency due to malnutrition. The patient was started on itraconazole, 400 mg/day. On 6 months follow-up, the patient has gained approximately 20 pounds, his skin lesions have improved and his symptoms have disappeared. He is no longer anemic and his liver function tests remain normal.

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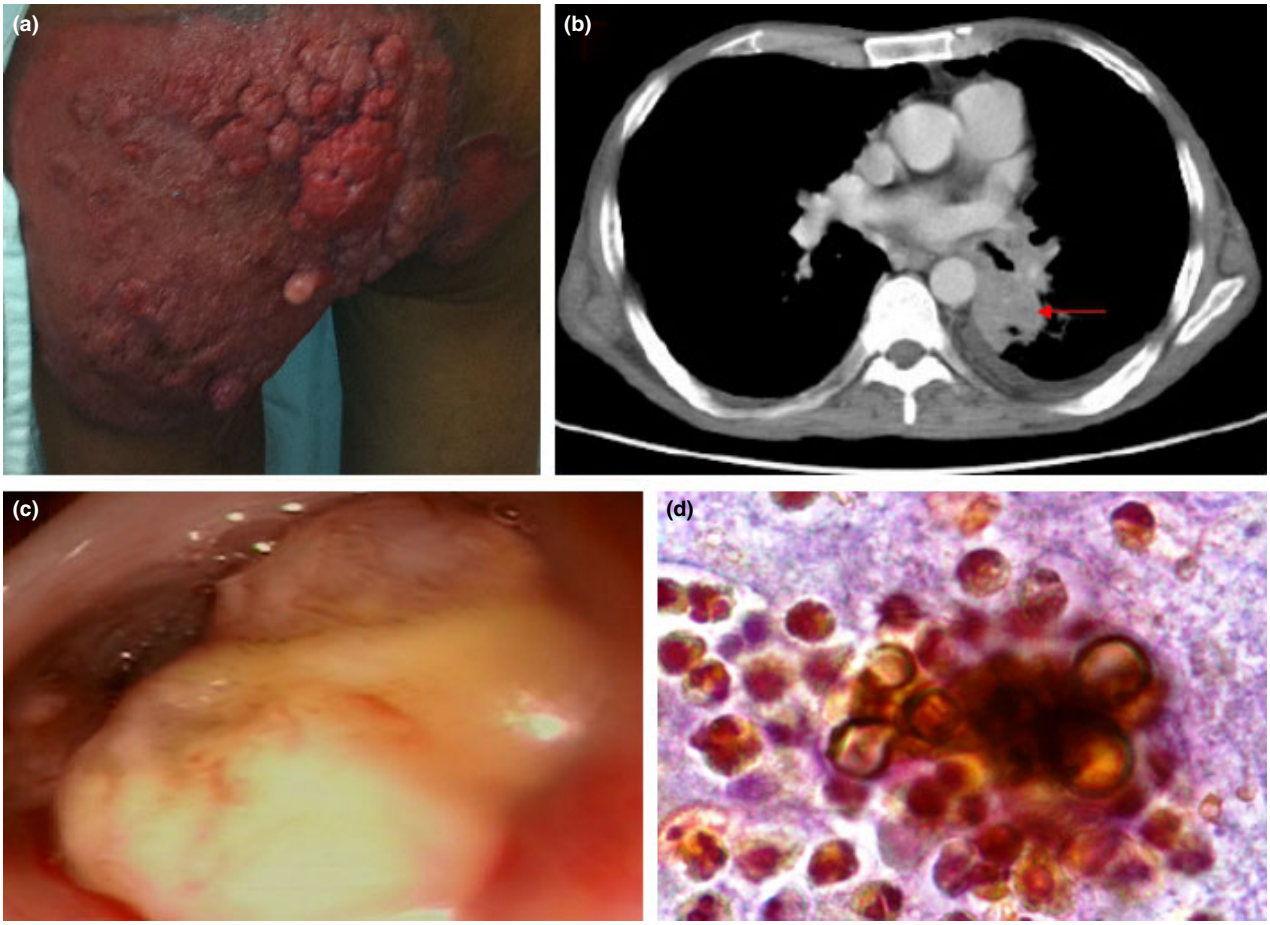

Figure 1. (a) Skin lesion. (b) Computed tomography image showing a left hilar mass. (c) Mass as visualized on endoscopy (arrow). (d) Medlar cells in histological analysis (hematoxylin–eosin, original magnification ×400). The image is a combination of four separate pictures. They were not digitally altered except for the arrow and letters.

Figure 2. Slide-culture showing brown, branching hyphae with dark elliptical conidia (lactophenol cotton blue stain, original magnification ×400).

DISCUSSION

Chromoblastomycosis is a slowly progressive cutaneous and subcutaneous mycosis. *F. pedrosoi* is the most commonly isolated fungi worldwide, and found in dirt and vegetation near tropical forests.[1,3] CBM presents first as papules in the skin, and cutaneous lesions can also be verrucous, tumoral, cauliflower-like and even ulcerative. However, extra-dermal manifestations are rare.

The diagnosis of CBM can be made using direct microscopy, culture and molecular methods. Traditionally, the finding of "Medlar cells" or "muriform bodies" is pathognomonic.[4,5] These are thick-walled, multiseptate, brown sclerotic cells found in scrapings of skin lesions after 10% potassium hydroxide application. Other differential diagnoses that must be considered are sporotrichosis, squamous cell carcinoma, protothecosis and verrucous tuberculosis.[4,6] The latter diagnosis was thoroughly ruled out, considering the lung lesions present in our patient.

Extra-dermal, or systemic, CBM is extremely rare. When present, there must be a suspicion of immunosuppression.

The only possible risk factor in our patient was a clinical state of malnutrition. CBM involvement has been reported in cerebrum,[7] cornea,[8] liver[9] and lymph nodes.[10] Here, we report a case of CBM with pulmonary involvement. The lung lesions were positive for Medlar bodies on analysis. In a case-based review of 34 patients with cutaneous CBM, 24% were found to have extracutaneous involvement, including tonsils, laryngotracheal area and pleural cavity.[11] However, lung parenchyma was not affected, and no histopathological confirmation of lung involvement was reported. The Medlar body represents an adaptive tissue form of the fungi known to cause cutaneous CBM, but systemic involvement has been usually demonstrated after visualization of fungal hyphae in extracutaneous tissue. Moreover, Medlar bodies have been found in brain tissue, and it has been demonstrated that they can germinate and form hyphae even in cutaneous lesions.[12] Medlar bodies have been also found in lymph nodes and in the liver.[9,10] We did not observe hyphae in the tissue obtained from the bronchial mass, but affection could be segmental, or this could be due to an extremely indolent (15 years) course of the disease.[12] We found only one previous report of pulmonary involvement of CBM, published in 1982.[13] CBM has been reported to appear in patients with chronic obstructive pulmonary disease, but the presentation was cutaneous, and chronic steroid use was identified as a risk factor.[14,15]

Treatment depends upon presentation. In limited cutaneous disease, cure is feasible by excision and antifungal therapy. However, extensive or systemic disease can only be controlled, and long-term antifungal therapy is required.[3,6] Relapse is common. Itraconazole and terbinafine are first-line agents, and they require high doses for up to 6–12 months.[3,6] Our patient received itraconazole in 400-mg/day doses, but the extension of the lesions prevented surgical therapy. He is recovering well.

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