

# Report of an imported cutaneous disseminated case of paracoccidioidomycosis

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## Summary

Paracoccidioidomycosis is a chronic progressive infection. It affects mainly the elderly and it is geographically limited to certain areas of Latin America. In Europe it is considered a rare imported infection. Here we report a case of paracoccidioidomycosis that occurred in a 27-year-old Ecuadorian patient living in Spain initially misdiagnosed as blastomycosis. The typical multi-budding yeast cells of *Paracoccidioides brasiliensis* were observed in Grocott stained samples. This case should alert Spanish mycologists, clinicians and pathologists about the possibility of patients who have travelled or lived outside Spain may suffer paracoccidioidomycosis or other imported mycoses.

## Key words

Mycoses, Paracoccidioidomycosis, Imported, Grocott stain

## Paracoccidiomycosis cutánea diseminada de importación

## Resumen

La paracoccidioidomycosis es una infección crónica progresiva. Afecta preferentemente a hombres mayores de 35 años, trabajadores rurales y está limitada geográficamente a ciertas zonas de Latinoamérica. En Europa la enfermedad es muy rara. Presentamos el caso de una paracoccidioidomycosis que afecta a un varón joven de 27 años, nacido en Ecuador, pero residente en España desde hace varios años. Su enfermedad fue diagnosticada inicialmente como blastomycosis. Las típicas células fúngicas de *Paracoccidioides brasiliensis* fueron posteriormente observadas mediante la tinción de Grocott. El presente caso quiere alertar a micólogos, clínicos y patólogos sobre la posibilidad de que también pacientes jóvenes nacidos o que hayan viajado a zonas endémicas, puedan padecer paracoccidioidomycosis u otras micosis importadas.

## Palabras clave

Micosis, Paracoccidioidomycosis, Importada, Tinción de Grocott

Paracoccidioidomycosis is a systemic mycosis geographically restricted to Latin America, caused by the dimorphic fungus *Paracoccidioides brasiliensis* [1,8]. The most common manifestation is pulmonary infection, but the pathogen can spread to other anatomical sites, including the oral mucous membrane, the lymph nodes and the

skin. Paracoccidioidomycosis is infrequent in children and young adults [9]. In Europe it is rare, and all case reported had a common clinical history the patients lived, worked or had visited endemic countries of Latin America [5,7,10]. We describe an imported case of disseminated infection caused by *Paracoccidioides brasiliensis* in a young patient from Ecuador.

## Case report

A 27-year-old Ecuadorian male farmer patient from Ibarra, arrived at Zaragoza, in Spain, in February 2003. His clinical history revealed that the patient had presented cutaneous lesions for 1 year with showed subcutaneous nodules in the skin of the right elbow, the left side back, the perinasal area and lymph nodes. The infection was previously diagnosed as blastomycosis and treated with ketoconazole (200 mg/day). In March 2003, a biopsy was taken from the skin on the left side of the patient's back. Although, this new biopsy revealed a chronic, necrotic lesion with epithelioid granulomas and round fungal cells,

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the patient was diagnosed again with blastomycosis and treated with ketoconazole (200 mg/day), but the lesions remained. The period of treatment with ketoconazole was 15 months. Two months later, other biopsy was taken from right subcutaneous elbow. The haematoxylin-eosin (H-E), and periodic acid-Schiff (PAS) revealed an inflammatory chronic granulomatous infiltrate consisting of giant cells, histiocytes, some lymphocytes and fungal cells, most of which were inside of the giant cells (Figure 1). We also observed multiple yeast-like cells with sparse multipolar budding when using Grocott stain (Figure 2). Culture was not performed. Based on the multi-budding yeast cells the patient was diagnosed with paracoccidioidomycosis and treated with itraconazole, 200 mg/day for 5 months. After treatment, the lesions healed and completely disappeared. During treatment the patient presented jaundice with no painful hepatomegaly suggesting of toxic hepatitis for the itraconazole. A computerized tomography scan showed dilation of the intrahepatic via and obstructive lithiasis. A cholecistectomy was performed and lithiasis was confirmed. Infection or other pathologies were not observed. X-ray of the thorax did not reveal pathology as was the case on previous diagnosis. Blood studies did not reveal increase of erythrocyte sedimentation rate or other parameters.

Few cases of imported paracoccidioidomycosis have been reported in Europe. For instance, in the last ten years, only 8 cases have been reported in this particular geographic area. Cases were reported in Germany [5], Italy [2], The Netherlands [10], Austria [7] and Spain [3,4]. All of these cases had several patterns in common. For example, all the patients had lived in South America (Brazil, Venezuela, or Ecuador) except for the patient in Austria, who had lived in Cuba, and all were over 59 years old. Also, all patients had a long silent period before the symptoms appeared. The disease is progressive when clinical manifestations become apparent, such as in acute juvenile form or chronic adult form. Paracoccidioidomycosis is not frequent in children or young adults, this population represents only 5% to 10% of all cases [9]. We reported here a case of disseminated paracoccidioidomycosis with the involvement of lymph nodes and skin in an Ecuadorian patient of only 27 years old. Diagnosis of paracoccidioidomycosis can be difficult because it could be misdiagnosed as tuberculosis, leprosy or other fungal infections such as blastomycosis, as indeed had happened in our case. Clinical suspicion is necessary because lesions may appear many years after patients have acquired the infection. Paracoccidioidomycosis should be suspected in patients who have travelled to endemic area and who presented weight lost and has pulmonary, mucosal, and cutaneous lesions. Diagnosis can be made by histopathology. Slides stained with PAS, H-E and mainly with Grocott are useful for visualizing typical fungal elements of *P. brasiliensis*. In our case, however, we observed typical fungal structures such as yeast-like multiple budding mimicking a pilot's wheel, only when using a Grocott stain. Several classes of therapies, such as sulphonamides, amphotericin B and azoles, have been used to treat paracoccidioidomycosis. However, with these drugs relapses has been observed

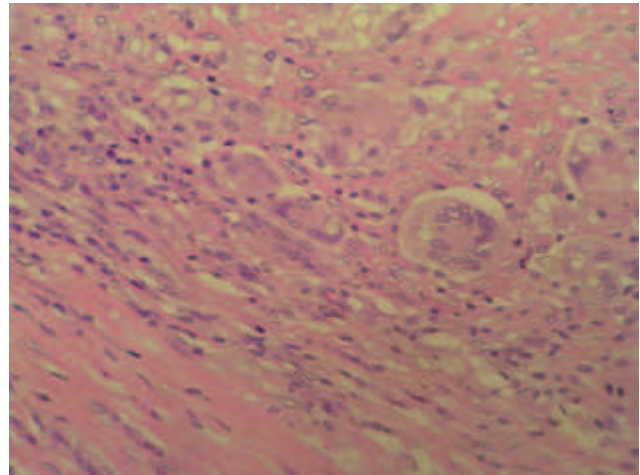


Figure 1. Histopathological section showing granulomatous reaction with numerous giant cells and some intracellular yeast. H-E x400.

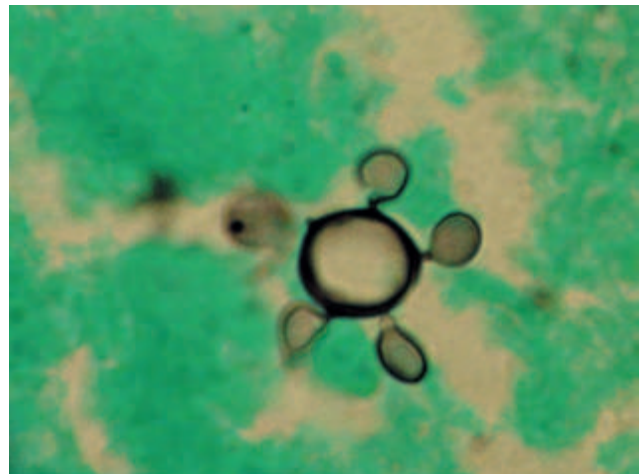


Figure 2. *Paracoccidioides brasiliensis* cells with multiple narrow-necked buds. Grocott stain x1000.

in 8%-25% of patients even with appropriate treatment [9]. Itraconazole is considered the antifungal of choice for most patients with a relapse rate of less than 5% [6]. In our case, the patient showed relapse with the initial therapy (ketoconazole), but responded successfully when treated with itraconazole.

Due to the increasing number of travellers coming into Spain from the endemic areas of paracoccidioidomycosis and the other restricted mycoses, pathologists and clinicians should be alert about the diagnosis and management of these imported infections.

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