

# Osteomyelitis of the wrist in a patient with disseminated paracoccidioidomycosis: a rare presentation\*

*Osteomielite de punho em paciente com paracoccidioidomicose disseminada: uma rara apresentação*

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**Abstract** Paracoccidioidomycosis is the most frequently found endemic systemic mycosis in Brazil. No symptoms are observed in the early phases of the disease. As the disease progresses, the patient may present disseminated involvement, but bone involvement is extremely rare. The present report is aimed at evaluating bone changes found on imaging studies in a patient with osteomyelitis of the wrist as a result of disseminated paracoccidioidomycosis.

**Keywords:** Paracoccidioidomycosis; Endemic mycosis; Osteomyelitis.

**Resumo** Paracoccidioidomicose é a micose sistêmica endêmica mais frequente no Brasil. No início, o paciente não desenvolve sintomas. Com a progressão da doença, o indivíduo pode apresentar envolvimento disseminado, sendo que o acometimento ósseo é extremamente raro. O objetivo deste artigo é avaliar as alterações ósseas encontradas em estudos de imagem em um paciente com osteomielite de punho decorrente de paracoccidioidomicose disseminada.

**Unitermos:** Paracoccidioidomicose; Micose endêmica; Osteomielite.

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## INTRODUCTION

Paracoccidioidomycosis is the most common endemic systemic mycosis in Latin America<sup>(1,2)</sup>. Such condition is acquired by inhalation of infected particles which reach the lungs, developing primary infection. In the early stages of the disease, the patient does not develop any symptom or otherwise present subtle and nonspecific symptoms. As the disease progresses, the patient presents severe involvement of several organs such as skin, mucosae, lungs and bones. Such an infection may progress with dissemination, but the disseminated presentation occurs in only 3–5% of cases<sup>(3)</sup>.

The present report is aimed at evaluating bone changes observed on imaging studies of a patient with osteomyelitis of the wrist secondary to disseminated paracoccidioidomycosis, as a contribution to the early diagnosis and treatment of this disabling disease.

## CASE REPORT

A male, 59-year old patient who as agricultural worker and with a long history of smoking attended an emergency unit in the city of Vitória, ES, Brazil, reporting progressive dyspnea for three months, ulcerative lesions on his skin and mucosae in association with pain and soft tissue enlargement in his right wrist, with serosanguineous secretion. Findings at physical examination included cervical and axillary lymph nodes enlargement, ulcerative lesions on the skin of his dorsal region and oral mucosa, besides a significant increase in volume of his right wrist.

Chest radiography demonstrated diffuse and confluent opacities in the middle thirds of the pulmonary fields and opacities corresponding to fibrotic scars in the lung bases. Radiography of the right wrist demonstrated a subtle decrease in the bone density of the distal ulna (Figure 1A).

Considering the radiographic findings, the patient was treated with antibiotics and referred to the center of infecto-parasitic diseases at Hospital Universitário Cassiano Antônio de Moraes. One month after the treatment, new radiographic images of the wrist and high resolution computed tomography (HRCT) of the chest were acquired in addition to fungal serology and cytology of the wrist secretion. The cytology revealed the presence of a high number of fungi compatible with *Paracoccidioides brasiliensis*.

Radiography of the right wrist demonstrated a lytic area in the distal ulna (Figures 1B and 1C). The patient was submitted to magnetic resonance imaging of the wrist, which demonstrated a significant involvement of the bone marrow in the distal ulna, development of bone abscess and fistulous trajectory, with involvement of the muscle and adjacent subcutaneous tissues, with intense contrast-enhancement (Figure 2). At chest HRCT multiple consolidations were observed in the middle and lower thirds of the lungs, besides septal thickening, cavitating nodules and bronchial walls thickening (Figure 3). Technetium and gallium scintigraphy was performed, confirming the presence of active disease in the lungs and right wrist (Figure 4). Contrast

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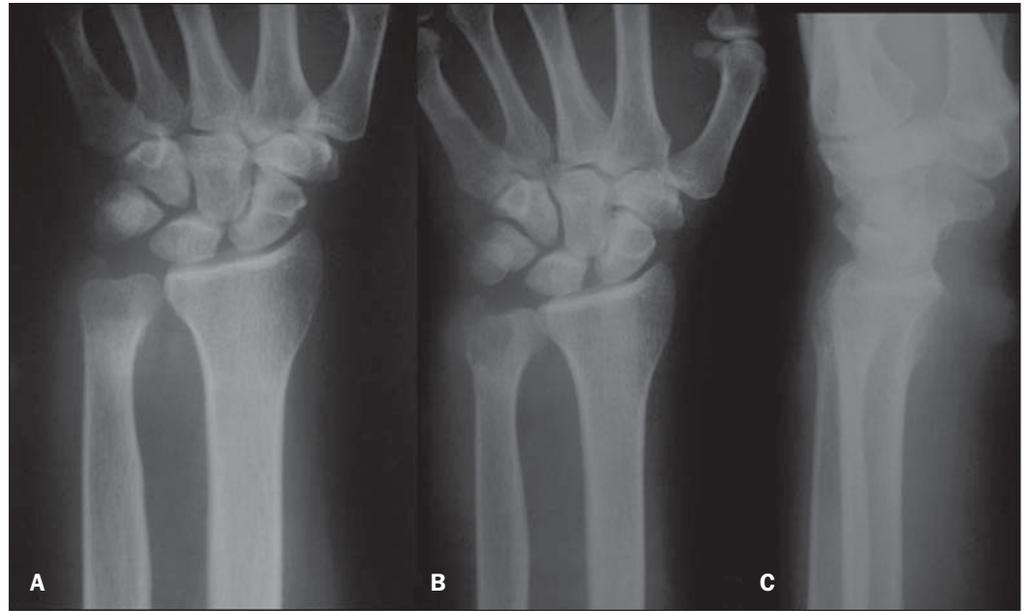
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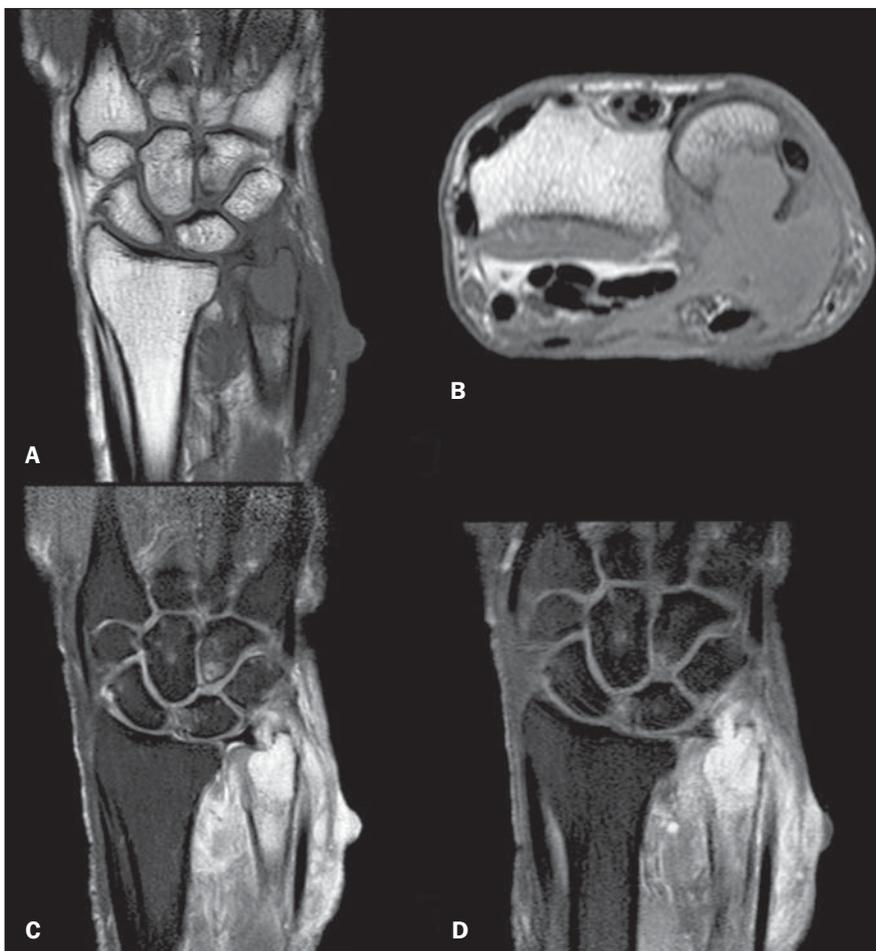
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**Figure 1.** Plain radiograph of wrist. **A:** The first radiograph of the right wrist demonstrates subtle decrease in bone density of the distal ulna and preserved joint spaces. **B,C:** Antero-posterior and lateral radiographic images of wrist acquired one month later, clearly identify a lytic area in the distal ulna.



**Figure 2.** MRI of right wrist. **A,B:** Coronal and axial T1-weighted images demonstrate intermediate signal intensity in the distal ulna bone marrow, with cortical destruction and fistulous tract. **C:** Coronal, T2-weighted image reveals a significant bone marrow edema and extension of the infectious process towards soft tissues. **D:** Contrast-enhanced coronal T1-weighted image demonstrates enhancement of the bone marrow as well as of the muscle and adjacent subcutaneous tissues.

uptake was not observed in other bones or in the central nervous system.

The patient was admitted and initially treated with Amphotericin B. After improvement of the respiratory symptoms and decrease in volume of soft tissue components in his right wrist, the patient was discharged with sulfamethoxazole-trimethoprim, remaining under regular follow-up on an outpatient basis.

## DISCUSSION

Paracoccidioidomycosis affects primarily adult individuals in their most productive phase, causing high social and economic impact. More than 90% of cases occur in male individuals<sup>(1,4)</sup>, representing a relevant public health problem because of its high disabling potential and high mortality in cases of disseminated disease<sup>(4,5)</sup>. Accurate epidemiological data on the disease are not available in Brazil, since Brazilian regulations do not require mandatory notification<sup>(6)</sup>.

Paracoccidioidomycosis may manifest in several organs, particularly in lungs, skin, mucosas and lymph nodes. Lungs are most commonly affected<sup>(7)</sup>, with radiographic changes in 60% of acute cases and in up to 80% of chronic cases<sup>(8)</sup>. Small opacities constitute the most common pulmonary findings, generally with bilateral and symmetric distribution of the le-



**Figure 3.** HRCT of chest demonstrates bilateral confluent opacities in pulmonary fields, cavitating nodules and bronchial walls thickening.

single bone involvement are reported in the literature.

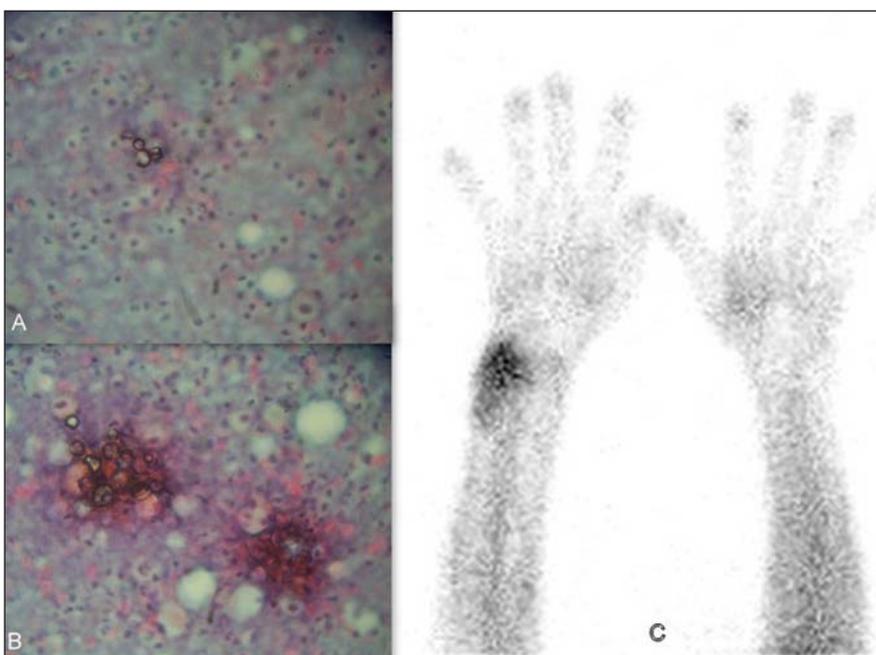
In many cases, osteomyelitis secondary to paracoccidioidomycosis is a late diagnosis, resulting in high morbidity and mortality.

Imaging studies are useful to evaluate the disease extent. Suspicious areas must be radiologically investigated. Bone scintigraphy can assess the whole body. Gallium scintigraphy can detect inflammatory activity.

Therefore, the identification of imaging findings in cases of osteomyelitis secondary to paracoccidioidomycosis is extremely relevant for early diagnosis and treatment, reducing morbidity.

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**Figure 4.** Scintigraphy and cytology of right wrist. **A,B:** Cytology of the right wrist secretion demonstrates the presence of a high number of fungi compatible with *Paracoccidioides brasiliensis*. **C:** Significant radiopharmaceutical uptake in the distal wrist, indicating the presence of active disease.

sions<sup>(4)</sup>. Bones involvement is extremely rare<sup>(3,9-11)</sup>.

The typical finding is a well-defined lytic area, either with or without a sclerotic halo that may be observed in any bone, either with or without involvement of soft tissues. The disease is generally multifocal. Differential diagnoses include other infectious diseases such as chronic bacterial os-

teomyelitis, tuberculosis and primary or metastatic tumors such as lymphoma and osteosarcoma.

The disease location is determined by hematogenous dissemination. It was first described by Pereira and Vianna in 1911, in a case with multiple bone lesions affecting the sternum, ribs, skull, tibia, sternoclavicular joint and shoulder. Few cases of