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Case report

Musculoskeletal ultrasound findings in paracoccidioidomycosis



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ABSTRACT

Paracoccidioidomycosis is a fungal infection endemic to South America. The infection is usually asymptomatic and mostly affects the upper and lower respiratory tracts with clinical-radiological dissociation. Joint involvement is rare with no specific pattern or radiological injury. We report a case of paracoccidioidomycosis in which the patient's initial symptoms were hoarseness and arthritis. After an ultrasound examination, we performed the differential diagnosis of other noninfectious arthropathies and analysis of the material collected, which revealed infection with the fungus *Paracoccidioides brasiliensis*.

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Achados ultrassonográficos osteomusculares na paracoccidioidomicose

RESUMO

A paracoccidioidomicose é uma infecção fúngica endêmica na América do Sul. A infecção geralmente é assintomática e afeta principalmente os tratos respiratórios superior e inferior, com dissociação clínico-radiológica. O envolvimento articular é raro, sem um padrão ou lesão radiológica específica. Apresenta-se um relato de caso de paracoccidioidomicose em que os sintomas iniciais do paciente foram rouquidão e artrite. Depois de um exame de ultrassonografia, foram feitos o diagnóstico diferencial de outras artropatias não infecciosas e a análise do material coletado, que revelou infecção pelo fungo *Paracoccidioides brasiliensis*.

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Palavras-chave:

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Introduction

Paracoccidioidomycosis is a fungal infection endemic to South America caused by the thermo dimorphic fungus *Paracoccidioides* sp.¹ The fungus has a spatially heterogeneous distribution, with areas of low and high endemicity, and the disease is acquired by airborne inhalation or via lesions in skin and mucosa. The infection is usually asymptomatic and mostly affects the upper and lower respiratory tracts with clinic-radiological dissociation. Even though rare, several extra pulmonary manifestations have been reported in the genitalia, gastrointestinal tract, intraspinal region, and central nervous system, and the association between paracoccidioidomycosis and cancer and HIV has been suggested. Joint involvement is rare with no specific pattern or radiological injury.²

We report a case of paracoccidioidomycosis in which the patient's initial symptoms were hoarseness and oligoarthritis. An ultrasound (US) was performed as an extension of the physical examination for possible differential diagnosis of other diseases that may produce similar joint changes. Articular manifestations of paracoccidioidomycosis include carpal tunnel syndrome, association with gouty arthritis in the proximal phalanx of the hallux, and pyoarthritides even in the absence of immunosuppression risk factors.^{3,4}

Osteoarticular involvement in paracoccidioidomycosis is variable, with 2.2-4% of bones and joints, and in the acute/subacute form of the disease it accounted for 20-26.4% of paracoccidioidomycosis cases.^{5,6} Skin involvement presents histologically as papillomatosis, with epidermal proliferation and formation of microabscesses.⁷

Interestingly, in this clinical case we performed guided ultrasound to collect articular material, which resulted in a more accurate diagnosis and detection of previously unreported ecotextural changes and cutaneous manifestations such as microabscesses.

Case report

The patient was a 55-year-old man, resident in the rural area, state of São Paulo, Brazil with a six-month history of joint pain.

He presented with asymmetric inflammatory pain in the left wrist and in the fingers, mostly in the 1st right distal interphalangeal (DIP) and 3rd proximal interphalangeal (PIP) joints, associated with hoarseness, which started at the same time as the other symptoms. In addition, he reported losing 3 kg since the onset of symptoms. The patient denied fever or contact with people with contagious diseases.

On physical examination, he had hoarseness, increased volume and temperature on palpation, in addition to hyperemia in the palm and thenar eminence of the left wrist and 1st right distal interphalangeal joint.

As a regular procedure in our practice, we performed a musculoskeletal ultrasound (MSUS) as an extension of the physical examination, which showed hypoechoic, heterogeneous collection in subcutaneous planes, extending to the flexor tendons of the left wrist and first right DIP, characterizing tenosynovitis and synovitis in dorsal recess of the 1st right DIP and 3rd right metacarpophalangeal (MCP) joints, with important inflammatory activity, shown by the presence of intense power Doppler signal (PD) (Figs. 1 and 2).

We performed an US-guided puncture of the collection and sent the material to the laboratory for bacilli tests, fungal and bacterial cultures, and direct mycological examination. The results were returned quickly, and direct mycological examination was positive for *Paracoccidioides brasiliensis*. Serologic tests for *Paracoccidioides* spp. were not realized and blood and wrist secretion cultures were negative for *Mycobacterium* spp. and fungi.

On further investigation, laryngoscopy revealed glottic cleft, chest radiograph thick reticular pattern affecting both lungs, chest computed tomography (CT) scan with cavitations (Fig. 3). The patient refused HIV testing.

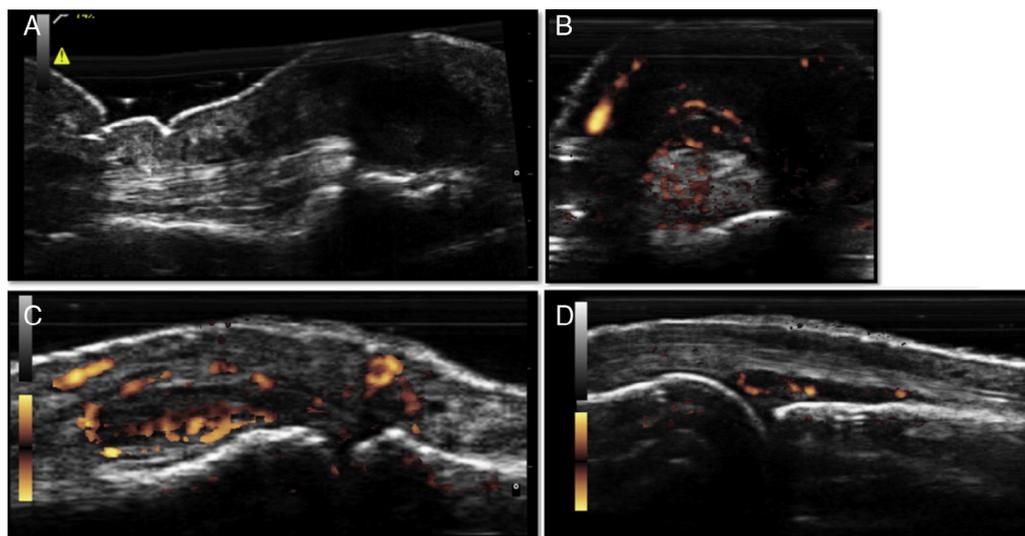


Fig. 1 – (A). Longitudinal US image of the palmar recess of the 1st right interphalangeal (DIP) joint, showing subcutaneous hypoechoic collection involving the flexor digitorum profundus tendon and (B) transverse scan with positive PD signal. US image showing dorsal recess synovitis with positive PD signal of (C) 1st right interphalangeal (DIP) and (D) 3rd right metacarpophalangeal (MCP) joints.

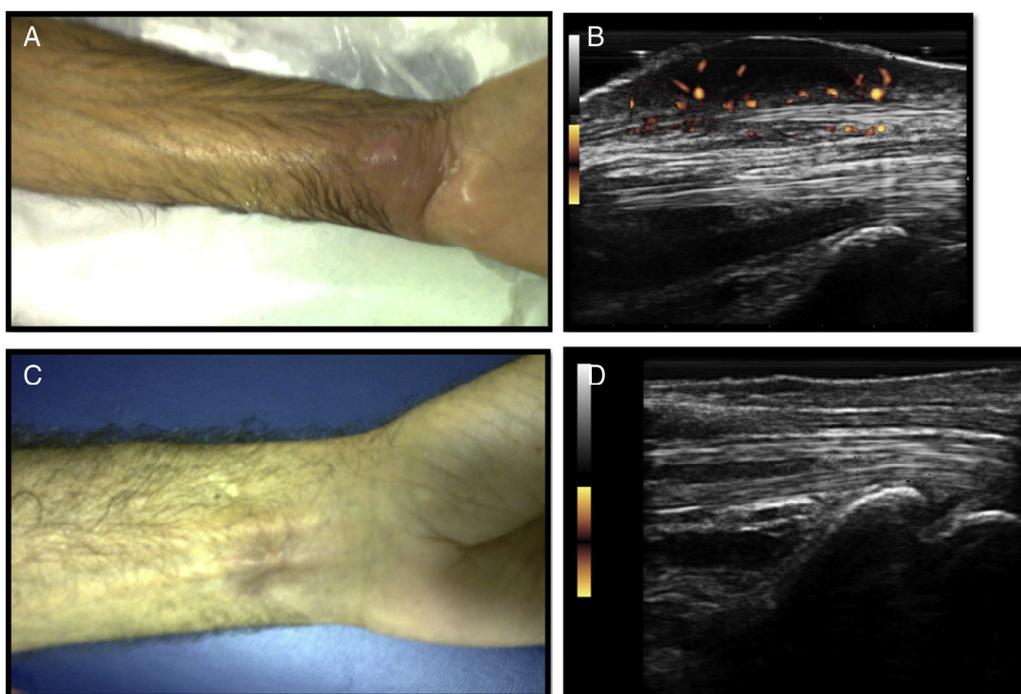


Fig. 2 – (A) Palmar aspect of the left wrist showing a floating lump. (B) US image of the palmar recess, with subcutaneous hypoechoic collection invading flexor tendons and presence of PD signal. (C), Palmar aspect of the left wrist after treatment. (D) US image of the palmar recess of the left wrist, showing no changes in flexor tendons after treatment.

The patient's treatment commenced with sulfamethoxazole 400mg/trimethoprim 80mg, two tablets three times a day.

Three months after treatment, US images showed the improvement of flexor tendon tenosynovitis in the left wrist and subcutaneous collection, without PD signal (Fig. 2).

Koch bacillus and bacterial culture results were negative. The patient currently undergoes follow-up in our institution.

Discussion

Paracoccidioidomycosis most commonly manifests as a chronic systemic mycosis in adults, affecting the lungs and

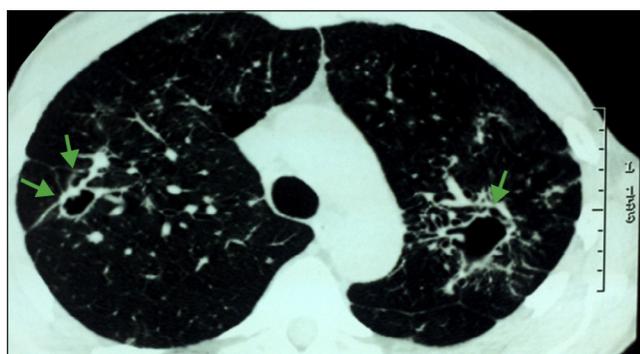


Fig. 3 – Chest high resolution CT scan, slice of 512 x 512 pixels, in a 55-year-old man, Caucasian, resident in the rural area and joint pain showing cavitations in both lungs, indicated by the arrows.

upper respiratory tract.⁸ Additionally, osteoarticular lesions occur in all age groups, but are more frequent in young subjects and, as in other clinical presentations of the disease, males are more frequently affected. There are no known risk factors for this kind of involvement, but localized traumas are believed to facilitate the establishment of the fungus in the osteoarticular system during fungemia.²

Many patients are sick carriers because of an imbalance between the production of Th1 and Th2 lymphocytes, with a predominance of the latter, and cytokines such as IL-4, IL-5, IL-10, and IL-18 that promote granuloma disruption.⁹⁻¹²

The reversed halo sign with pneumonitis and reorganization in the chest CT scan is highly suggestive of lung infection by paracoccidioidomycosis, even though other fungal and granulomatous infections may have similar symptoms.¹³ Direct mycological examination is the simplest, least expensive laboratory diagnosis, but negative results should be followed by fungal culture and more specific tests such as PCR and ELISA.¹⁴

The treatment is prescribed according to the economic conditions of patients and availability of medications in the primary care network. However, the duration and the choice of the drug to treatment are still unclear. Both itraconazol 200mg/day for 6-9 months in mild forms and 12-18 months in moderate forms, or sulfamethoxazole 800-1.200mg+trimethoprim 160-240mg every 12h for 12 months in mild forms and 18-24 months in moderate forms are indicated.¹⁵

On clinical examination, there were a few symptoms of note: hoarseness, a small abscess in the palm of left wrist, the fact that the patient lived in a rural area, and the absence

of lower respiratory tract symptoms, which could be detected only in complementary exams. Analysis of the fluid drained from the wrist abscess helped complete the diagnosis. The joint changes detected by high-resolution ultrasound showed great synovial involvement and subcutaneous involvement. However, no bone damage, the most frequent osteoarticular manifestation, nor ulcerations in skin or mucosa caused by fungal dissemination were detected, probably due to the early diagnosis.

In conclusion, ultrasound exams should be part of routine medical care as an extension of rheumatologic physical examination to help formulate more accurate differential diagnoses in patients with mono- or oligoarthritis, mainly in infectious arthropathies such as tuberculosis, leprosy, septic arthritis, spondyloarthropathies, and also paracoccidioidomycosis.

Conflicts of interest

The authors declare no conflicts of interest.

REFERENCES

1. Queiroz-Telles F, Escuissato DL. Pulmonary paracoccidioidomycosis. *Semin Respir Crit Care Med*. 2011;32:764-74.
2. Mendes RP. Bone and joint lesions. *Paracoccidioidomycosis*. 2nd ed. Boca Raton: CRC Press; 1994. p. 331-8.
3. Bonilla-Abadía F, Vélez JD, Zárate-Correa LC, Carrascal E, Guarín N, Castañeda-Ramírez CR, et al. Over infection by *Paracoccidioides brasiliensis* in gouty crystal arthritis. *Case Rep Med*. 2012;1-3.
4. Saccente M, Woods GL. Clinical and laboratory update on blastomycosis. *Clin Microbiol Rev*. 2010;23:367-81.
5. Trad HS, Trad CS, Elias J Jr, Muglia VF. Radiological review of 173 consecutive cases of paracoccidioidomycosis. *Radiol Bras*. 2006;39:175-9.
6. Bellissimo-Rodrigues F, Bollela VR, Da Fonseca BA, Martinez R. Endemic paracoccidioidomycosis: relationship between clinical presentation and patients' demographic features. *Med Mycol*. 2013;51:313-8.
7. Costa MAB, Carvalho TN, Araújo Júnior CR, Borba AOC, Veloso GA, Teixeira KS. Extra-pulmonary manifestations of paracoccidioidomycosis. *Radiol Bras*. 2005;38:45-52.
8. Sant'Anna GD, Mauri M, Arrarte JL, Camargo H Jr. Laryngeal manifestations of paracoccidioidomycosis (South American blastomycosis). *Arch Otolaryngol Head Neck Surg*. 1999;125:1375-8.
9. Fortes MR, Miot HA, Kurokawa CS, Marques ME, Marques SA. Immunology of paracoccidioidomycosis. *An Bras Dermatol*. 2011;86:516-24.
10. Benard G. An overview of the immunopathology of human paracoccidioidomycosis. *Mycopathologia*. 2008;165:209-21.
11. de Brito T, Franco MF. Granulomatous inflammation. *Rev Inst Med Trop Sao Paulo*. 1994;36:185-92.
12. Corvino CL, Mamoni RL, Fagundes GZ, Blotta MH. Serum interleukin-18 and soluble tumour necrosis factor receptor 2 are associated with disease severity in patients with paracoccidioidomycosis. *Clin Exp Immunol*. 2007;147:483-90.
13. Marchiori E, Zanetti G, Hochegger B, Irion KL, Carvalho AC, Godoy MC. Reversed halo sign on computed tomography: state-of-the-art review. *Lung*. 2012;190:389-94.
14. Teles FR, Martins ML. Laboratorial diagnosis of paracoccidioidomycosis and new insights for the future of fungal diagnosis. *Talanta*. 2011;85:2254-64.
15. Menezes VM, Soares BGO, Fontes CJF. Drugs for treating paracoccidioidomycosis. *Cochrane Database Syst Rev*. 2011:CD004967, <http://dx.doi.org/10.1002/14651858>.