

## Bilateral paracoccidioidomycotic iliopsoas abscess associated with ileo-colonic lesion

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### ABSTRACT

This case report shows the clinical development of a patient with systemic paracoccidioidomycosis presenting with lymphatic-intestinal manifestation. The patient initially had a substantial clinical improvement but had a recrudescence after six months of sulfamethoxazole-trimethoprim oral treatment, with the emergence of feverish syndrome, lumbar pain, and intermittent claudication, characterizing a bilateral iliopsoas muscle abscess, necessitating clinicosurgical therapeutics.

**Keywords:** Paracoccidioidomycosis. Psoas abscess. *Paracoccidioides brasiliensis*.

### INTRODUCTION

Paracoccidioidomycosis (PCM) is a systemic mycosis endemic in Latin America, whose agent is the dimorphic fungus *Paracoccidioides brasiliensis*, which manifests itself in a chronic presentation in over 90% of the cases. Its prevalence is greater among males from 30 to 60 years old, and lungs are the most frequent compromised organs. Its acute/subacute form with generalized lesions involves multiple organs and is rare and severe<sup>1,2</sup>.

We herein present a case of disseminated PCM, with involvement of lymphatic and gastrointestinal systems, and a subsequent development of bilateral iliopsoas muscle abscess (IPA), which constitutes a rare complication of the disease.

### CASE REPORT

A 24-year-old rural man presenting fever, asthenia, weight loss (20kg in the period), dull abdominal pain, diarrhea and hematochezia for six months. After four months of the initial symptoms, he developed disseminated subcutaneous nodules, which began draining a purulent secretion when he was admitted to our hospital.

On physical examination, we found bilateral cervical, inguinal, and axillary lymphadenomegaly and a painful abdomen at superficial palpation, especially in the right iliac fossa.

The laboratory tests revealed a hemoglobin level of 8.9g/dL; total leukocyte count of 6,000/mm<sup>3</sup> (eosinophil count of 60/mm<sup>3</sup>); C-reactive protein of 188mg/dL; viral serology negative for HIV and hepatitis B and C; nonreactive Venereal Disease Research Laboratory (VDRL) test; negative sputum alcohol acid-resistant bacilli (AARB) test in three samples (Auramine and Ziehl Neelsen); negative sputum

fungal smear; normal renal and hepatic function panels; and negative microbiological stool tests. An abdominal and pelvic computed tomography (CT) demonstrated mesenteric lymphadenomegaly, with a hypodense center. Colonoscopy evidenced multiple deep ulcers with changeable sizes and forms along the whole colon, intercalated by normal mucosa, and a deformed ileocecal valve, which favored the diagnosis of infectious ulcerative proctocolitis (**Figure 1**). Anatomopathological examination of the colonic mucosa revealed some areas of erosion covered in fibrino-leukocytic exudate, inflammatory infiltrate in corion, with some multinucleated giant cells, and lots of round shaped yeast, in various sizes, compatible with ileum-colonic PCM (**Figure 2**). A biopsy of a cervical lymph node also was performed, and in the anatomopathological examination, we found the *Paracoccidioides brasiliensis*, with non-caseating granulomas containing multinucleated giant cells and epithelioid histiocytes, presenting a steering wheel shape, compatible with PCM granulomatous lymphadenitis.

During hospitalization, the patient was treated with venous sulfamethoxazole-trimethoprim (SMX-TMP) and amphotericin B (amphoB) deoxycholate, receiving a cumulative dose of 1,900mg after

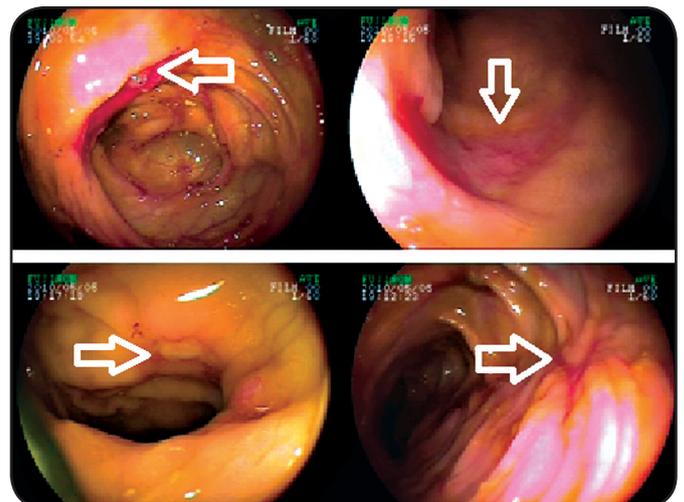


FIGURE 1 - Colonoscopy revealing the cecum and the ileocecal valve, a large ulcer, and an ulcer with fibrin (arrows).

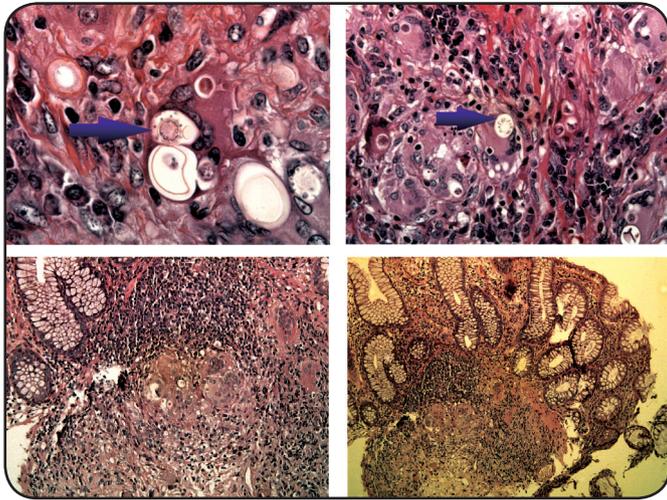
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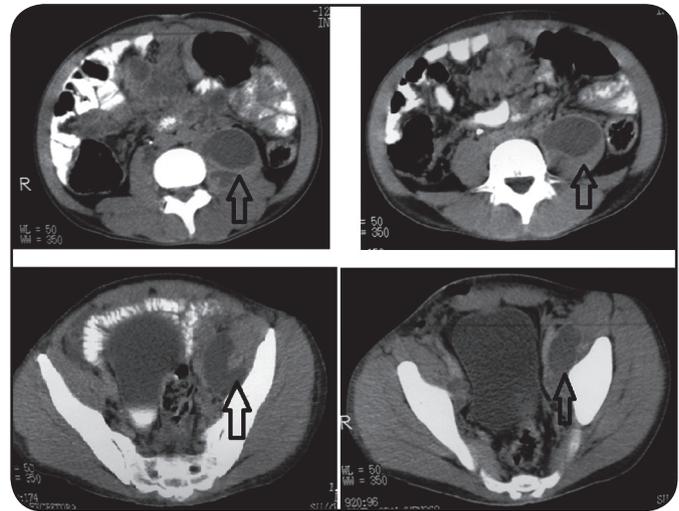
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**FIGURE 2 - Anatomopathological exam:** fragments of colonic and ileal mucosa demonstrating areas of erosion covered in fibrino-leukocytic exudate, inflammatory infiltrate in corion, with some multinucleated giant cells and many round-shaped yeast (compatible with *Paracoccidioides brasiliensis*) in various sizes (arrows), compatible with ileum-colonic paracoccidioidomycosis.



**FIGURE 3 - Abdominal and pelvic computed tomography scan:** mesenteric lymphadenomegaly, enlargement of the left iliac and psoas muscles, with inner voluminous liquefaction, which extended from the level of the third lumbar vertebra (L3) to the pelvis (arrows).

38 days. A complete regression of lymphadenopathy occurred, the patient gained weight, and the diarrhea ceased. After 40 days from admission, he was discharged, with a prescription of a daily use of SMX-TMP 800 + 160mg.

The patient returned 40 days later, presenting recrudescence of the initial symptoms, including pain in the left hip, limited extension of the homolateral hip, limping, and fever. Furthermore, cultures were obtained, and an anatomopathological examination of an inguinal lymph node biopsied revealed *Paracoccidioides brasiliensis*. He was rehospitalized and treated with amphotericin B plus oral itraconazole and sulphadiazine.

Abdominal and pelvic CT showed mesenteric lymphadenomegaly, enlargement of the left iliac and psoas muscles, with inner voluminous liquefaction, which extended from the level of the third lumbar vertebra (L3) to the pelvis. Moreover, a smaller collection was observed in the right psoas muscle at L3 level, and left pyelocaliceal dilation was present (**Figure 3**).

An ultrasound (US)-guided lumbar percutaneous puncture of the collection was performed at L3 level, and a Cystofix® 12 Fr drain was placed inside it. There was immediate drainage of 100ml of thick purulent secretion, which revealed the presence of *Paracoccidioides brasiliensis*. Bacterial and fungal cultures of this specimen were negative.

The patient kept the initial symptomatology after eight days of drainage, being therefore submitted to a new CT scan, that demonstrated reduction of the left psoas abscess, but another collection was found involving the homolateral iliac muscle. He was then submitted to an US-guided inguinal puncture of this abscess, using 14G Jelco, with immediate drainage of 50ml of purulent secretion. The lumbar drain was left in place for 18 days.

Control CT scan 20 days after drainage showed minimal residual bilateral iliopsoas collection, and the pyelocaliceal dilation was no longer observed. After having received a cumulative dose of 2.9g of amphotericin B and 60 days of oral itraconazole and sulfadiazine, he was discharged.

Ambulatory follow-up six months after treatment showed no signs of relapse, maintaining oral itraconazole and SMX-TMP usage.

## DISCUSSION

Paracoccidioidomycosis can manifest itself in two distinct clinical forms: acute/subacute or chronic. The acute/subacute form represents 3 to 5% of PCM cases, being more common in the first three decades of life, especially in children<sup>3</sup>. It evolves rapidly, with significant general state impairment, fever, hepatosplenomegaly, lymphadenomegaly, together with digestive, osteoarticular, and cutaneous manifestation. Mesenteric lymph node hypertrophy can lead to bowel obstruction or choledochal compression. Moreover, it can impair oral drugs and nutrient absorption, aggravating the nutritional state of the patient.

This report evidences PCM involvement in an immunocompetent young adult presenting a subacute severe disseminated form, with colitis and systemic lymphadenomegaly, especially in the retroperitoneum, which fistulized not only to the skin but also to the lumbar musculature. The maintenance of the *Paracoccidioides brasiliensis* foci was strictly related to the relapse of a severe form of PCM, requiring prolonged clinicosurgical therapeutics to control the infection.

The therapeutic choice depends on the severity of the PCM, and the options are sulfas, azolics, and amphotericin B. The great efficacy of the sulfa and azolic drugs justifies the restricted use of amphotericin B, which is more toxic and is exclusively parenteral. Amphotericin B is reserved for more severe and systemic forms, which compromise structures beside the lungs, mucosa, and skin, and when there is imminent risk of death. Only a few studies have been held in order to define its optimal treatment. Only two randomized trials have been published so far, neither of which have collected sufficient data to assess response or cure rates<sup>4,5</sup>. The duration of treatment is still a matter of debate, since no randomized trials have compared different treatment durations, and are based mainly on data from open studies and specialist opinions<sup>6</sup>. In this report, the patient had initially been treated with venous SMX-TMP and amphotericin B because of the severity of the manifestations, and there was a great initial response. After being discharged, a prolonged oral SMX-TMP therapy proceeded, with a

good compliance. However, at this stage of treatment, the disease relapsed, manifesting as *cold* retroperitoneal abscesses. Therapeutics at that moment was based on antifungal association because of the disease severity and life threatness.

The iliopsoas muscle abscess (IPA) is a retroperitoneal collection in the space defined by the iliac and psoas muscles, usually determined by a bacterial infection. It is considered a rare medical entity; nonetheless, with the advent of CT scan for evaluation of patients with sepsis with undetermined focus, the diagnosis of IPA has become more frequent. Its emergence is related to two mechanisms: primary, with hematogenic spread, and secondary, when related to adjacent structures infection.

The diagnosis, evolution and treatment of IPA are well described for bacterial infection. Iliopsoas abscess related to PCM is even more rare and there is little information about this PCM entity<sup>7</sup>. In the patient reported, the most likely mechanism of IPA formation was the fistulization of retroperitoneal lymph nodes into the iliopsoas muscle compartment, simultaneous to cutaneous fistulization of the superficial lymph nodes.

The classical triad symptoms, characterized by fever, lumbar pain, and claudication, are present in only 30% of the cases<sup>8</sup>. Other symptoms include inguinal bulge, malaise, anorexia, and weight loss. In 60% of the cases, the US confirms the diagnosis, with the CT scan considered the gold standard for diagnosis.

Historically, the initial IPA treatment was based on extraperitoneal surgical drainage, associated with adequate antimicrobial therapy. The first IPA radiologic (US, CT) guided percutaneous drainage occurred in 1984<sup>9</sup>, and since then, the best therapeutic option has been on debate. So far, the treatment choice is the guided percutaneous drainage, as it is the least invasive and associated with lower morbidity and mortality<sup>10</sup>; however, many local and systemic factors of each patient may influence the therapeutic choice<sup>11</sup>.

After the first drainage, there was only partial improvement in the patient reported, requiring the repetition of the CT scan, which showed that the left IPA was composed of two distinct compartments, requiring a new US-guided percutaneous puncture. Therapeutic failure after percutaneous drainage is rare, as described by Castadimir et al, who revealed that this method was efficacious in 21 of 22 patients; nonetheless, when it happens, surgical treatment is indicated<sup>12</sup>. The right IPA was of little volume, and we opted to treat it with antibiotics only, a plausible treatment for small abscesses.

The main complications of IPA are deep venous thrombosis, bowel obstruction, ureteral obstruction, and sepsis. In this case, pyelocaliceal dilation was due to extrinsic compression and was fully reverted after drainage, with no renal sequela.

Paracoccidioidomycosis reveals itself as a variable clinical entity that ought to be remembered in the clinical diagnosis of a localized or systemic disease, with possible involvement of the lungs, skin, mucosa, central nervous system, retroperitoneal musculature or lymph nodes, intestine, adrenal, kidneys, or prostate and likely association with tuberculosis, neoplasms, and sepsis. In this report, we presented

a patient with multiple organ compromise because of disseminated PCM, apparently with antifungal resistance, necessitating broad-spectrum antibiotics and radiologic-guided percutaneous drainage of *cold* retroperitoneal abscesses, who evolved with slow recovery despite all therapeutics. Although the first description of PCM goes back over 100 years, its optimal treatment is not yet well-defined. The lack of information about diagnosis and treatment may contribute to the development of unusual forms of presentation that are capable of producing significant damage and death.

## ABSTRACT IN PORTUGUESE

### Abscesso paracoccidioidomicótico bilateral do iliopsoas associado ao acometimento íleo-colônico

Este relato de caso descreve a evolução clínica de paciente com paracoccidioi-domicose sistêmica com manifestação linfática-intestinal. O paciente evoluiu inicialmente com melhora clínica acentuada e recrudescência após seis meses de uso de SMX-TMP pela via oral, com o surgimento de síndrome febril, dor lombar, e claudicação intermitente, caracterizando um abscesso bilateral do músculo ileopsoas, com necessidade de terapêutica clínico-cirúrgica.

**Palavras-chaves:** Paracoccidioidomycose. Abscesso do psoas. *Paracoccidioides brasiliensis*.

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