

LATE OUTCOME OF *PARACOCCIDIOIDES BRASILIENSIS* ISOLATED INFECTION ON THE HIP

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SUMMARY

Infections caused by *Paracoccidioides brasiliensis* are always a diagnostic challenge, even in endemic areas due to its multiple clinical presentations and involved organs.

This paper describes the evolution of the only case found in literature in which the disease affected just one hip.

Keywords: Paracoccidioidomycosis; Osteomyelitis; Hip joint.

INTRODUCTION

Paracoccidioidomycosis is an infection rarely involving locomotive apparatus. The disease is caused by *Paracoccidioides brasiliensis*, and is usually neglected or confused with other neoplasias or bacterial infections.

The objective of this study is to describe the evolution of a single case of paracoccidioidomycosis reported in literature⁽¹⁾, which affected hip joint as the only manifestation of the disease.

CASE REPORT

A 22 year-old, female, mulatto, rural worker patient, reports a progressive pain on right hip starting 6 months before, getting worse upon movement and restraining her job activities. At physical examination, she revealed a pale mucosa, hypotrophic right thigh, hip movement restrain at 100° flexion, 0° extension, 30° abduction, 15° adduction, 20° inner rotation, 20° outer rotation. Negative Thomas & Trendelenburg's test. No palpable lymphonode or visceromegaly.

Pelvic X-ray images evidenced a well-bounded lithic injury at the lower region of the right femoral neck trespassing adjacent cortical bone, with absence of periosteal response,

reduction of joint space and flattening of the upper region of femoral head (Figure 1). The hemogram showed changes evidencing anemia (Hb 9,5g/dL), leukocytosis (14,000/mm³ with neutrophilia) and increased blood-sedimentation speed (58 mm/h). Initial diagnostic suspects were towards hematogenic sub acute osteomyelitis or neoplasia. Magnetic resonance imaging showed an extensive injury involving femoral head and neck, with bone destruction and infiltration of adjacent soft tissues, joint edema and capsule thickening. Aspiration biopsy of the injury was performed, and the histological analysis revealed fragments of fibrous-connective tissue showing a chronic granulomatous process and a florid reaction of giant cells. Rounded structures birefringent similar to yeasts with characteristics of *Paracoccidioides brasiliensis* spores were seen. Counterimmunoelectrophoresis specific to paracoccidioidomycosis was positive for dilutions above 1/4.

No other supplementary investigative test (chest X-ray, cranial, chest and abdominal computed tomography) detected additional injuries in any other organ. Similarly, the tri-phase bone scintiscan with technetium (Tc 99m) showed only an increased captivation of the radiopharmaceutical on the right

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hip, with absence of other injuries in any portion of the skeleton, which led us to the conclusion that this was a localized form of the disease.

The non-surgical treatment was initiated with daily doses of sulfamethoxazole 2400mg combined with trimethoprim 480mg for 2 years. After 5 months of therapy, an important improvement of the symptoms was noticed by the patient, and pelvic X-ray image showed that the lithic area of the femoral neck was partially filled by bone, with only a remaining reduction of joint space (Figure 2). After two years, when drug therapy was finished, patient was shown to be asymptomatic, pelvic X-ray images

showed remodeling on femoral neck region (Figure 3) and when comparing magnetic resonance images at baseline and two years later, a reduction of bone edema both at the acetabulum and at femoral head and neck, of joint edema, and of the response on adjacent soft tissues (Figure 4). The counterimmunoelectrophoresis specific to paracoccidioidomycosis was negative.

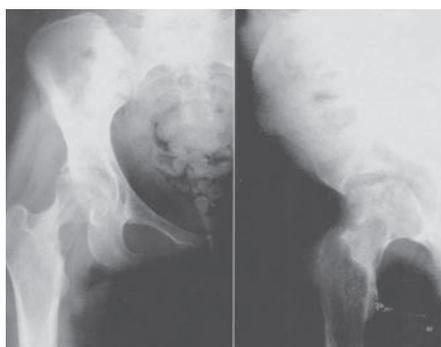


Figure 1 – Hip X-ray image at baseline, showing a lithic injury at the lower region of femoral neck, and a flattening on the anterosuperior region of femoral head.



Figure 2 – X-ray image of the hip showing partial recovery of femoral neck injury and joint space reduction in 5 months of treatment.



Figure 3 – X-ray images of the hip showing neck remodeling, restored roundness of femoral head, with joint space reduction after 2 years of treatment.

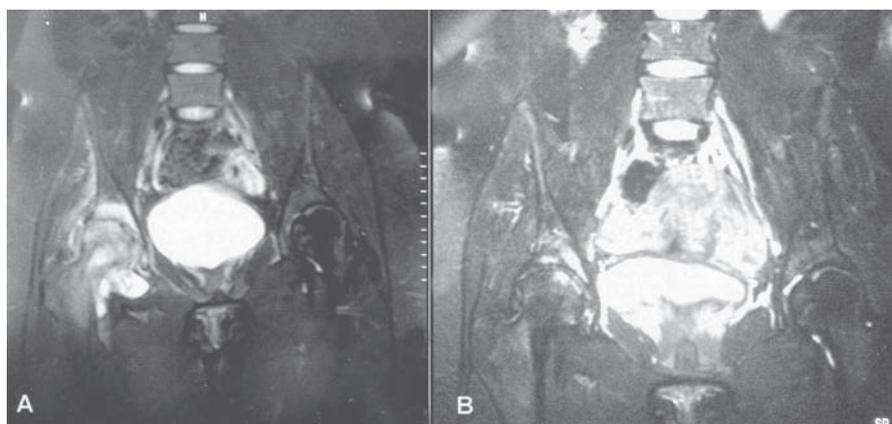


Figure 4 – Magnetic resonance image, time T2 with fat saturation, (A) pre- and (B) post-treatment, showing injury recovery with bone edema reduction, both at femur and at acetabulum, as well as the reduction of joint edema and of the lithic area at lower neck region.

DISCUSSION

Most of the information available about bone-joint injuries caused by *Paracoccidioides brasiliensis* were observed in disseminated forms of the disease, with the involvement of multiple organs. Isolated bone-joint affection forms of the paracoccidioidomycosis are rare events and little described in literature. Figueiredo et al.⁽²⁾, by analyzing data gathered from Medline, Embase and Lilacs within the period of 1966 to January 2001, studied 345 cases of joint fungal infections, but only five cases were due to *Paracoccidioides brasiliensis*. Amstalden et al.⁽³⁾ published nine paracoccidioidomycosis cases in which the locomotive apparatus

was the major clinical manifestation of the disease, and, only in two (one involving the femur and the other involving the elbow) no injuries were found in other organs.

Paracoccidioides brasiliensis is commonly found in South American and Central American soils. Contamination occurs via inhalation and the majority of the cases is limited to a self-limiting lung infection, with few or no symptoms. In some cases, especially in immunodepressed individuals, the disease

may evolve to chronic pulmonary infection or may be spread to other organs such as liver, spleen, lymphonodes, adrenal glands, joints and bones, among others.

The appearance of the bone injury at X-ray images is described as a lithic, well-bounded area, with or without sclerotic borders that may occur in any bone, but commonly affects clavicle, scapula and ribs ^(4,5). Differential diagnosis for this kind of injury include bacterial osteomyelitis, tuberculosis, lymphoma, and osteosarcoma ⁽⁴⁾.

Diagnosis is confirmed when the microorganism is found, so, it is mandatory to perform a biopsy of the involved tissue. Fulciniti et al. ⁽⁶⁾ reported the first case in which the presence of the fungus was confirmed by aspiration biopsy with thin needle.

Therapy with anphotericin B is effective, but causes frequent complications such as fever, trembling and nephrotoxicity, and should be reserved for more severe forms of the disease ^(3,7). Sulfametoxazole combined with trimethoprim, and ketoconazole are more frequently

used, and treatment must last at least two years, according to literature ⁽⁷⁾. The absence of fungi in subsequent microscopic examinations or negative counterimmunoelectrophoresis tests are important parameters for evaluating treatment effectiveness. However, there are no criteria confirming that the patient is cured of paracoccidioidomycosis. The possibility of recurrences ⁽⁸⁾ justifies patients being followed up for long periods.

In brief, the involvement of the locomotive apparatus by *Paracoccidioides brasiliensis* as the only clinical manifestation of the disease is an uncommon event, and few orthopaedic doctors are experienced with this kind of condition, either for providing a diagnosis or for recommending a therapy. This fact is also relevant in non-endemic areas, because the continuous migration of people may change territorial distribution of infectious diseases. This case report shows the evolution of the first case described in literature where the hip was identified as the only site affected by the disease.

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