

# Tropical pyomyositis in a patient with systemic lupus erythematosus and HTLV 1/2 infection

Hérica Cristiani Barra de Souza<sup>1</sup>, Barbara Nascimento de Carvalho<sup>1</sup>, Marina Gonçalves Veras de Morais<sup>1</sup>,  
Guilherme Zager Monteiro<sup>1</sup>, Fabiano Takaaki Emori<sup>2</sup>, Luiz Carlos Latorre<sup>3</sup>

## ABSTRACT

Tropical pyomyositis (TP) is an unusual infectious disease of skeletal muscles, caused by bacteria, and often associated with immunodeficiency conditions. The involvement of deep pelvic muscles, such as the iliac muscle, is even rarer. The association of systemic lupus erythematosus (SLE) and PT is seldom reported in the literature. Because SLE involves a state of immunosuppression resulting from both the disease itself and its medicamentous treatment, SLE patients are at higher risk for developing infections, such as PT. Infection by HTLV 1/2 is increasingly identified and associated with autoimmune diseases, such as SLE. This is a case report of PT in the pelvic muscles of a female patient with SLE, chronic kidney failure, on hemodialysis, and HTLV1/2 infection, admitted to the Hospital Heliópolis, in the city of São Paulo, Brazil.

**Keywords:** pyomyositis, systemic lupus erythematosus, human T-lymphotropic virus 2, human T-lymphotropic virus 1.

[Rev Bras Reumatol 2011;51(1):97-103] ©Elsevier Editora Ltda.

## INTRODUCTION

Tropical pyomyositis (TP) is an infectious disease of skeletal muscles, whose major agent is *Staphylococcus aureus*. It is typical of developing countries and of tropical areas. Tropical pyomyositis is not common and is often associated with immunodeficiency. It affects mainly young adults and children, with a preference for large muscle groups.<sup>1-4</sup>

Tropical pyomyositis is classified as primary and secondary. The primary form is due to hematogenous dissemination favored by basic immunodepression, *S. aureus* being its major agent. The secondary form is due to the extension of an infection from neighboring structures, also favored by immunodepression or post-operative conditions, and its most frequent agents are *Escherichia coli* and *Bacteroides spp.*<sup>3,4,6</sup>

The association between systemic lupus erythematosus (SLE) and TP has been rarely reported in the literature. Systemic lupus erythematosus is an immunodepression condition due to both the disease itself and its medicamentous treatment. Thus, SLE patients are at risk for developing infections.<sup>7,8</sup>

The authors report a case of TP in the iliac muscle of a female patient with SLE, chronic kidney failure, on hemodialysis, and HTLV1/2 infection, as a complication of the underlying disease.

## CASE REPORT

VSF is a 25-year-old female patient, originating from the rheumatology outpatient clinics of the Hospital Heliópolis, in the Brazilian city of São Paulo.

Submitted on 01/07/2010. Approved on 11/15/2010. We declare no conflict of interest.

Complexo Hospitalar Heliópolis, São Paulo, SP, Brazil.

1. Medical resident at the Service of Rheumatology of the Hospital Heliópolis - Medical Residency

2. Medical resident at the Service of Radiology at the Hospital Heliópolis, São Paulo, Brazil

3. Assistant physician of the Service of Rheumatology and director of the Medical Clinics of the Hospital Heliópolis; PhD in Epidemiology by the Faculdade de Saúde Pública of the Universidade de São Paulo

Correspondence to: Hérica Cristiani Barra de Souza. CEREDO - Serviço de Reumatologia do Hospital Heliópolis. Rua Caçapava, nº49, Conj. 17. Jardim Paulista, São Paulo, SP, Brazil. CEP: 01408-010. E-mail: hericabarra@yahoo.com.br.

The patient reported the following personal history: SLE for six years; nephritis for five years; chronic kidney failure (on hemodialysis for four months); positive serology for HTLV 1/2 detected four months before; and chronic demyelinating polyradiculopathy diagnosed by use of electroneuromyography four months before. The patient also reported two hospitalizations as follows: from 01/03/2009 to 01/20/2009, due to severe community pneumonia (CP); and from 03/04/2009 to 05/01/2009, due to cutaneous vasculitis, CP, and sepsis resulting from a catheter-related infection, when multiresistant *S. aureus* was isolated. The patient was then treated with vancomycin for 21 days.

The patient reported that, in the preceding month, a nodule appeared in the anterior face of the right leg, with progressive volume increase, pain, and local rash. In the last week, the lesion worsened and was accompanied by chills, fever, and pain in the right hip, which worsened with movement. The patient's physical examination showed a regular general health state, pale mucosae (2+/4+), diffuse abdominal tenderness on deep palpation, no signs of peritoneal irritation, muscle atrophy in upper and lower limbs, and grade III muscle strength in lower limbs and grade IV in upper limbs. Each pretibial region (right and left) showed a 3-cm nodule, which was soft, red, warm, and tender to touch (Figure 1).

The nodule was punctured and the purulent content released was sent to analysis. Treatment with Clindamycin was then started.

Initial laboratory tests revealed the following: significant leukocytosis (19,280 cells per mm<sup>3</sup>) with left shifting; lactic dehydrogenase of 751U/L; urinalysis showing 230,000



**Figure 1**  
Presence of abscesses; pretibial abscesses in a patient with systemic erythematosus lupus and HTLV-1/2 infection.

leukocytes; negative urine culture; and negative serology for HIV.

Bacterioscopy of the abscess content revealed intracellular Gram-positive cocci in pairs, which, through culture, proved to be multiresistant *S. aureus* (sensitive to vancomycin). That agent was also identified on the initial samples of blood culture and hemodialysis catheter-tip culture.

Computed tomography (CT) of the pelvis showed bilateral sacroiliitis, bilateral collection on the topography of the iliac muscle, and signs of osteomyelitis of the sacroiliac joints (Figure 2A). That finding was also present on the abdominal ultrasound performed on 07/29/2009.

After bacterioscopy, the antibiotic was changed to vancomycin (1 g every four days, dose adjusted to kidney function). Based on the results of the abdominal ultrasound and pelvic CT, open surgical drainage was performed on 07/29/2009, with lateral access to the retroperitoneum and bilateral placement of a tubular-laminar drain. The collection content was sent to culture, which also evidenced the presence of multiresistant *S. aureus*. Antibiotic was maintained for 42 days. The patient showed progressive clinical and radiological improvement, and was discharged from the hospital on 09/04/2009, with satisfactory clinical recovery, negative cultures, and a significant reduction in the lesion on pelvic CT (Figure 2B).

## DISCUSSION

Tropical pyomyositis is a severe pathology that most frequently affects the thigh, gluteal, and trunk muscles; impairment of a single muscle is the rule (60%-70%).<sup>9</sup>

Three stages characterize the disease. The first presents as initial myositis without abscess formation, accompanied by low fever, pain, and muscle stiffness. Diagnosis is rarely established at this phase. At stage 2, a deep single (or multiple) intramuscle abscess forms, usually 10 to 21 days from symptom onset, accompanied by fever, muscle edema, and more localized pain. Aspiration reveals a purulent material. Most diagnoses are made at this stage, as was the case of our patient, who had primary stage 2 TP.<sup>4,5,10</sup>

If not properly treated, the patient can evolve to stage 3, characterized by high fever, severe pain, fluctuation of the muscles involved, and signs of toxicity, which can progress to septic shock, and, eventually, death.<sup>1,2,5,9</sup>

In the case reported, *S. aureus* was isolated in blood culture samples and abscess aspirate. That finding is in accordance with data from the literature suggesting that to be the etiologic agent in more than 90% of the cases, followed by *Streptococcus spp*,

and *E.coli*. Although the mechanism of muscle aggression involves bacteremia, cultures are positive in less than 5% of the cases, suggesting that such bacteremia is transient.<sup>1,4,6,9</sup>

In addition to the most affected muscle groups already cited, involvement of the following muscles has been reported:

paravertebral, cervical, iliac, psoas, piriform, abdominal, calf, deltoid, and biceps. Although extensive muscle areas are often affected, usually no elevation of muscle enzymes occurs.<sup>1,2,9,11</sup>

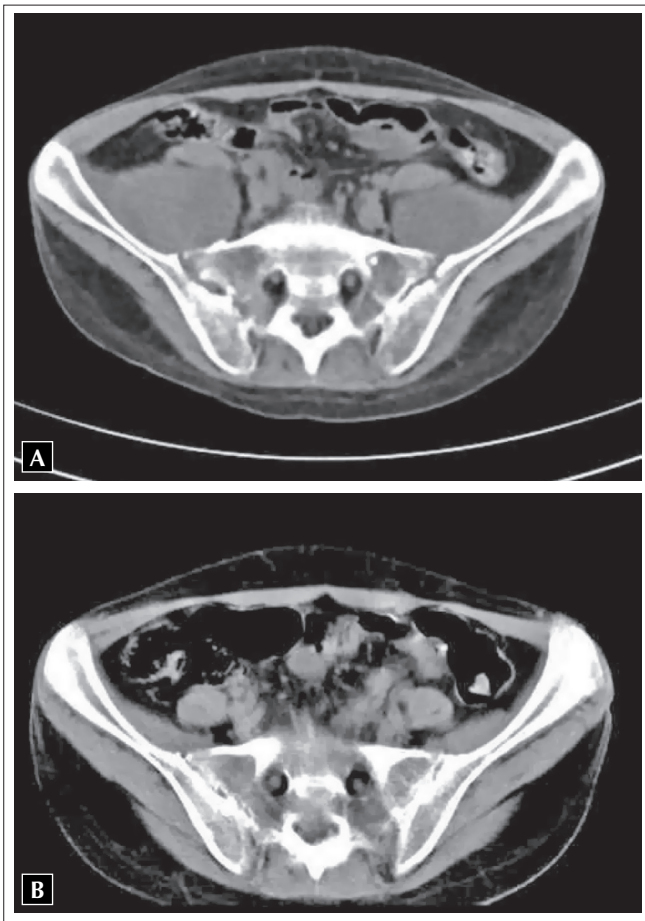
Brazil has approximately 2.5 million HTLV 1/2-seropositive individuals, and that pathology is endemic in the city of Salvador, Bahia state (1.8% of prevalence).<sup>12,13</sup>

Some studies have suggested that HTLV is the trigger and perpetrator of some articular inflammatory diseases, because it induces an exuberant lymphocytic proliferative response in the immune system. Thus, it has been implicated in autoimmunity induction. Of the autoimmune rheumatic diseases associated with that infection, the following stand out: Sjögren's syndrome, rheumatoid arthritis, polymyositis, and SLE. In 2006, Carvalho *et al.*<sup>14</sup> reported that, in their case series, 0.7% of the patients with HTLV-1 had SLE, but most studies are unable to establish such association so precisely. In the case of our patient, the HTLV coinfection was an adjuvant of SLE for immunosuppression, favoring the opportunistic infection.

To help in diagnosing TP, image exams can be used, preferably ultrasound and CT, which are useful to locate the affected muscle and to guide puncture and biopsy. The exams should be serially performed for clinical and treatment follow-up.<sup>1,2,9,10</sup> In our case, the iliac muscle involvement was identified on pelvic CT, and the abscess volume was confirmed on ultrasound.

Therapy for TP is based on antibiotics and percutaneous or surgical drainage of the abscess. Puncture with surgical drainage seems fundamental in most cases, along with antibiotic therapy, to avoid disease progression to stage 3, in which mortality increases.<sup>4</sup> Initially, antibiotic therapy was chosen, delaying surgical drainage, because of the patient's high surgical risk due to her underlying pathologies. In addition, the lateral surgical access through the retroperitoneum is also technically difficult. However, clinical and radiological worsening occurred and the surgical procedure was successfully performed. Antibiotic therapy was maintained in the post-operative period.

Tropical pyomyositis should be remembered as a severe complication in immunosuppressed patients. Its insidious onset can delay diagnosis, limiting prognosis. Early clinical suspicion, imaging diagnosis, and adequate therapy are decisive for the satisfactory outcome of such cases.



**Figure 2**  
**(A)** Computed tomography of the pelvis. Huge collection in the iliac muscle bilaterally, and osteomyelitis of the sacroiliac joints in a patient with systemic erythematosus lupus and HTLV-1/2 infection. **(B)** Computed tomography of the pelvis. Aspect of the lesions described in Figure 2A; after surgical drainage of pyomyositis of the iliac muscle bilaterally.

## REFERENCES

### REFERÊNCIAS

1. Takayasu V, Onuchic MHF, Campos FPF, Albuquerque RP. Miosite tropical. *Rev Hosp Clin Fac Med São Paulo* 1993; 48:112-15.
2. Marques GC, Filho GLM, Valiatti M, Hamra A, Valiatti J, Mastrocola Jr A *et al.* Piomiosite tropical: estudo retrospectivo de 27 casos. *Rev Bras Reumatol* 1995; 35:193-200.
3. Hernández FJG, Román JS, Medina CO, Romero LM, López JM, Wichmann I. Absceso del músculo iliopsoas y lúpus eritematoso sistêmico. *An Med Interna* 2003; 20(4):198-200.
4. Chiedozi LC. Pyomyositis: review of 205 cases in 112 patients. *Am J Surg* 1979; 137:250-55.

5. Isdale AH, Foley DF, Butt WP, Wright V. Psoas abscess in rheumatoid arthritis - an inperspicious diagnosis. *Brit J Rheumatol* 1994; 33(9):853-58.
6. Berge VM, Marie S, Kuipers T, Jansz AR, Bravenboer B. Psoas abscess: report of a series and review of the literature. *Neth J Med* 2005; 63(10):413-16.
7. Walsh TR, Reilly JR, Hanley E, Webster M, Peitzman A, Steed DL. Changing etiology of iliopsoas abscess. *Am J Surg* 1992; 163:413-16.
8. Iliopoulos AG, Tsokos GC. Immunopathogenesis and spectrum of infections in systemic lupus erythematosus. *Semin Arthritis Rheum* 1996; 25:318-36.
9. Marques GC, Valiatti M, Pereira ER, Barbieri V, Mauro FGL. Piomiosite tropical de localização cervical em pacientes com Lupus Eritematoso Sistêmico. *Rev Bras Reumatol* 1996; 36:47-50.
10. Braz AS, Fernandes JMC, Couto RAL, Szajubok JCM, Moura Jr JE, Chahade WH. Piomiosite tropical: descrição de quatro casos. *Rev Bras Reumatol* 2000; 40:81-6.
11. Tokuda N, Himura I, Harada M, Tsubaki H, Matsushima M, Kuwashima A. Pyomyositis of iliopsoas muscle with infection of renal cyst: a case report. *Hinyokika Kyo* 1994; 40:139-4.
12. Cruz BA, Catalan-Soares B, Proietti F. Manifestações reumáticas associadas ao vírus linfotrópico humano de células T do tipo I (HTLV-I). *Rev Bras Reumatol* 2005; 45 (2):71-7.
13. Romanelli LCF, Caramelli P, Proietti ABF. O vírus linfotrópico de células T humanos tipo I: quando suspeitar da infecção? *Rev Assoc Med Bras* 2010; 56(3):340-7.
14. Carvalho MMN, Giozza SP, Santos ALMA, Carvalho EM, Araújo MI. Frequência de doenças reumáticas em indivíduos infectados pelo HTLV 1. *Rev Bras Reumatol* 2006; 46(5):315-22.