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**Tropical Pyomyositis: case report**

**Piomiosite tropical: relato de caso**

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ABSTRACT

# Introduction: Tropical pyomyositis is an infectious disease that affects the skeletal muscle, appearing as a diffuse inflammation or a rapidly progressive myonecrotic process. Staphylococcus aureus is the most common microorganism, representing 90% of cases. The diagnosis can be delayed because patients generally do not seek care for the first symptoms, and because it is a rare disease, doctors are still not familiar with the entity. Case report: The case refers to a 42-year-old male patient with diabetes mellitus, hypothyroidism, anemia, thrombocytopenia, and hypoalbuminemia who developed tropical pyomyositis with multiple muscle abscesses, requiring prolonged antibiotic therapy and surgical drainage. Conclusion: Pyomyositis is not a well know disease and can be fatal if not diagnosed early.

**Keywords:** Tropical pyomyositis, muscle abscess, muscle infection.

RESUMO

Introdução: A piomiosite tropical é uma doença infecciosa que afeta o músculo esquelético, aparecendo como uma inflamação difusa ou um processo mionecrótico rapidamente progressivo. Staphylococcus aureus é o microorganismo mais comum, representando 90% dos casos. O diagnóstico pode ser tardio porque os pacientes geralmente não buscam cuidados com os primeiros sintomas e, por ser uma doença rara, pode surpreender um médico ainda não familiarizado com a entidade esta clínica. Relato de caso: o caso refere-se a um paciente do sexo masculino com 42 anos de idade com diabetes mellitus, hipotireoidismo, anemia, trombocitopenia e hipoalbuminemia que desenvolveu pimiosite tropical com múltiplos abscessos musculares, e que necessitou de antibioticoterapia prolongada e drenagem cirúrgica. Conclusão: A piomiosite não é uma doença bem conhecida e pode ser fatal se não for diagnosticada precocemente.

**Palavras-chave:** Piomiosite tropical, abscessos musculares, infecção muscular.

INTRODUCTION

Sriba, in 1885, was the first to describe pyomyositis, a disease predominant in tropical regions and rarely found in temperate regions. Currently, this pattern of incidence has been changing, and pyomyositis has also been found in temperate climates1.

Under normal circumstances, skeletal muscle tissue is intrinsically resistant to bacterial infections. In the tropical pyomyositis, the microorganisms reach the skeletal muscles during a transient bacteremia, finding favorable conditions and triggering the infection2,3,4.

The microorganism most commonly found in crops is Staphylococcus aureus, appearing in about 90% of cases in tropical areas and 60% in non-tropical regions.5

Tropical pyomyositis usually affects individuals with some immunological deficiency, becoming an important complication among those affected by the human immunodeficiency virus (HIV), which is facilitated by the infection itself, antiretroviral therapy, or infections caused by parasites and mycobacteria.6

The tropical disease occurs in all age groups with a peak incidence in two of them, 2 to 5 years and 35 to 40 years, with a male to female ratio of 4: 1. The non-tropical disease presents a peak of 30-50 and 60-70 years, with a 3: 1 ratio2.

Tropical pyomyositis is not a well know disease yet, and its initial characteristics are very nonspecific, easily being confused with other more prevalent pathologies, including arboviruses. A delay in the recognition of the disease results in a greater quantity of examinations, interventions and days of hospitalization, generating high costs for the institution, besides negatively influencing the prognosis, because of the imminent risk of sequelae or death.

The objective of the report is to describe the case of a patient hospitalized with tropical pyomyositis at a Tertiary Hospital in the municipality of Juazeiro do Norte - Ceará, and to emphasize the importance of an early diagnosis, as well as the quick institution of treatment.

CASE REPORT

The information contained in this study was obtained through a review of the medical records and related literature. The data were only obtained after approval of the Ethics Committee in Research and the signing of the secrecy Term of the medical record.

A 43-year-old male patient, Caucasian, obese, married, native from Juazeiro do Norte - Ceará, with a case of diabetes mellitus, hypertension and hypothyroidism diagnosed approximately 3 years ago, using levothyroxine 125mcg, metformin 500mg per day, enalapril 20mg/day and simvastatin 40mg / day. It started 16 days ago, with symptoms of asthenia and diffuse myalgia, without association with other signs or symptoms, obtaining partial improvement with analgesics. On the next day, it evolved with increased pain in the lower limbs, more prominent on the left, radiating to the ipsilateral gluteal region, making it difficult to ambulate and not being responsive to the usual analgesics. Subsequently, it was reported a pain increase and an edema in the knee and in the right elbow. On the third day, from the beginning of the treatment, he was unable to ambulate due to pain (Analog Visual Scale: 10).

He presented a normal condition in general at the examination, hypoxic +/4+, afebrile, blood pressure 160x90mmHg, capillary glycemia 180mg/dl. Cardiac and respiratory prophylaxis without alterations.

Right lower limb: Edema 3+ /6+, painful to the digitopression in the leg and thigh region, with hyperemia, increase in the temperature of the limb and difficulty of movement of the knee and hips:

* Lower left limb: Edema 2+ /6+ in the calf, with increased temperature to the touch, hyperemia and pain the digitopression and movement of the foot;
* Right upper limb: semi-flexed limb with restriction of elbow extension due to pain, 2+ /6+ edema, increased temperature, digitopression pain and hyperemia in the joint region;
* Upper left limb: edema + /6 +, with increased temperature in the region of the elbow joint, without limitation of movement (Table 1).

In the emergency room, puncture was performed in the right knee with the exit of purulent contents, requiring surgical drainage of the joint. Antibiotic therapy was started with ceftriaxone 2g 24/24h and oxacillin 2g 4/4h, with hospitalization.

In view of the extension of the clinical picture, it was performed an investigation through images of the inflammatory foci, and multiple muscle abscesses were visualized (Table 2). Early nuclear magnetic resonance was not feasible due to intense pain during the positioning, even with medication, being initially performed only USG and RNM one week after.

After one week of use of ceftriaxone and oxacillin, it was not observed an improvement in pain and/or reduction of edema/inflammation, and Meropenem 1g 8/8h and vancomycin 1g 12/12h were prescribed. The results of the blood cultures were negative, preventing guided antibiotic therapy.

A conservative treatment with the new antibiotic regimen was observed for another month. A significant reduction of abscesses in the upper limbs was observed, but an unsatisfactory response was obtained in the reduction of abscesses in the gluteus and left trochanter and right thigh, and a surgical approach was required for drainage.

Antibiotic therapy was prolonged, totaling three months, when a complete reduction of all abscesses and infiltrative procedures was obtained and the patient was discharged (Table 2).

DISCUSSION

Reports of blunt trauma or vigorous exercise of the muscle groups involved are reported in 20-50% of cases. It is also suspected that a change in the immune system may be a causal factor, especially by the inadequate action of T lymphocytes against the staphylococcus7. In this case, there was no history of recent trauma, which would fit as the entrance door for the infection, and a transthoracic echocardiogram was performed to exclude the formation of emboli due to bacterial endocarditis. Among immunity modifying factors, there were diabetes mellitus and decompensated hypothyroidism (ultra-sensitive TSH: 66.17mIU /L), which may be involved as facilitators of the spreading of the infection. An autoimmune disease research was started, after finding an anti-nucleus factor and reagent chromosomal metaphase plate with title of 1/1280 - homogeneous nuclear pattern, splenomegaly, thrombocytopenia before diagnosis. This last investigation was not conclusive until the moment of the hospital discharge, being referred for outpatient follow-up.

In addition, the patient had anemia and hypoalbuminemia (Table 1). Another important factor for the development of pyomyositis is the presence of a deficient nutritional status, as well as the presence of anemia and hypoproteinemia (hypoalbuminemia), factors observed with a significant factor among patients studied in Nigeria and India8,9. The diagnosis is sometimes delayed because patients usually do not seek care during the first symptoms, and because it is a rare disease, doctors are still not familiar with the entity. For the differential diagnosis of pyomyositis, it is important to observe the local epidemiology, since it varies from arbovirus to osteomyelitis, deep vein thrombosis, cellulitis, hematoma, tumors, synovitis, septic arthritis and, for iliopsoas pyomyositis, appendicitis, diverticulitis and others10. At the onset of the disease, signs and symptoms were like the arboviruses ones, especially considering the local epidemiology. With the clinical worsening of the edema and knee pain, and after the puncture of suppurative content, the hypothesis of septic arthritis was pertinent. With the ultrasonographical investigation, when the multiple collections were found (Table 2), it was more likely to be a case of tropical pyomyositis. Doppler ultrasonography was performed on the lower limbs to rule out superficial or deep venous thrombosis. There was no evidence of abdominal disease.

Pyomyositis is most often found as a localized infection in skeletal muscle, but may also present as diffuse inflammation or a rapidly progressive myonecrotic process. The disease can affect any group or a muscle unit, but the disease commonly affects the large muscles of the body. The muscle most commonly affected is the quadriceps muscle (65%), followed by the gluteal muscles (35%). One possible explanation for the greater involvement of these muscles may be the greater degree of movement, which may cause subclinical trauma, which is a predisposing factor. In 12-60% of the cases, the multiple-segment enveloping occurs 5, 11. Overall results are satisfactory if the disease is diagnosticated and treated in the early stages. Late stages present an increased morbidity and prolonged hospitalization, which may result in continuity extension to the nearby bone or region, causing a compartmental syndrome or distant infection, such as pericarditis, endocarditis, myocarditis, pulmonary and cerebral abscesses, renal failure, septicemia, and even death. Mortality ranged from 0.5% to 2%.²

According to Larkin *et al.* and Drosos, the clinic will depend on the general stage presented by the patient at the time of the investigation.

- The first stage, the so-called "invasive stage" - about 2% of patients at diagnosis. It lasts around ten days with minimal signs of inflammation. The patients present with muscular pain, fever, leukocytosis and elevation of HSV. Other possible symptoms that may arise are anemia and eosinophilia;

- The second is the "suppurative stage" - about 90% of the patients. It is present between 10 and 21 days after the onset of symptoms. The inflammatory signs are more intense, with muscular tension and edema, fever and leukocytosis;

- The third stage is the "final stage" - only 5% of patients. There is high fever, toxicity and occasionally septicemia and even coma.

At the time of diagnosis, the patient was in the second stage of the disease - suppurative stage - and there were signs of inflammation accentuated with tension and muscular edema, leukocytosis (Table 1) and, unlike the typical picture, absence of fever. Blood culture was negative. In individuals with comorbidities, such as immunodeficiency virus (HIV) infection, diabetes mellitus, hematopoietic disorders and other conditions that occur with defective neutrophil function, an atypical presentation is commonly found13. Blood cultures are positive only in 5% of cases, while in non-tropical cases septicemia has been reported more frequently in about one third of patients2.

The diagnosis of pyomyositis is often difficult due to the lack of specific clinical features. They also overlap with symptoms with common endemic febrile diseases, making clinical suspicion often low. Generally, leptospirosis, malaria, dengue, other viral fevers, polymyositis, septic arthritis, osteomyelitis, cellulitis, lymphangitis and deep venous thrombosis should be considered as differential diagnosis14.

Once diagnosed, pyomyositis requires an early prescription of antibiotics and evaluation of surgical drainage. Anti-staphylococcal medication is traditionally the drug of choice; broad-spectrum antibiotic coverage for anaerobic infections, especially in patients without immunological involvement. The duration of treatment lasts until the complete reduction of abscesses, normal leukocytes and absence of fever for at least one week. If the patient is at a late stage with secondary dissemination of infection of the involved muscles, the recommendation is four to six weeks of parenteral antimicrobial therapy. With the advent of drug resistance, the right choice of antibiotics could significantly improve the outcome15. Our patient required repeated surgical drainage along with extended spectrum antibiotics for a prolonged time (12 weeks), insulin, levothyroxine adjustment, and supportive measures to control the infection.

Pyomyositis is not a well know disease, and can be fatal if not diagnosed early. The initial signs and symptoms are very nonspecific, making it often underdiagnosed. A high level of suspicion is important in diabetic patients and/or patients with other types of immunological deficiency; especially in the occurrence of fever and myalgia without significant elevation of muscle enzymes. S. aureus is the most common microorganism, although it may not be visualized in all blood cultures. Immediate antibacterial treatment is essential in management and surgical intervention, which should not be postponed if relevant in the case. The prognosis can be excellent if the disease is promptly identified and treated correctly.

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Table 1: Laboratory tests

|  |  |  |  |
| --- | --- | --- | --- |
| Laboratory tests | Result  | Laboratory tests  | Result |
| Hemoglobin | 11,3g/dL | Amylase | 74U/I |
| Hematocrit | 33,4 % | Lipase | 28,7 U/I |
| Leukocytes | 10800/mm³ | Total cholesterol | 86mg/dL |
| Segmented | 86% | Lactate | 2,9 U/l |
| Bats | 6% | Direct Coombs | Negative |
| Monocytes | 6% | Anti-HIV | Negative |
| Typical / atypical lymphocytes | 7%/0 | Enzyme immunoassay - dengue | Non-reactive |
| Platelets | 46000u/L | PTH | 8,10pg/mL |
| Sodium | 113mE/L | TSH | 66,17 mIU//L |
| Potassium | 3,3mE/L | Free T4 | 0,41 ng/dl |
| CRP  | 41,85mg/dL | Anti-thyroglobulin AC | 2,6 IU/mL |
| ESR | 105mm/h | AC anti-HCV | Non-reactive |
| Urea | 58,9mg/dL | Anti-HBs / Anti-HBc IgM / HBs Ag | Non-reactive |
| Uric acid | 5,2mg/dL | Rheumatoid factor | 8,6 UI/mL |
| Creatinine | 1,14mg/dL | Quick test - American visceral leishmaniasis | Negative |
| Albumin / globulin ratio | 0,2 | FAN - Anti-metaphase and anti-core plate | Reagent (title - 1/1280) |
| Albumin | 1,34g/L | AC anti – SM | <0,1 U/dL |
| Hemoculture (2 samples) | Negativa | C3  | 188 mg/dL |
| Transferrin | 80mg/dl | C4 | 23,9 mg/dL |
| Transferrin saturation index | 20% | CH50 | low (<60 U CAE) |
| Ferritin | 974,76ng/ml | AC anti-cardiolipin IgG / IgM | Non- reactive |
| Iron | 29ug/dl | AC lupus anticoagulant | <1,2 |
| Total iron binding capacity | 145ug/dl | AC anti-parietal cell | Non-reactive |
| Folic acid | 5,05ng/ml | B12 vitamin | 1.342,1 pg/ml |
| Insulin | 14,8uU/mL | Peptide C | 3,4ng/mL |

Source: Patient record.

Legend: CRP - C reactive protein; ERS - erythrocyte sedimentation rate; PTH - Parathormone; TSH - thyroid stimulating hormone; AC-antibody; FAN - anti-nuclear factor; HIV - human immunodeficiency virus.

Table 2: Ultrasound and magnetic resonance imaging

|  |  |  |
| --- | --- | --- |
| Region  | Ultrasonography Nuclear  | Nuclear magnetic resonance |
| Right thigh | Subcutaneous infiltrative edema in the distal third. Presence of collection / abscess in third distal of anterior and medial thigh, measuring respectively 4.2 x 0.8cm and 4.0 x 1.8cm. | Collection involving vast intermediate and lateral 3.8 x 0.8cm in axial axes presenting wide margin and contour with femoral cortical but without signs of periosteal reaction. No joint effusion. |
| Right knee |  |  |
| Right upper limb | Lateral compartment presenting at least two hypoechoic collections, measuring 2.7 x 0.4cm and the other (super - lateral) measuring about 1.3 x 0.8 cm. | Collections with partially defined limits with intramuscular component in the distal aspect of the vast intermediate 4.2 x 1.8 x 0.9 cm with apparent communication with the joint capsule and extension for subcutaneous planes. |
| Left thigh | Venous Doppler: Absence of images compatible with deep or superficial thrombosis in the lower right limb. |  |
| Left hip | Subcutaneous infiltrative edema in the elbow region and posterior face of the forearm. Large abscess in the olecranon fossa. | Right Elbow |
| Left leg |  | Poorly defined collections of inflammatory aspects with hyposignal T1 and hypersignal T2, located in myoadiposal planes, involving the biceps and triceps brachii muscles, the largest one measuring 7.4 x 3.6 x 1 cm with intense contrast enhancement. |
| Left upper limb | Subcutaneous infiltrative edema. |  |

Source: Patient record.