



CASE REPORT

Tropical Pyomyositis: Case Report

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Tropical pyomyositis is an infectious disease that affects skeletal muscle and may appear as a diffuse inflammation or a rapidly progressive myonecrotic process. *Staphylococcus aureus* is the most common microorganism, accounting for 90% of cases. The diagnosis is sometimes late because patients usually 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: A 42-year-old male patient with diabetes mellitus, hypothyroidism, anemia, thrombocytopenia, and hypoalbuminemia developed tropical pyomyositis with multiple muscle abscesses, requiring prolonged antibiotic therapy and surgical drainage.

Conclusion: Pyomyositis is a little known disease and if not diagnosed early can be fatal.

Introduction

The first case of pyomyositis was described by Scriba in the 19th century, being found endemically in tropical regions. Until recently, pyomyositis was considered uncommon in the temperate zone. Currently there is an increase in the number of cases of pyomyositis in temperate climates [1].

In tropical regions, it affects 1% to 4% of hospitalizations. In temperate regions there are few incidence data. In a review of 100 North American cases over 20 years, incidence was not determined but 10% mortality was reported [2,3].

Under normal circumstances, skeletal muscle tissue is intrinsically resistant to bacterial infections. In tropical pyomyositis, microorganisms reach the skeletal

muscles during transient bacteremia - surgical manipulation of infected oral tissues or even routine dental manipulations, contaminated lower urinary tract catheterization - and it find favorable conditions, as immunosuppression conditions, that trigger infection [2,4,5].

Staphylococcus aureus is the most common microorganism, accounting for 90% of cases. In non-tropical regions, the frequency of staphylococcal infection is lower - about 60% [6].

Tropical pyomyositis is also becoming more common in people infected with HIV. Mechanisms include damage caused by the infection itself, zidovudine therapy, infections caused by parasites and mycobacteria, and deficiency in host defenses [7].

The disease has two peaks of incidence: In childhood (2 to 5 years) and in adulthood (35 to 40 years), 80% male. Non-tropical disease presents a peak of 30-50 and 60-70 years, with a 3:1 ratio [2].

Tropical pyomyositis is not a well-known disease, and its initial characteristics are nonspecific and can be easily confused with other more prevalent pathologies, including arboviruses. The delay in recognition results in a greater number of exams, interventions and days of hospitalization, can generate high costs for the institution; besides negatively influencing the prognosis, the imminent risk of sequelae or death.

The objective of the report is to describe the case of a patient that was hospitalized with tropical pyomyosi-

tis at a tertiary hospital in the municipality of Juazeiro do Norte, in the northeast Brazil, and to point out the importance of their early recognition, as well as the rapid institution of treatment.

Method

The information contained in this study was obtained by means of a review medical records and literature revision. The data were only obtained after obtaining the favorable opinion of the Ethics Committee in Research and the signing of the Term of Secrecy of the medical record.

Case Report

Patient male aged 43 years, white, obesity grade 2, married, from northeast of Brazil, a region of tropical climate, with diabetes mellitus, hypertension and hypothyroidism diagnosed for approximately 3 years, using levothyroxine 125 mcg, metformin 500 mg/day, enalapril 20 mg/day and simvastatin 40 mg/day. The patient reported asthenia and diffuse myalgia, beginning 15 days prior to admission, without association with other signs or symptoms, obtaining partial improvement with analgesics. The following 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 responsive to usual analgesics. One day after hospitalization, he reports worsening of pain and presence of knee and elbow edema, both on the right. On the third day, from the beginning of the clinical condition, he could not wander due to pain (Analog Visual Scale: 10).

In to the Physical examination, he presented normal general state, afebrile, normal blood pressure, capillary glycemia 180 mg/dl. Cardiac and respiratory probe without changes.

- Right lower limb: Swollen, painful to the digitopression in the leg and thigh region, with hyperemia, increase of the temperature of the limb and difficulty of movement of the knee and hip;
- Lower left limb: Swollen 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, swollen, temperature increase, digitopression pain and hyperemia in the joint region;
- Upper left limb: Swollen, with increased temperature in the region of the elbow joint, without limitation of movement.

In the emergency room, the first conduct was puncture in the right knee with exit of purulent contents, requiring surgical drainage of the join. The drainage was interrupted after 24 h due to the absence of flow through the drain. Antibiotic therapy was started with ceftria-

xone 2 g 24/24 h and oxacillin 2 g 4/4 h, with hospital admission. The antibiotic therapy was chosen empirically, based on the sensitivity profile of the community bacteria, and considering the diagnostic hypothesis of septic arthritis, giving coverage to gram negative and gram-positive germs.

In view of the extension of the clinical condition, it was realized an investigation through images of the inflammatory foci, and multiple muscle abscesses were visualized. The images were available in the medical record, only the reports of the examinations (Table 1). Early nuclear magnetic resonance was not feasible due to intense pain to the positioning, even with medication, being initially performed only USG and one week after the MRI.

After one week of use of ceftriaxone and oxacillin, no improvement in pain and/or reduction of edema/inflammation was observed, and Meropenem 1 g 8/8 h and vancomycin 1 g 12/12 h were started, due to lack of clinical response. The results of the blood cultures were negative, preventing guided antibiotic therapy. The result of the drained pus culture was positive for methicillin-sensitive *S. aureus*, but there was no good clinical response to the therapy instituted initially, even with favorable *in vitro* sensitivity profile.

Conservative treatment using the new antibiotic regimen was maintained 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 to drainage was necessary.

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

Discussion

Analyzing the case, we observe that, there was no history of recent trauma, which would fit as the entry point of the infection. Transthoracic echocardiography was performed to exclude the formation of emboli due to bacterial endocarditis.

Search for autoimmune disease was started, after finding anti-nucleus factor and reagent chromosomal metaphase plate with a title of 1/1280 - homogeneous nuclear pattern, splenomegaly, thrombocytopenia values 46000 u/L (reference values: 150000 a 450000) prior to the diagnosis. This last investigation was not conclusive until the moment of hospital discharge, being referred for outpatient follow-up. Investigation was made for systemic lupus erythematosus, but the patient did not meet the diagnostic criteria, and remained in clinical follow-up to anticipate the diagnosis in case of later manifestation. In relation to thrombocytopenia, there was an improvement in laboratory levels even during hospi-

Table 1: 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 × 0.8 cm and 4.0 × 1.8 cm.	Collection involving vast intermediate and lateral 3.8 × 0.8 cm in axial axes presenting wide margin and contour with femoral cortical but without signs of periosteal reaction. No joint effusion.
Right knee	Lateral compartment presenting at least two hypo-echoic collections, measuring 2.7 × 0.4 cm and the other (super - lateral) measuring about 1.3 × 0.8 cm. Venous Doppler: Absence of images compatible with deep or superficial thrombosis in the lower right limb.	Collections with partially defined limits with intramuscular component in the distal aspect of the vast intermediate 4.2 × 1.8 × 0.9 cm with apparent communication with the joint capsule and extension for subcutaneous planes.
Right upper limb	Subcutaneous infiltrative edema in the elbow region and posterior face of the forearm. Large abscess in the olecranon fossa.	Right Elbow 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 × 3.6 × 1 cm with intense contrast enhancement.
Left thigh	Subcutaneous infiltrative edema.	Inflammatory/infectious appearance collections in myoadiposal planes exhibiting intense post-contrast parietal enhancement: - Vast lateral and intermediate: Measuring 4.5 × 2.1 cm with intramuscular extension. - Medial and rectus femoris: Two juxtaposed and confluent collections measuring 2.8 × 0.8 cm.
Left hip	Important edematous/inflammatory subcutaneous infiltrate, without significant collection.	Inflammatory/infectious appearance collections in myoadiposal planes exhibiting intense post-contrast parietal enhancement: - Maximum and average gluteus measuring 8.9 × 7.7 × 3.7 cm (13 ml). - Adductor magno 5 × 1.7 × 1.6 cm (7 ml).
Left leg	Subcutaneous infiltrative edema in the posterior face of the leg. Distal third of the medial gastrocnemius presenting collection/abscess measuring around 4.1 × 1.2 cm.	Inflammatory/infectious appearance collections in myoadiposal planes exhibiting intense post-contrast parietal enhancement. - Soleus: Two small collections poorly defined 0.5 × 0.3 cm and 0.6 × 0.5 cm. - Tibial anterior: 1.1 × 0.4 cm.
Left upper limb	Subcutaneous infiltrative edema in the elbow region and anteromedial face of the forearm.	Left elbow Net collection of well-defined margins, located at the transition between the subcutaneous planes and the pronator round muscle, measuring 2.8 × 2.3 × 0.9 cm showing peripheral enhancement after contrast infusion.
Transthoracic echocardiography	Within the normality.	Unrealized.

Source: Patient record.

talization. In addition, the patient had anemia (HB: 11.3 g/dl RV: 13.5-18) and hypoalbuminemia, values: 1.34 g/dL (RV: 4.01-4.78 g/dL) (Table 2).

At the beginning of the clinical condition, the signs and symptoms were similar to arboviruses, especially taking into account local epidemiology. With the worsening of the edema and pain in the knee, and after puncture of suppurative content, the hypothesis of septic arthritis was pertinent. With ultrasonographic investigation, when multiple collections were found (Table 1), 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.

At the time of diagnosis, the patient was in the second stage of the disease - suppurative stage - there were signs of inflammation accentuated with tension added to muscular edema, 10800 leukocytes (RV: 4000 a 10000) (Table 2), unlike the typical clinical presentation, absence of fever. Blood culture was negative. The muscular biopsy was dispensed by the clinical characteristics of the patient, which left no doubt as to the diagnosis.

Our patient required repeated surgical drainage along with broad-spectrum antibiotics for extended time (12 weeks), insulin, levothyroxine adjustment, and supportive measures to control infection.

The literature reports of blunt trauma, in which the-

Table 2: Laboratory tests.

Laboratory tests	Result	Laboratory tests	Result
Hemoglobin*	11.3 g/dL	Amylase	74 U/l
Hematocrit*	33.4%	Lipase	28.7 U/l
Leukocytes*	10800/mm ³	Total cholesterol	86 mg/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*	46000 u/L	PTH*	8.10 pg/mL
Sodium*	113 mE/L	TSH [†]	66.17 mIU//L
Potassium*	3.3 mE/L	Free T4*	0.41 ng/dl
CRP [†]	41.85 mg/dL	Anti-thyroglobulin AC	2.6 IU/mL
ESR [†]	105 mm/h	AC anti-HCV	Non-reactive
Urea [†]	58.9 mg/dL	Anti-HBs/Anti-HBc IgM/HBs Ag	Non-reactive
Uric acid	5.2 mg/dL	Rheumatoid factor	8.6 UI/mL
Creatinine [†]	1.14 mg/dL	Quick test - American visceral leishmaniasis	Negative
Albumin/globulin ratio	0.2	FAN - Anti-metaphase and anti-core plate	Reagent (title - 1/1280)
Albumin*	1.34 g/L	AC anti-SM	< 0.1 U/dL
Hemoculture (2 samples)	Negative	C3	188 mg/dL
Transferrin*	80 mg/dl	C4	23.9 mg/dL
Transferrin saturation index	20%	CH50	low (< 60 U CAE)
Ferritin [†]	974.76 ng/ml	AC anti-cardiolipin IgG/IgM	Non-reactive
Iron*	29 ug/dl	AC lupus anticoagulant	< 1.2
Total iron binding capacity*	145 ug/dl	AC anti-parietal cell	Non-reactive
Folic acid*	5.05 ng/ml	B12 vitamin	1.342.1 pg/ml
Insulin	14.8 uU/mL	Culture pus	Methicillin-sensitive <i>S. aureus</i>

Source: Patient record.

Legend: PCR: C Reactive Protein; VHS: Erythrocyte Sedimentation Rate; PTH: Parathormone; TSH: Thyroid Stimulating Hormone; AC: Antibody; FAN: Anti-Nuclear Factor; HIV: Human Immunodeficiency Virus; *Exams with values below normal; †Exams with values above normal.

re is extravasation of blood into the muscle, or vigorous exercise of the involved muscle groups are reported in 20-50% of the cases of pyomyositis. It has also been suggested that an abnormality of the immune system may be an underlying cause in many cases, especially by the inadequate action of T lymphocytes against *staphylococcus*. Some people may be colonized by *S. aureus*, but because they have an active immune system, they are not affected by the transient presence of this organism in the bloodstream [8].

Among immunity modifying factors, there were diabetes mellitus which may be involved as facilitators of the spread of the infection. There is no formal link between diabetes and pyomyositis, but it is widely reported in the literature the high incidence of pyomyositis in patients with diabetes. An interesting fact to consider is the increase in cases of pyomyositis in this population, which increased from 8% in studies between 1971-1991 to 31% in the most current studies [9].

In diabetics, the predisposition to infections occurs primarily in individuals with longstanding disease and with a lack of glycemic control. Damage to the humoral and cellular defense system, combined with damage to the nerves and blood vessels, can act as facilitators of infectious entities. Malnutrition is another important factor for the development of pyomyositis [9].

Anemia and hypoproteinemia (hypoalbuminemia) were seen in a significant proportion of patients in studies in Nigeria and India [10,11]. The diagnosis is sometimes late because patients usually do not seek care for the first symptoms, and because it is a rare disease, doctors are still not familiar with the entity. The differential diagnosis depends on the region and includes osteomyelitis, deep venous thrombosis, cellulitis, hematoma, tumors, synovitis, septic arthritis and, for iliopsoas pyomyositis, appendicitis, diverticulitis and other causes of peritonitis [12].

Pyomyositis usually presents as a skeletal muscle infection, but it also appears as a diffuse inflammation or a rapidly progressive myonecrotic process. Although any skeletal muscle may be involved, the disease has a predilection for large muscles in the body. The most commonly involved site is the quadriceps muscle (65%), followed by the gluteal muscles (35%). The involvement of several muscles occurs in 12-60% of the pyomyositis [5]. The possible reason for involvement of the pelvic girdle and lower extremity muscles may be the greater degree of movement, which may cause subclinical trauma to the muscles making them susceptible [13]. Overall results are satisfactory if the disease is recognized and treated in the early stages. Late stages present an increased morbidity and prolonged hospitalization, whi-

ch may result in local extension to adjacent bone or junction, cause a compartment syndrome, cause remote 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 and collaborators (1994), Drosos (2005), Shepherd (1983), the clinic will depend on the stage presented by the patient at the time of the investigation, this classification is valid for tropical and non-tropical pyomyositis, whose differentiation is only geographical [3,14-16]:

- First stage, the so-called "invasive stage", only 2% of patients. It lasts about ten days and the signs of inflammation are minimal. Patients present with muscle pain, fever, leukocytosis and elevation of HSV. Other findings may be anemia and eosinophilia.
- The second is the "suppurative stage". Usually this phase is between 10 and 21 days after the onset of symptoms. More than 90% of patients present at this stage. Signs of inflammation are most accentuated with muscle tension and edema, fever and leukocytosis.
- The third stage is the "final stage". The patient is actually sick with high fever, toxicity and occasionally septicemia and coma can be found. About 5% of patients are presented at this stage.

Atypical presentation may occur in patients with co-morbid conditions such as immunodeficiency virus (HIV) infection, diabetes mellitus, hematopoietic disorders, and other conditions that occur with defective neutrophilic function [17]. Blood cultures are positive only in 5% of cases, whereas in non-tropical cases septicemia has been reported more frequently in about one third of patients [2].

The diagnosis of pyomyositis is often difficult to establish 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, deep vein thrombosis should be considered as differential diagnosis [18]. For diagnosis the culture of the abscess or muscle biopsy can be performed [2].

Once diagnosed, pyomyositis requires early institution of antibiotics and evaluation for surgical drainage. Anti-staphylococcal medication is traditionally the medicine of choice; broad-spectrum antibiotic coverage for anaerobic infections, especially in patients without immune compromise. With the emergence of drug resistance, the right choice of antibiotics would significantly improve outcome [19]. The duration of treatment is until the complete reduction of abscesses, normal leukocytes and absence of febrile for at least one week. If

the patient presents in the late phase with secondary dissemination of infection of the involved muscles, the recommendation is four to six weeks of parenteral antimicrobial therapy [20].

Conclusion

Pyomyositis is a poorly understood condition and it may be fatal if not diagnosed early. The initial signs and symptoms are nonspecific, making it often underdiagnosed. A high level of suspicion is important in diabetic patients and/or patients with other types of immune compromise; mainly in the occurrence of fever and myalgia without significant elevation of muscle enzymes. *S. aureus* is cited in the literature as the most common microorganism, although it cannot be seen in all blood cultures. Immediate antibacterial treatment is essential in management and surgical intervention, which when relevant, should not be postponed. The prognosis remains excellent if the disease is promptly identified and treated correctly.

References

1. Chattopadhyay Bitoti, Mukhopadhyay Mainak, Chatterjee Atri, Biswas Pijush Kanti, Chatterjee Nandini, et al. (2013) Tropical pyomyositis. *N Am Journal Med Sci* 5: 600-603.
2. Chauhan S, Jain S, Varma S, Chauhan S (2004) Tropical pyomyositis (myositis tropicans): Current perspective. *Postgrad Med J* 80: 267-270.
3. Christin L, Sarosi GA (1992) Pyomyositis in North America: Case reports and review. *Clin Infect Dis* 15: 668-677.
4. Larkin Julie A, Shashy Ronald G, Poblete Sarah JP (1999) Nontropical Pyomyositis. *Hospital Physician* 67-71.
5. Chawla S, Bansal M, Chawla L (2016) Tropical pyomyositis: A report of two cases. *Med J DY Patil Univ* 9: 657-660.
6. Crum-Cianflon Nancy F (2008) Bacterial, fungal, parasitic, and viral myositis. *Clin Microbiol Rev* 21: 473-494.
7. Batista Rodrigo Siqueira, Gomes Andréia Patrícia, Nacif Marcelo Souto, Guerra Juliana Elvira Herdy, Monte-Alvo Cristiano Ramos, et al. (2004) Manifestações reumáticas da síndrome de imunodeficiência adquirida (AIDS). *Revista Brasileira de Reumatologia* 44: 339-346.
8. Navivan Mitrakrishnan Rayno, Yudhisdran Jevon, Kandeepan Thambyaiah, Kulatunga Aruna (2015) Tropical pyomyositis as a presenting feature of subclinical leukemia: A case report. *Journal of Medical Case Reports* 9: 39.
9. Seah Michele YY, Anavekar Sadanand N, Savige Judy A, Burrell Louise M (2004) Diabetic Pyomyositis: An uncommon cause of a painful leg. *Diabetes Care* 27: 1743-1744.
10. Pozzilli P, Leslie RD (1994) Infections and diabetes: Mechanisms and prospects for prevention. *Diabet Med* 11: 935-941.
11. Cheidozi LC (1979) Pyomyositis: Review of 205 cases in 112 patients. *Am J Surg* 137: 255-259.
12. Malhotra P, Singh S, Sud A, Kumari S (2000) Tropical pyomyositis: Experience of a tertiary care hospital in north-west India. *J Assoc Physicians India* 48: 1057-1059.
13. Golçalves Angelica de Oliveira, Fernandes Nurimar Conceição (2005) Piomiosite tropical. *An Bras Dermatol* 80: 413-414.

14. Sharma Aman, Kumar Susheel, Wanchu Ajay, Sharma Kusum, Sharma Navneet, et al. (2010) Clinical characteristics and predictors of mortality in 67 patients with primary pyomyositis: A study from North India. *Clin Rheumatol* 29: 45-51.
15. Drosos Georgios (2005) Pyomyositis. A literature review. *Acta Orthop Belg* 71: 9-16.
16. Shepherd JJ (1983) Tropical myositis: Is it an entity and what is its causes? *Lancet* 322: 1240-1242.
17. Taguchi Bruno Borges, Francisco José de Arimatea, Campos Pompeu Tome Ribeiro, Teixeira Carlos Osvaldo, Teixeira Maria Aparecida Barone (2013) Piomiosite tropical: Correlação anatomoclínica. Relato de caso. *Rev Bras Clin Med. São Paulo* 11: 194-196.
18. George P, Bendigeri M (2013) Tropical pyomyositis-an emerging multi-disciplinary emergency. *Trop Med Surg* 1: 114.
19. Olson Douglas P, Soares Sarita, Kanade Sandhya V (2011) Community-acquired MRSA pyomyositis: Case report and review of the literature. *J Trop Med* 2011: 970848.
20. Lemonick DM (2012) Non-tropical pyomyositis caused by methicillin-resistant *Staphylococcus aureus*: An unusual cause of bilateral leg pain. *J Emerg Med* 42: e55-e62.