Case Report

Purulent Infectious Myositis

Miosite infecciosa purulenta

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ABSTRACT

Purulent infectious myositis is a rare infection of the skeletal muscle, with Staphylococcus aureus as the commonest primary etiologic agent, which occurs more frequently in men than women. Its mild and nonspecific initial manifestation can lead to a delayed diagnosis, resulting in complications such as endocarditis, osteomyelitis, and sepsis, subsequently affecting the patient's prognosis. Herein, we present the case report of an adolescent female with no significant health history, who developed infectious myositis as a result of an insult secondary to mild trauma to the right upper limb. Medical diagnostic imaging was essential to both confirm the diagnosis and assess the extent of muscle involvement. Prolonged antibiotic therapy effectively treated the infection, and therefore no surgery was needed.

Headings: Pyomyositis; Staphylococcus infection; Antibacterial agents; Case reports

RESUMO

Miosite infecciosa purulenta é uma infecção rara do músculo esquelético, mais frequente em homens, na qual o principal agente etiológico envolvido é o *Staphylococcus aureus*. Por suas manifestações iniciais leves e inespecíficas, o diagnóstico por vezes é retardado, levando a complicações tais como endocardite, osteomielite e sepse, impactando no prognóstico do doente. É relatado aqui um caso de uma adolescente sem antecedentes mórbidos que desenvolveu miosite infecciosa em consequência a um insulto secundário a trauma leve em membro superior direito. A complementação com exames radiológicos foi fundamental para a confirmação do diagnóstico e avaliação da extensão do acometimento muscular. A antibioticoterapia prolongada foi eficaz na resolução do quadro, sem necessidade de abordagem cirúrgica.

Descritores: Piomiosite; Infecções estafilocócicas; Antibacterianos; Relato de Caso.

INTRODUCTION

Purulent infectious myositis (PIM), formerly known as tropical pyomyositis, is an infection characterized by the presence of an intramuscular abscess due to bacterial infection, which is primarily attributed to *Staphylococcus aureus*[1,2]. From an epidemiological standpoint, this disease typically affects men in their first or second decade of life[2,3]. While the pathogenesis of PIM remains unclear, it is thought to be related to transient bacteremia that develops in the context of some preexisting muscle abnormality[3].

Herein, we present a case report of PIM in an adolescent female with no significant health history, whose complementary medical diagnostic imaging evaluation, including magnetic resonance imaging (MRI) and positron emission tomography (PET), was essential to both confirm the diagnosis of PIM and to demonstrate the extent of muscle involvement. Despite the magnitude of involvement, prolonged antibiotic therapy was successful in treating the infection, therefore no surgery was needed.

CASE REPORT

A 14-year-old female, with no known prior comorbidities, presented with pain in her left shoulder after she incurred a mild trauma during a recreational game seven days prior. The patient sought immediate medical assistance, but in the absence of any phlogistic signs upon physical examination performed during her initial evaluation, she was treated with an anti-inflammatory medication to resolve the supposed muscle strain. After three days, the patient returned with fever of 39 °C, as well as worsening arthralgia. Blood counts and shoulder ultrasonography (US) were performed; however, the results showed no changes. Therefore, the patient was discharged again only with symptomatic medication. Her condition progressed beyond the initial arthralgia, however, and the patient began to have limited movement of other joints, such as her knees and hands, as well as pustular lesions erupted in her lower limbs. Twelve days after the onset of symptoms, she was taken to the emergency room at Emílio Ribas Institute of Infectious Diseases, presenting sleepy, feverish, eupneic, normotensive, with pustules on the lower limbs (Figure 1), swelling in the fourth metacarpophalangeal joint of the right hand (Figure 2), swelling and heat in the left knee, no phlogistic signs in the shoulders, and decreased limb strength (Grade 3). The remainder of the physical examination was normal. Considering the tentative diagnosis of reactive arthritis secondary to an infectious condition, antibiotic therapy with ceftriaxone was started. Table 1 shows the laboratory tests performed upon her admission. In the following days, the patient showed increased signs and symptoms (fever, arthralgia, limbs weakness), while new erythematous and painful subcutaneous nodules appeared in the trunk, upper limbs, and lower limbs. An US was performed on the knees bilaterally (which showed no relevant changes) as well as the abdomen (which showed hepatomegaly and nephropathy). The patient also underwent an MRI of the chest wall, which showed multiple small collections located in the belly of the left pectoralis major muscle, adjacent to the left sternoclavicular joint, as well as bone marrow signal alteration in the medial end of the left clavicle, suggesting osteomyelitis (images not available). The patient then underwent a PET scan with fluorodeoxyglucose (FDG) to better analyze the extent of the disease, the results of which showed multiple areas of densification in the muscular and subcutaneous planes with hypoattenuating centers in the thorax and pelvis, and dissemination to all four limbs (Figure 3a). Several blood culture samples collected during the patient's hospitalization tested negative; however, in a single urine culture sample collected on the day of admission, there was growth of methicillin-susceptible *Staphylococcus aureus* (MSSA). The clinical, laboratory, and radiographic findings led to a differential diagnosis of PIM. As a result of this diagnosis, the patient's antimicrobial regimen was changed to oxacillin and clindamycin starting on the 11th day of her admission. In light of the disseminated abscesses, the pediatric surgery team chose not to perform multiple punctures and/or incisions for drainage, reserving this possibility in case of clinical worsening (sepsis), which did not happen. After eight weeks of intravenous antibiotic treatment, as well as progressive clinical, laboratory, and radiographic improvement (Figure 3b), the patient was discharged from hospital with a prescription of cephalexin concerning outpatient treatment of osteomyelitis.

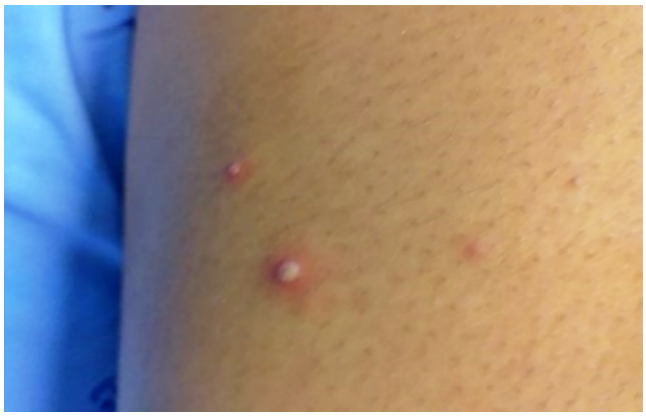

Figure 1. Skin pustules on lower limb

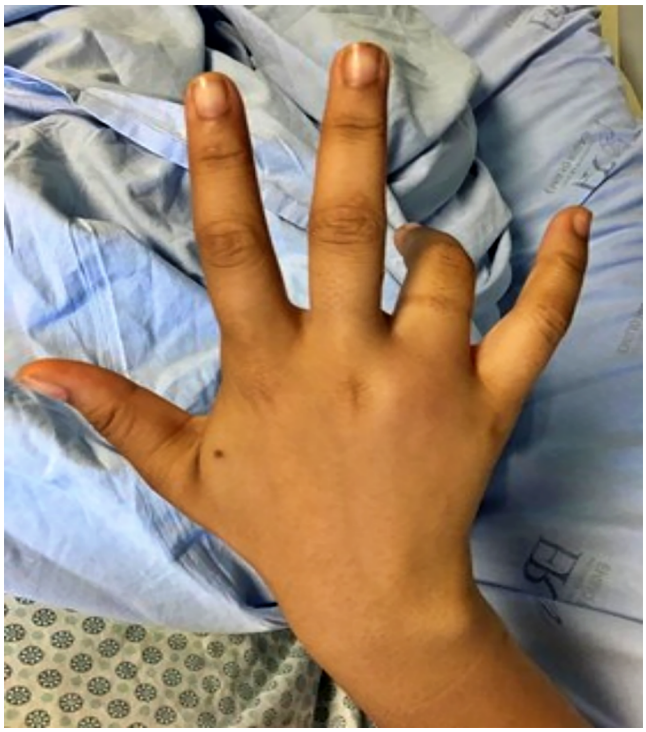

Figure 2. Arthritis in the fourth metacarpophalangeal joint of the right hand

Table 1. Laboratory tests performed upon hospital admission

Blood count	Hemoglobin: 12.3 g/dL; Hematocrit: 34.8%; Total leukocytes: 24,300/mm^3 (metamyelocytes, 2%; rods, 4%; neutrophils, 82%; lymphocytes, 5%; monocytes, 7%); platelets: 77,000/mm^3
C-reactive protein (CRP)	396.60 mg/L (reference value 5–10 mg/dL)
Creatine phosphokinase (CPK)	652 U/L
Transaminases (TGO/TGP)	152 U/L; 202 U/L
Bilirubin	Total, 3.87 mg/dL; Direct, 3.21 mg/dL
Urea/Creatinine	126 mg/dL; 2.21 mg/dL
Sodium/Potassium	133 mmol/L; 4.2 mmol/L
Urine type 1	pH, 6.0; blood, +++; protein, ++; leukocytes, 800,000/mL; erythrocytes, 250,000/mL; negative nitrites, with the presence of bacteria

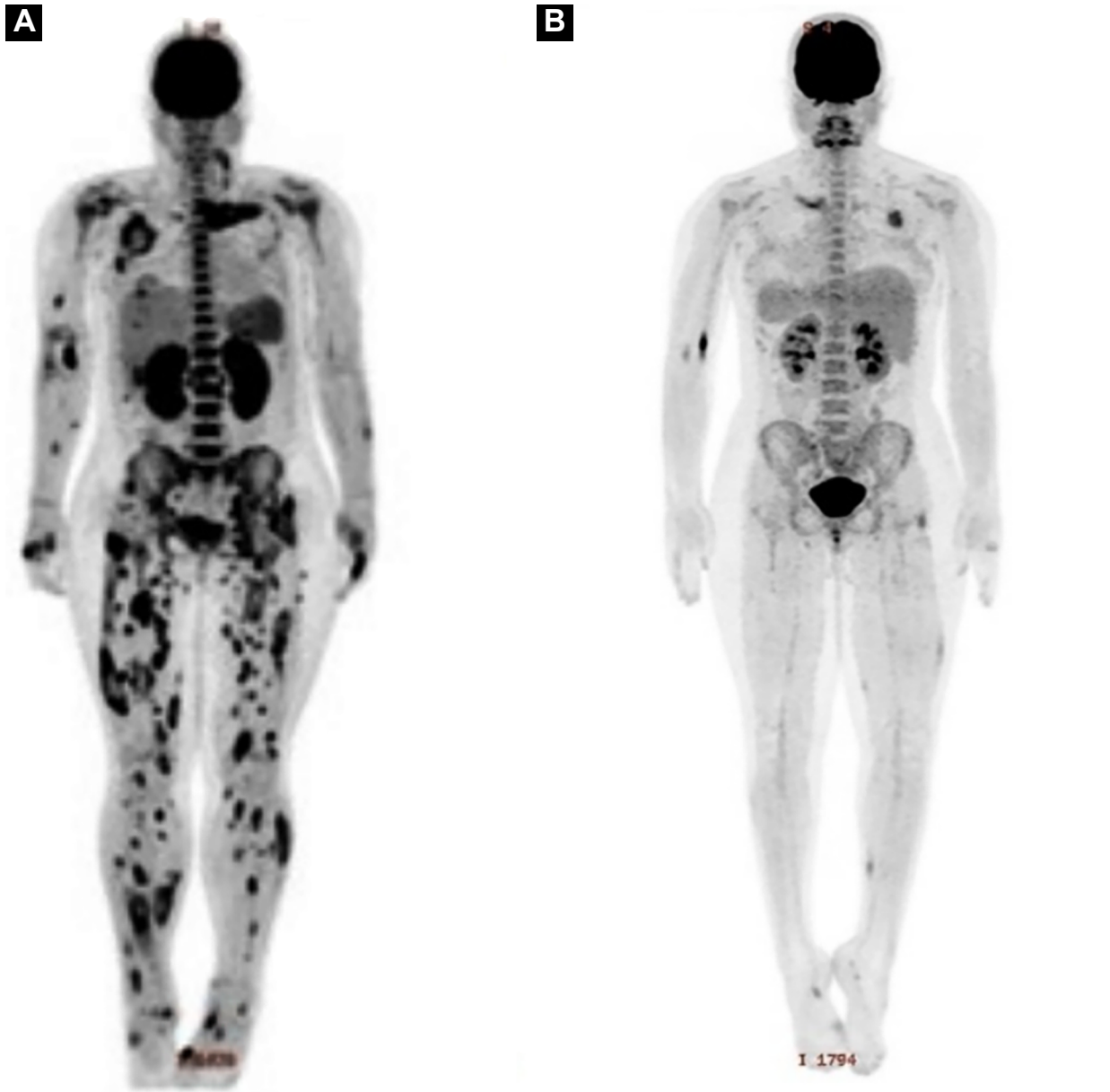

Figure 3. (a) PET scan with multiple densifications in muscle planes; (b) progress of PET scan after eight weeks of intravenous antibiotic therapy

DISCUSSION

Tropical pyomyositis was initially described by Scriba[4] in 1885; however, it wasn't until 1971 that Levin et al.[5] described it in the geographic context of temperate regions. Since its original description, due to the numerous cases documented globally, the regional nomenclature has been replaced by the term "PIM",[6] which refers to a subacute, deep infection of skeletal muscles, infectious disease that accounts for up to 4% of surgical hospital admissions in some countries [7-9]. Early detection and treatment are considered to be the most effective interventions for PIM to minimize complications and reduce mortality, which can reach 23%[8,10]. People with any type of immunodeficiency, diabetes mellitus, cancer, kidney failure, malnutrition, autoimmune disease, or a history of preexisting trauma are more predisposed to develop PIM. It is important, however, to highlight the potential of this disease to affect immunocompetent patients[6–8,10], as in the case described herein.

PIM can be categorized as primary or secondary, based on the route of infection. The former is intrinsically related to bacteremia, while the latter is associated with contiguous focus. The origin of the infection in primary PIM, however, occurs neither by continuity, nor by inoculation or penetrating injury[6,11,12], as seen in the present case. The pathogenesis of PIM is thought to involve transient bacteremia associated with a prior skeletal muscle insult or traumatic injury, even a mild one such as that related to rigorous physical exercise. Other predisposing conditions have also been suggested, such as factors that affect skin integrity (atopic dermatitis, intravenous or intradermal drug use), nutritional deficiencies, and viral myositis. In most cases, however, the specific event which led to the development of PIM remains unclear[1,3,6,12].

When evaluating the disease topography and muscle groups involved, PIM can be categorized as focal or generalized[6,13]. Any muscle group can be affected, and most abscesses are solitary, with only 12-40% of cases being multifocal[14]. The muscle groups most affected are the glutes, quadriceps, and iliopsoas[14,15], although some studies have also reported the involvement of the abdominal wall, cervical, foot, flank, forearm, and calf muscles[2,3,14,16]. In the case of the patient presented herein, she had extensive and varied multifocal involvement, including the hand, which, according to relevant literature, is the rarest segment to be affected, corresponding to less than 1% of the cases reviewed[3].

The primary form of PIM has three distinct progressive stages, which represent the progression from diffuse inflammation to the formation of a localized abscess, which may or may not cause sepsis[3,6]. The first stage, known as the invasive stage, occurs over a period of 10–21 days, with general nonspecific symptoms such as fever, vague muscle aches, and poorly localized edema, sometimes described as "woody induration" - few patients present with this stage. The second stage is the purulent or suppurative stage, and involves chills along with sustained fever, focal muscle pain, and increased edema. During this phase, exudate develops, which progresses to abscess formation. In this stage, a needle aspiration procedure can reveal purulent fluid. Most patients present at this stage. Finally, the third, or late stage, is characterized by obvious local signs, such as erythema and fluctuation, as well as systemic manifestations, including clinical signs of sepsis[1,2,6]. Complication percentages range from 9 to 66%, with sepsis being the most serious one. If not treated, contiguous or hematological dissemination may occur, which may subsequently progress to extra-muscular complications such as meningitis, myelitis, necrotizing fasciitis, septic pulmonary embolism, intra-abdominal abscesses, renal failure, pericarditis, cardiac tamponade, and osteomyelitis, among others[6].

It can be difficult to diagnosis PIM in its early stages due to the lack of overlying skin changes combined with nonspecific findings on physical examination. Therefore, a high index of suspicion is necessary to achieve an early diagnosis, as differential diagnoses include fever of undetermined etiology, orthopedic muscle conditions such as muscle strain, rupture, or hematoma, tumors such as osteosarcoma, osteomyelitis, septic arthritis, and thrombophlebitis. In cases where the abdominal muscles are affected, there may even be acute abdominal mimics[1,3,6,15]. In the laboratory, the acute phase biomarkers are increased, in addition to leukocytosis with neutrophilic predominance in the complete blood count tests; muscle enzymes such as creatine phosphokinase (CPK) and aldolase can be normal or slightly elevated[3,9,13,15,16]. Infectious agent are not frequently identified in blood cultures, occurring in up to 35% of cases[1,11,12,17]. The culture of any purulent material obtained by the puncture or drainage of abscesses, on the other hand, may be negative in 15–30% of samples[1]. In cases where it is possible to identify the microorganism, *Staphylococcus aureus* is the most common[6,15,18], found in up to 90% of the isolates[1,6,17]. Other gram-positive and negative agents, such as aerobes, anaerobes, mycobacteria, and fungi, have also been reported[1,3,11,14].

Medical diagnostic imaging plays a key role in the early and accurate diagnosis and evaluation of PIM. Radiographs have limited function due to their low sensitivity, and are therefore more useful in excluding other processes, such as osteomyelitis or orteosarcoma[3,18].

US is very useful in the detection of superficial collections, in addition to allowing the guided puncture or drainage of abscesses[1,3,6]. Computed tomography (CT) and MRI are more sensitive in terms of demonstrating swelling of the affected muscle(s), corresponding to necrosis or collections. These lesions may reveal a contrast-enhanced peripheral ring, consistent with an abscess capsule which develops during the suppurative stage of the disease. MRI plays an essential role in timely and early diagnosis, particularly in the initial phase of the disease, allowing for the precise localization and improved delineation of the extent of muscle involvement[18]. While gallium or technetium scintigraphy can detect multifocal disease, single-photon emission computed tomography (SPECT) and PET scan can also be used for the same purpose, albeit at a higher cost[6,18].

Abscess drainage should be performed in conjunction with an appropriate antimicrobial therapy for the treatment of PIM. When there is an early diagnosis, that is, one in the first stage, the disease can be treated solely with antibiotic therapy, especially in children[6,7]. The case described by Niamane et al.[13] also showed therapeutic success with the exclusive use of antibiotic therapy as we have reported in the case presented herein, a decision which was determined by the multifocal distribution of PIM and the fact the patient was immunocompetent. The total recommended treatment time for PIM of bacterial etiology is 4-6 weeks, with the first 1-2 weeks of intravenous antibiotic therapy[3,6,15] using an anti-staphylococcal drug resistant to inactivation by penicillinase[1] - in selected cases, anti-methicillin-resistant *Staphylococcus aureus* (MRSA) coverage is required[6].

CONCLUSION

The case presented herein clearly illustrates an incidence of PIM as a potentially serious and widespread disease affecting an immunocompetent adolescent female, having no direct relationship with any infectious point of entry, and with the most common etiologic agent (*S. aureus*) found only in urine culture. Despite the extensive distribution of affected muscle groups and the lack of surgical drainage of multiple abscesses, the patient showed good clinical improvement with prolonged antibiotic therapy.

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