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Original Research Article

A Challenging Case of Tropical Pyomyositis; Revisiting an Old Foe

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Abstract

Tropical pyomyositis is an unfrequent disease characterized by skeletal muscle suppuration mainly occurring in tropical countries, few cases have been observed in India. The patients present with fever, localized swelling & pain. Mainly lower limb muscles are involved, other muscles involved are the chest wall, back, forearm & abdominal wall with a history of trauma or intramuscular injections. Due to non-specific signs & symptoms, early diagnosis is often missed Laboratory investigations reveal low hemoglobin levels, leucocytosis with neutrophil predominance, and increased levels of acute phase reactants. Diagnosis can be confirmed by pus aspiration from involved muscles & culture sensitivity shows Staphylococcus aureus. Ultrasonography, Computed tomography scan, and Magnetic resonance imaging can be done to confirm the findings. The first antibiotic of choice is Cloxacillin. Aggressive management should be instituted with incision and drainage. The clinician should be aware & familiar with the presentations of this disease as it can be life-threatening but with aggressive management it is curable.

Introduction

Scriba, in the 19th century first gave the description of pyomyositis, a disease affecting skeletal muscles. Levin et al in 1971 reported the first case from a temperate region. It is most common in tropical regions like Asia, tropical Africa, Oceania, and the Caribbean islands, but cases have been reported in temperate regions of the world. [1] Based on its geographical endemicity, various terminologies have been used like tropical pyomyositis, myositis tropicans, tropical skeletal muscle abscess, and tropical myositis Around 1% to 4% of hospitalized patients suffer from this disease. [2] A review by Christin L in 1992 on 100 North American cases over 20 years observed 10% mortality. [3] It mainly affects immune-compromised individuals, patients on chemotherapy, undergone organ transplantation or with diabetes

mellitus, chronic kidney disease, asplenia, scleroderma, rheumatoid arthritis, Human immunodeficiency virus infection, and acquired immune deficiency syndrome & Felty's syndrome. [4] In healthy individuals, it is observed in athletics, after vigorous exercise & bicycle accidents in the tropics. Peak incidence is observed in monsoon season (July to September), but cases are noted throughout the year. [5]

Skeletal muscle tissue is intrinsically resistant to bacterial infections but in pyomyositis, transient bacteremia can result in infection of skeletal muscles. Transient bacteremia can occur during common dental treatments or contaminated lower urinary tract catheterization. [6] Also parasitic infections, immunocompromised state. [7] The disease has male predilection & peak incidence is observed at 2 to 5 years & 35 to 40 yrs of age in tropical regions. In non-tropical disease, the peak incidence is noted at 30-50 and 60-70 years, with a male-to-female predilection of a 3:1 ratio. [8] Initial clinical features are nonspecific & recognition & diagnosis of the disease is difficult. Treatment delays may result in increased hospitalization days, financial burden & significant morbidity & mortality. [6] The purpose of this case report is to report the rare clinical condition of pyomyositis & discuss its clinical features, treatment & management & thus guiding the clinician in early diagnosis to reduce mortality.

Case Presentation

A 13-year-old girl presented with persistent abdominal pain for 7 days in addition to bilateral lower limb swelling & extreme tenderness over the right lower limb, severe enough to precipitate a fall in the bathroom 3 days prior to admission. The patient had a history of fever for 5 days within the 15 daysprior to admission at our centre for which was diagnosed as Dengue (based on an NS1 positive report) and received home-based care consisting of intravenous fluids and supportive treatment by a local practitioner for the subsequent 5 days, following recovery.

On clinical examination, the patient had a toxic look, anasarca with warm extremities The pulse was 168 beats/min, respiratory rate was 48 breaths/min, oxygen saturation was 90% on room air, Non-invasive blood pressure(NIBP) was 86/55 mmHg, CRT>3 sec. Sensorium: EVM:15/15, no meningeal signs. ABG: PH:7.28, pCO2 23.5 mmHg pO2 76mmHg HCO3-10.9 mmol/L lactate - 6.87. Chest X-ray and ECG were normal. Initial laboratory examination showed elevated white cell count with neutrophilic predominance with raised inflammatory markers (C reactive protein-199 mg/L, procalcitonin-36.15 ng/mL). 2D Echocardiogram ruled out any underlying cardiac impairment and the initial color doppler of lower limb vessels was normal. The patient was then started on empiric broad-spectrum antibiotic therapy with Meropenem, resuscitated with intravenous fluids & blood components, on top of supplying oxygen by mask along with supportive therapy. However, despite all these efforts, within 48 hours there was no improvement in condition. Furthermore, the patient´s level of consciousness deteriorated on day 3 with unilateral pupil dilatation necessitatingendotracheal intubation for airway protection and aggressive measures to reduce intracranial pressure. CT Head showed diffuse parenchymal swelling with effacement of sulcal effacement. Significant redness of the right lower limb was also noted, requiring a repeating doppler study whichat this point revealed diffuse subcutaneous edema and several collections in the myoadiposal planes without any associated joint effusions. This was completed with a CT abdomen which revealed multiple collections in the myoadiposal planes with sparing of joints. Subsequently needle aspiration of purulent collections with

culture sensitivity of samples and blood culture revealed MRSA sensitive to Vancomycin, linezolid, and clindamycin. Consequently, Vancomycin was added in addition to source control with repeated surgical debridements in the context of refractory septic shock.

During the initial 2 weeks, a tracheostomy was performed due to prolonged ventilatory support, and CRRT was required for stabilization and offloading. She was making good progress with progressively reduced supportive therapy with preserved wound healing. On day 30 was successfully decannulated from tracheostomy and on day 33 patient was finally discharged.

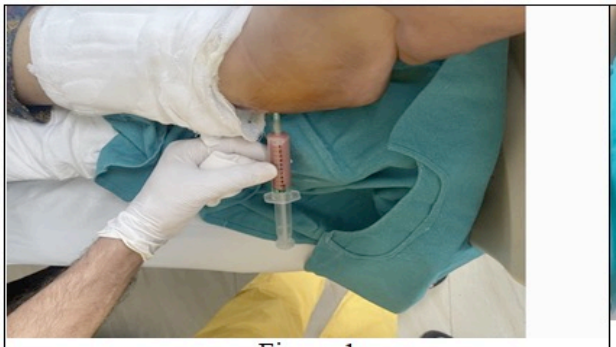
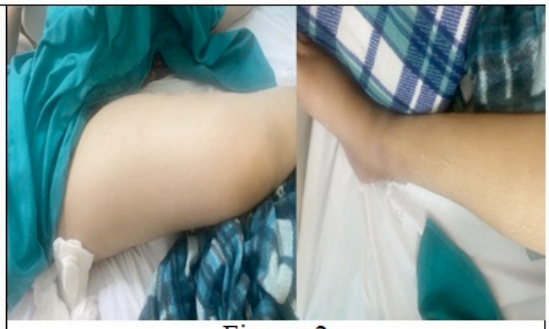


Figure 1



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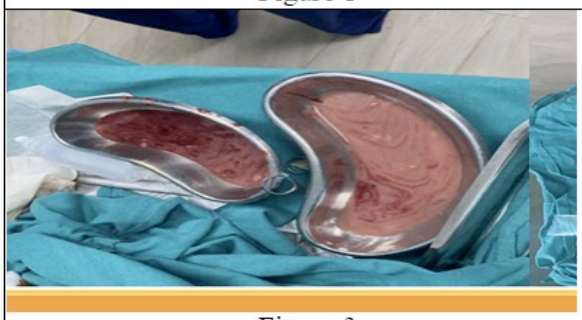

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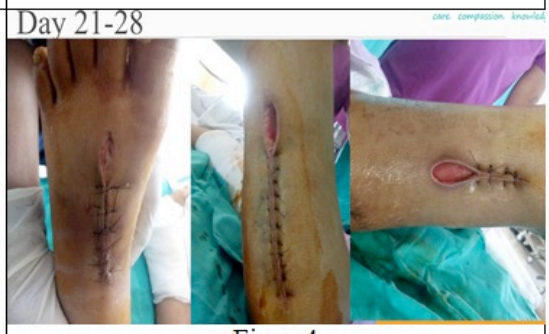

Figur 4

Discussion

Tropical pyomyositis, often seen in tropical countries, is a disease characterized by suppuration within skeletal muscles manifesting as single or multiple abscesses. [1,2] It has not been widely reported in India but there have been sporadic case reports. [9]

The term ‘‘tropical pyomyositis" is restricted to primary muscle abscesses arising within the skeletal muscle. It is not used to describe either (a) intermuscular abscesses, (b) abscesses extending into muscles from adjoining tissues such as bone or subcutaneous tissues or (c) those secondary to the previous septicemia. Staphylococcus aureus is the organism most commonly cultured from abscesses, with up to 90% of cases in tropical areas and 75% of cases in temperate countries. Group A Streptococcus accounts for another 1–5% of cases and other organisms uncommonly implicated are Streptococcus (groups B, C, G), Pneumococcus, Neisseria, Haemophilus, Aeromonas, Serratia, Yersinia, Pseudomonas, Klebsiella, and Escherichia. Rarely Salmonella, Citrobacter, Fusobacterium, anaerobes, and Mycobacterium are seen. [3]

In tropical regions, pus cultures are sterile up to 15–30% of cases, and 90–95% of patients also, have negative blood cultures. Blood cultures are positive in 20–30% of cases in temperate regions. Etiopathogenesis remains unclear as muscles are generally resistant to bacterial infection. Damaged muscle’s susceptibility to hematogenous invasion by bacteria is

increased with resultant abscess formation.10 In 20–50% of cases, a history of blunt trauma or vigorous exercise of the involved group of muscles is forthcoming. Other possible mechanisms include nutritional deficiencies and viral and parasitic infections besides abundant iron is available after trauma, resulting in the profuse growth of bacteria. Underlying abnormality of the immune system may be the predisposing factor in many cases. Some researchers have proved that the lymphocytes, particularly T-cells, in patients with tropical pyomyositis are not primed adequately against staphylococcus during infection. [11] Other predisposing factors include a preceding viral infection (especially arboviruses, as in our case) or nematode infection (Dracunculus medinensis) but their role in pathogenesis remains uncertain. [12] Moreover, as these abscesses are intermuscular rather than intramuscular, these are not included under tropical pyomyositis. Intravenous drug abusers and HIV are other important risk factors for tropical pyomyositis. The disease is seen in all age groups, although young males are themost susceptible range. Maximum incidence is seen at 10–40 years of age with a male preponderance ratio of 1.5:1. Frequently involved muscles are quadriceps, glutei, pectoralis major,serratus anterior, biceps, iliopsoas, gastronemius, abdominal and spinal muscles. Usually, a single group of muscles is affected but in 12–40% of cases, multiple groups are involved either sequentially or simultaneously. [13]

Several stages of progression are noted. This first stage called the "invasive stage" is characterized by an insidious onset of variable fever, painful firm swelling, and negligible systemic symptoms with or without erythema (as the infection is deep-seated). The area is tender with a wooden consistency. Aspiration at this stage may not yield pus. Firm swelling, absence of erythema, and mild pain may divert attention away from an infectious etiology. The invasive stage may resolve itself, confusing with fibromyalgia, or may progress to the next stage of suppuration. The second phase acknowledges for “Suppurative stage". From the second to the third week, abscess forms in the muscle. Temperature spikes with more severe systemic symptoms mark the suppurative stage. Most cases are present at this time. There may be an absence of classical signs of abscess, fluctuation,and erythema, as the overlying muscle is tense. Needle aspiration at this stage yields pus. Regional lymph nodes are not involved.

Finally, the third phase is the “late stage”: If the abscess remains untreated, dissemination of infection occurs. Bacteriemia, followed by septicemia, septic shock, acute renal failure, and metastatic abscesses are complications that are described. [14,15]

Atypical presentations are not uncommon. The patient may present acutely with fever and chills or local symptoms. In rare occasions, the initial presenting picture may be toxic shock syndrome. [16] At times, a prolonged invasive phase might cause the patient to present with pyrexia of unknown origin. Rarely, it may present as an acute abdomen or spinal cord compression or compartment syndrome, which can be easily mistaken for cervicobrachial neuralgia when localized to neck muscles. [17] Tropical pyomyositis is a great masquerader. Its differential diagnosis is wide and includes pyrexia of unknown origin, septic arthritis, muscle contusion osteomyelitis, cellulitis, muscle hematoma, deep vein thrombosis, muscle rupture or muscle strain, osteosarcoma of muscle, trichinosis, leptospirosis, and polymyositis. [8] Occasionally, the invasion by Cysticercus cellulose is characterised by fever, muscle tenderness and eosinophilia, which needs to be differentiated. Rarely, trypanosomiasis or toxoplasmosis causing focal or diffuse myopathy presents as severe myalgia mimicking pyomyositis. It

must also be distinguished from spontaneous gangrenous myositis caused by Streptococcus pyogenes characterized by gangrenous necrosis which prompts emergent debridement. A patient presenting with muscle pain, fever, and/or leucocytosis should include tropical pyomyositis in its list of differentials. [8]

Early diagnosis remains critical. Muscle biopsy also helps to exclude osteosarcoma, polymyositis, trypanosomiasis, toxoplasmosis, cysticercosis, and trichinosis. Ultrasound characteristics include hypoechoic areas with an increase in muscle bulk. Computed tomography (CT)/magnetic resonance imaging (MRI) are the best imaging techniques for early diagnosis. CT shows areas of low attenuation with loss of muscle planes and a surrounding rim of contrast enhancement as characteristic of pyomyositis. CT is also useful in differentiating tumours, haematoma, and thrombophlebitis from the abscess. However, CT alone may be unreliable in distinguishing muscle abscesses from swollen muscles. MRI shows hyperintense rim on T1 weighted images with peripheral enhancement on gadolinium DTPA scan. [18,19,20,21]

Once the diagnosis is established, attention should turn to aggressive management. Surgical debridement and drainage, accompanied by parenteral antistaphylococcal b-lactamase-resistant penicillin (cloxacillin 1–2 g every six hours) is the initial recommended choice.

For methicillin-resistant staphylococcus (MRSA), vancomycin in a dose of 15 mg/kg to a maximum of 1 g given every 12 hours is a suitable alternative. Another glycopeptide, teicoplanin, in a dosage

of 400 mg/day in a single dose is equally effective. For vancomycin-intermediate sensitive staphylococcus, linezolid or dalfopristine-quinapristine are effective alternatives. [22]

If group A streptococcus is isolated from the pus, treatment should be changed to crystalline penicillin, while awaiting culture and sensitivity reports. For Gram-negative bacilli, the addition of an aminoglycoside, that is, gentamicin in doses of 5–6 mg/kg/day intravenously, should be considered in addition to cephalosporins. The possibility of associated anaerobic infection is an indication for introducing metronidazole (20– 30 mg/kg/day) intravenously or orally at intervals of eight hours.

Broad spectrum empirical antibiotics against Gram positive, Gram negative and anaerobic organisms should be administered in immunocompromised patients. In addition to anti- staphylococcal antibiotics, patients should also receive aminoglycosides and clindamycin.

Secondary spread of metastatic infection from involved muscles usually requires long courses of up to four to six weeks of parenteral high-dose antimicrobial therapy. Otherwise, treatment should be continued until the wound is clean, the leucocyte count becomes normal and the patient is afebrile for 7–10 days. While on appropriate antimicrobials, post-surgical drainage continuation or recurrence of fever suggests the presence of other foci, development of drug resistance, or, less commonly, drugfever. [23]

Although there are no recommendations to prevent pyomyositis, nasal carriage may be eliminated in patients with a history of pyomyositis or bacteraemic staphylococcal infection, with topical mupirocin nasal formulation, rifampicin (600mg each day) or cloxacillin (500 mg four times a day) for 10 days are other alternate agents. [24]

The natural history is progressive suppuration with either spontaneous drainage and gradual resolution or eventual bacteraemia, and secondary infection leading to fatal outcomes. Despite advances in diagnosis and treatment, mortality varying from 0.5 to 2% still occurs. [8]

Conclusion

Immediate initiation of appropriate antibiotics and surgical debridement and drainage are required to avoid complications. The prognosis remains excellent if the disease is promptly identified and correctly treated. Awareness among clinicians should be spread about this not so frequent clinical entity. Suspicion should be raised in patients having fever, and myalgia without increased levels of muscle enzymes. Staphylococcus aureus is the most frequently cultured microorganism. The gold standard in the diagnosis is pus aspiration from the intramuscular abscess. Aggressive management with antibiotics & surgical drainage should be instituted for the prevention of severe sequelae & better prognosis.

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