American Journal of Infectious Diseases

Case Reports

Purulent Infectious Myositis: Its Anguish and Ubiquitous!

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Article history
Received: 10-08-2020
Revised: 25-09-2020
Accepted: 05-10-2020

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Abstract: Purulent Infection Myositis (PIM) which was formerly called as tropical myositis is an acute or sub-acute primary infection of striated muscles. It affects all age groups, both sexes and occurs in tropical as well as temperate climates. Most frequently encountered culprits are skin commensals like staphylococci aureus. It is predominantly reported in immunocompromised individuals and with pre-existing muscle abnormalities like strain, trauma, parasite or another inciting event. Gram-negative bacilli myositis in an immunocompetent individual is rare. We report a case of *Escherichia coli* PIM in an immunocompetent patient with no risk factors and muscle abnormality. This helps early consideration of proper antibiotic therapy in an immunocompetent patient with pyomyositis and helps avoid complications associated with pyomyositis.

Keywords: Tropical Myositis, Gram Negative Bacteria, Immunocompetent, *Escherichia coli*

Background

Purulent Infectious Myositis (PIM) is a subacute purulent infection of the skeletal muscles (Crum-Cianflone, 2008). The first reported case of PIM was reported in 1971 (Levin *et al*., 1971). Previously it was referred to as 'Tropical myositis' considering most of the reported cases were from tropical countries (Habeych *et al*., 2020). Now it's considered a ubiquitous disease affecting people in temperate as well as tropical climate. Hence the terminology has been changed to purulent infectious myositis (Shepherd, 1983). It affects all age groups, both sexes and both immunocompromised and immunocompetent individuals (Habeych *et al*., 2020). Extensive PIM is most commonly affects the immunocompromised individual with staphylococcus aureus as the most common etiologic agent identified (Habeych *et al*., 2020). PIM is associated with significant morbidity and mortality due to complications like Meningitis, septic emboli, sepsis, compartment syndrome, septic arthritis, pancarditis and multi-organ dysfunction (Al-Najar *et al*., 2010).

Case Presentation

A 34 years lady with no previous known comorbidities presented to our institute with complaints of pain in all limbs for 7 days duration following development of furuncle over left buttock 12 days back for which she didn't take any medications. Three days into illness she developed high grade fever, pain and swelling of left thigh associated with difficulty in walking. Her menstrual history was normal and her obstetric history is 12 years of married life with 2 children both were full-term normal vaginal delivery. There was no history of any substance abuse. With these complaints' patient visited a nearby hospital where her blood investigations done which showed high Creatinine Phosphokinase (CPK) values of 6000 units/dl. MRI of b/l thigh was done which showed swelling/oedema/inflammation involving the pelvic, gluteal and bilateral thigh muscles appearing hypointense on T1 and hyperintense on T2w images with no obvious collection or mass consistent with inflammatory myositis (Fig. 4). During this period her symptoms worsened with development of severe pain, swelling and redness in all four limbs (proximal>distal) (Fig. 1). She was referred to our centre with a provisional diagnosis of inflammatory myositis. On presentation she was bedridden with severe pain in minimal of activities.

On examination, she was conscious oriented, febrile with a temperature of 39.3 C and other vitals were normal. She had pain in all limbs on both active and passive movements. Erythema was present on medial side of bilateral upper limb and thighs which were warm and tender on light palpation. Power could not be assessed due to pain on all limb movements. All other systemic examination was within normal limits.

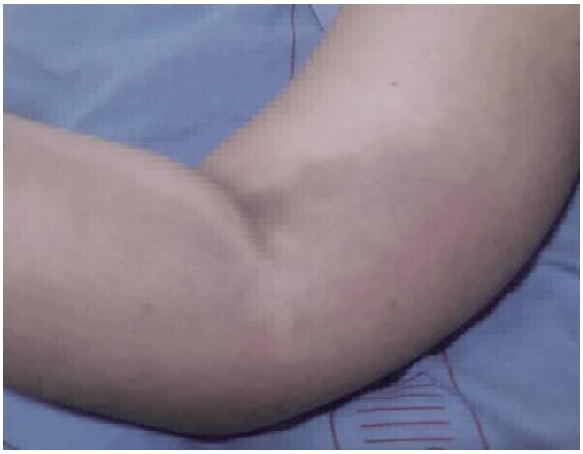

Fig. 1: Erythematous indurated area in the medial side of the right arm and forearm suggestive of pus collection

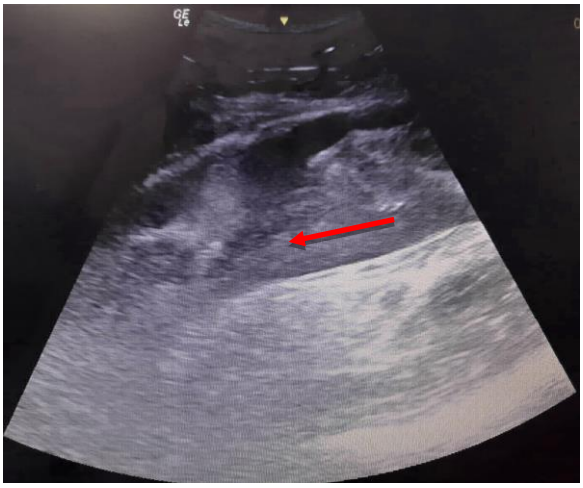

Fig. 2: A large pocket of the collection with moving internal echoes is seen in the intermuscular plane of the left thigh

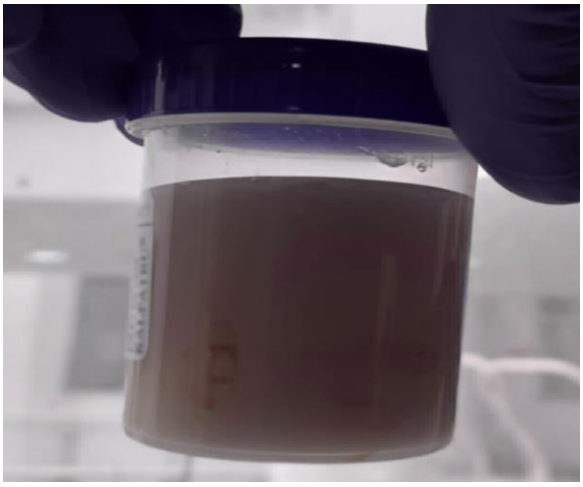

Fig. 3: Pus aspirated form the site

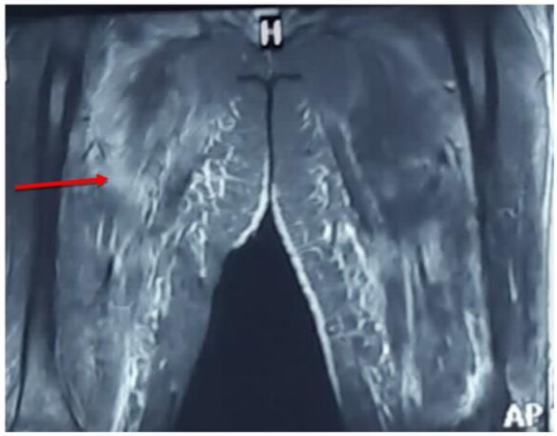

Fig. 4: Magnetic Resonance Imaging (MRI)-Diffuse swelling/ oedema/inflammation seen involving the pelvic, gluteal and bilateral thigh muscles appearing hyperintense on T2w images with no obvious collection or mass

Investigations if Relevant

Her Total leucocyte count (14,450 cells/ml), C-reactive protein (694 mg/dl) and Erythrocyte sedimentation rate (ESR-117 mm/h) were elevated. Procalcitonin was also elevated (32.5 ng/dl) and a provisional diagnosis of tropical pyomyositis was considered. Bedside USG (Fig. 2) showed pus (Fig. 3) collection in intermuscular planes in b/l upper and lower limbs. Aspiration revealed thick, turbid, yellow coloured pus which was sent for culture. Transthoracic echo was done to rule out infective endocarditis which was normal. Her electromyography study was done which showed features of inflammatory myopathy (increased spontaneous activity, insertional activity and Polymorphic small-amplitude Motor unit action potential with increased recruitment). Her immunoglobulin profile (IgG, IgM, IgA and IgE) as sent to rule out immunodeficiency which came to normal. Her autoimmune myositis panel (Ku, Mi-2beta, PM-Sc100, PM-Sc75, JO-1, SRP, PL-7, PL-12, EJ, OJ and RO-52) was negative. Her CPK (4324 IU/mL) was elevated. Her blood culture was sterile while pus culture revealed Escherichia Coli sensitive to amikacin. She was advised for magnetic resonance imaging pelvic region and bilateral lower limb to look for the extent of a collection in lower limbs which showed multiple peripherally enhancing intercommunicating intramuscular intercommunicating intramuscular collection in bilateral thigh involving all three compartments with largest in medial compartment of left thigh (2.2×8.9×10 cm) are also seen in left thigh (Fig. 5). Her fever subsided with treatment but swelling persisted despite repeated drainage. Pigtail insertion in the left thigh was done and rest of the sites were drained under USG guidance. Her Total leucocyte and count, CPK NAC and other inflammatory markers are normalized over next 3 weeks and she was discharged in stable condition.

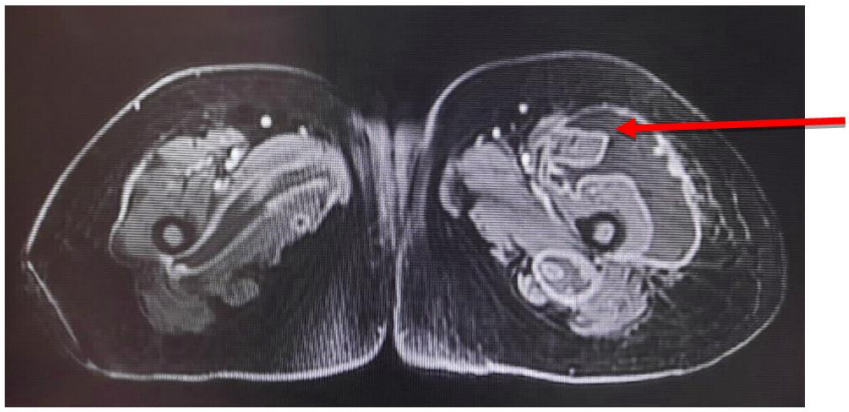

Fig. 5: MRI - the large collection measures- 2.2×8.9×10 cm (AP X TR × CC) in medial compartment of right thigh.

Treatment

Patient was started on a broad-spectrum antibiotic (piperacillin-tazobactam) initially and later antibiotics were changed to Amikacin according to sensitivity. She was continued on same antibiotic for 21 days. She was advised for protein diet and regular physiotherapy. Pigtail insertion in the left thigh was done and rest of the sites were drained under ulatrasound guidance.

Out Come and Follow-up

Her Total leucocyte and count, CPK NAC and other inflammatory markers normalized over next 3 weeks and pigtail removal was done. She was discharged in stable condition.

Discussion

Purulent infection myositis is formation of pus in intermuscular planes most commonly encountered during bacteraemia by gram positive organisms. There are multiple hypotheses proposed for the pathogenesis of PIM. Trauma leading to invasion by pathogenic organism was the most prevalent hypothesis initially as most of PIM cases were young males and muscle affected in these patients were those used to get injured during daily work (Diamandakis and Grose, 1994; Theodorou *et al*., 2007). The next hypothesis of PIM was due to vitamin deficiency (thiamine) (Engel, 1981) and parasites (*Toxocara canis*, *trichinella*, *Dracunculus medinesis*) (Habeych *et al*., 2020) with concurrent bacteremia. Presently PIM is considered to occur as a complication of transient bacteraemia in patients with pre-existing muscle abnormality (strain, trauma, parasite, or another inciting event) (Domínguez-Pinilla *et al*., 2015). Evolutionarily iron sequestration in intracellular spaces act as defence mechanism against bacterial proliferation. Muscle damage releases free iron from myoglobin providing adequate environment for bacterial proliferation and predisposing patients to PIM. Viral infection, hepatitis B carrier state, IV drug abuse, malnutrition, viral infection, Immunodeficiency and malignancy are the common predisposing conditions for pyomyositis (Habeych *et al*., 2020). Our patient did not have any comorbidities and extensive workup did not reveal any immunodeficiency state.

The most common etiologic agent for PIM is staphylococcus aureus followed by streptococci pyogens and less frequently *Escherichia Coli* (Sharma *et al*., 2011). *E. coli* is most commonly associated with urinary tract infection frequently identified in the skin around perineal region (Foxman, 2002). Multiple risk factors are typically common with the extraintestinal manifestation of *E. coli* (Russo and Johnson, 2003). PIM is a rare extraintestinal manifestation of *E. coli* (Cooke *et al*., 2010). *E. coli* PIM is typically most common with immunocompromised individuals and it is rare in immunocompetent individuals (Vigil *et al*., 2010). In our patient furuncle over the buttock region may have led to bacteraemia with seeding of different muscles ultimately leading to pyomyositis.

PIM divided into three clinical stages, stage I: Invasive stage characterized by bacterial invasion of muscle manifesting as fever associated with muscle pain, stage II: Purulent or suppurative stage characterized by purulent collection. Most of the patients present to health care in the second stage of the disease. Stage III is the late-stage characterized by complications of myositis like compartment syndrome, infective endocarditis, sepsis, multi organ dysfunction (Habeych *et al*., 2020). Our patient also presented to us in stage II that is the purulent stage.

In the management of PIM, gram-negative coverage is generally advocated for patients with multiple comorbidities and immunocompromised states (Habeych *et al*., 2020). Our case highlights the need of early gram-negative antibiotic therapy in PIM to avoid complications and reduce morbidity and mortality.

PIM is a great masquerade, a wide range of differential diagnoses and lack of early specific signs (Shepherd, 1983). Extensive pyomyositis must be differentiated from polymyositis (Chauhan *et al*., 2004). In our case, initial differentiation was difficult because the patient is a middle-aged female presented with extensive myositis without any comorbidities. She was diagnosed as PIM based on pus culture report and MRI findings.

Learning Points/Take Home Messages 3-5 Bullet Points

1. Extensive PIM can occur in an immunocompetent individual by gram-negative bacteria.
2. Early aggressive antibiotics therapy should be instituted to avoid complications and gram-negative cover should be considered
3. PIM can mimic as polymyositis in the early stages
4. Its associated with significant morbidity to patient with prolonged hospital stay with severe pain and disability

Acknowledgment

The authors would like to acknowledge all India Institute of Medical Sciences, Rishikesh.

Author's Contributions

Mohan. S.: Manuscript preparation, data collection, study design.

Mahendra Kumar Meena: Manuscript preparation, Manuscript editing.

Ravi Kant: Manuscript review, guidance, corresponding author.

Ethics

This article is original and contains unpublished material. The corresponding author confirms that all of the other authors have read and approved the manuscript and no ethical issues involved.

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