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Methicillin-resistant Staphylococcus aureus Pyomyositis inducing Guillain-Barré Syndrome - A rare and unexpected clinical presentation

Piomiositis por Staphylococcus aureus resistente a la meticilina que induce el síndrome de Guillain-Barré: una presentación clínica rara e inesperada

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Abstract

Tropical pyomyositis is characterized by deep suppurative skeletal muscle infection most commonly by *Staphylococcal Aureus* (S. aureus) with increasing incidence of infection by community acquired methicillin resistant S. aureus(CA-MRSA). The initial clinical presentation is generally non-specific and requires a high index of suspicion. We report the clinical course of a child from subtropical area of North India who developed multiple deep pyogenous collections, complicated with CA-MRSA septicaemia and followed by unusual complications consistent with Guillain-Barré Syndrome.

Keywords: MRSA(Methicillin resistant Staph aureus), Panton-valentine leucocydin(PVL), Vancomycin, Guillain-Barré Syndrome.

Resumen

La piomiositis tropical se caracteriza por una infección supurativa profunda del músculo esquelético, más comúnmente por Staphylococcal Aureus (S. aureus) con una incidencia creciente de infección por S. aureus resistente a la meticilina adquirida en la comunidad (CA-MRSA). La presentación clínica inicial es generalmente inespecífica y requiere un alto índice de sospecha.Presentamos el caso clínico de un niño del área subtropical del norte de la India que desarrolló múltiples colecciones piógenas profundas, complicadas con septicemia por CA-MRSA y seguidas de complicaciones inusuales compatibles con el síndrome de Guillain-Barré.

Palabras clave: MRSA (Staph aureus resistente a meticilina), leucocidina de Panton-valentine (PVL), vancomicina, síndrome de Guillain-Barré.

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Introduction

Pyomyositis is a purulent infection of skeletal muscles. Since this was mainly found in tropical countries, it was considered endemic and also called “tropical Pyomyositis'. However, presently this condition is increasingly being reported from temperate regions also. Though it is found more commonly in adults, it can affect both children (commonest age range of 4-5 years) and adolescents. *Staphylococcus aureus* (S. aureus) infection is the most common (90%) cause for morbidity. This is increasingly being replaced by community-acquired methicillin-resistant S. aureus (CA-MRSA). [1] The source of infection is commonly from a skin lesion spreading by a hematogenous route to an injured skeletal muscle. The trauma to the muscle leads to sequestration of elemental iron from myoglobin, which is inherently protective against bacterial invasion of the muscle. [2] The most common muscle groups involved are those around the pelvis and lower limbs. The most common complications being arthritis, osteomyelitis, pneumonia and even sepsis and meningitis in rare cases. In this case report, we present the clinical course of a female child hailing from a subtropical area of north India who developed multiple deep pyogenous collections, complicated with staphylococcal septicaemia, followed by an uncommon presentation consistent with Guillain-Barré Syndrome (GBS).

Case Presentation

A 9 years old female child presented with complaints of fever of 02 days, generalized bodyache and diffuse pain in bilateral lower limbs. Initial examination revealed axillary temperature of 101F with generalized tenderness in bilateral thighs. She was vaccinated as per age and had appropriate weight for age without any peripheral signs of chronic disease, malignancy, malnutrition or immunocompromised status. Initial investigations revealed Hemoglobin(Hb)=9.8g/dl, Total Leukocyte count (TLC) = 5000/cumm, platelets= 78000/cumm. Dengue, Malarial parasite and Typhoid were negative. Sonogram (USG) study of abdomen was normal. Child was admitted as case of Viral fever and started on symptomatic treatment (injection Paracetamol at 15 mg/kg/dose, maintenance iv fluids). The child however, developed a high grade fever with chills and rigor (maximum 104F) alongwith pain in both lower limbs and inability to walk approx. 48 hrs after admission. Physical examination revealed poorly localised tenderness involving both calves and thighs. USG of bilateral thighs and legs failed to show any infective or thrombotic focus. Repeat blood investigations revealed leukocytosis (TLC=17600/cumm) and raised CRP levels (50 mg/L). Patient was started on a broad spectrum antibiotic (Inj ceftriaxone 1gm twice a day). However, child deteriorated further and at 72 hrs post admission, developed respiratory distress with

tachypnoea (54/min) and chest retractions with low oxygen saturation of 89% at room air. Chest auscultation revealed reduced air entry on right side of chest in the mid-lower zones. Other systemic examination were normal. Chest radiograph revealed right middle and lower zone consolidations. The computed tomograpy (CT) chest revealed septic emboli and pneumatoceles in bilateral lungs with predominantly subpleural and peripheral distribution. Patient was further investigated with lab tests viz. Weil felix, blood culture and for COVID-19. The antibiotics were revised with addition of Inj Tiecoplanin (300 mg iv once a day), Tablet Azithromycin (250 mg per oral once a day) and Tablet Hydroxychloroquin (HCQ)(150 mg twice a day). In the mean time, patient required continuous positive airway pressure (CPAP) for subsequent 03 days and remained febrile (max. 103.6F) with raised CRP, ESR upto 59, Leukocytosis =15400/cumm with neutrophillia and toxic granules on peripheral blood smear (PBS). Serum biochemistry and liver enzymes were within normal limits except serum lactate dehydrogenase (634 IU). Weil Felix serology and RT-PCR for COVID-19 was negative. Blood culture was positive for *Staph Aureus* (Methicillin resistant MRSA type) which was sensitive to Teicoplanin, and hence same antibiotics were continued. Hb levels dropped to 6.0 gm% with stool positive for occult blood. Packed red blood cell (PRBC) transfusion (250 ml) was administered. Respiratory distress settled over the next 03 days but high grade fever with daily spikes, positive CRP and raised ESR persisted till day 09 of admission. On day 10 of admission, a diffuse swelling in right leg mimicking cellulitis appeared and physical examination revealed tenderness in bilateral thighs. USG bilateral thighs revealed multiple pus pockets. Magnetic resonance (MRI) T2W1 and short T1 inversion recovery (STIR) images of B/L lower limbs confirmed multifocal pyogenous collections in the inter and intramuscular regions of anterior and medial compartments of both thighs. In view of patient manifesting with spikes of high grade fever despite antibiotic therapy, a decision for surgical drainage was taken and approx. 8ml and 5ml of pus was drained from right and left thighs respectively. However, patient still remained febrile till day 13 and was found to have another pus pocket in left calf which also needed surgical drainage. Antibiotics were now changed to Inj Vancomycin (400 mg iv thrice a day). Patient still continued to have fever till day 15 and then complained of weakness in both lower limbs. Physical examination revealed flaccid weakness (power 2/5 in knee and hip extensors, 2/5 in ankles) with hyporeflexia. MRI dorsal spine was done to investigate lower limb weakness. Incidentally hydro-syringomelia was detected involving D6 toD8. In view of acute flacid paralysis with hyporeflexia, a suspicion of post infection Guillain-Barré Syndrome was made and a diagnostic Lumbar puncture was performed. CSF study revealed albuminocytological dissociation (raised protein=54 mg/dl without any rise in cell counts). Nerve conduction velocity study could not be performed due to nonavailability. Intravenous Immunoglobulins IVIG 2gm/kg was administered over the next 4 days. The weakness made gradual recovery over the next 06 days. On day 21, patient had only minimal residual weakness in left lower limb with power improved to 4/5 left side (ankle, knee) and full recovery on the right side. She was discharged on day 22 on oral antibiotics (Tab Linezolid 150 mg twice a day) for another 08 days with total duration of antibiotic therapy summing to 04 weeks. Patient remained asymptomatic during follow-up and Neurosurgical consultation for syringomyelia dorsal spine was taken.

Figure 1. CT Chest showing (arrow mark pointing) multiple air filled cavities and soft tissue densities in B/L lung fields. Also seen are few peripheral wedge shaped air space opacities suggestive of septic emboli B/L Lung fields

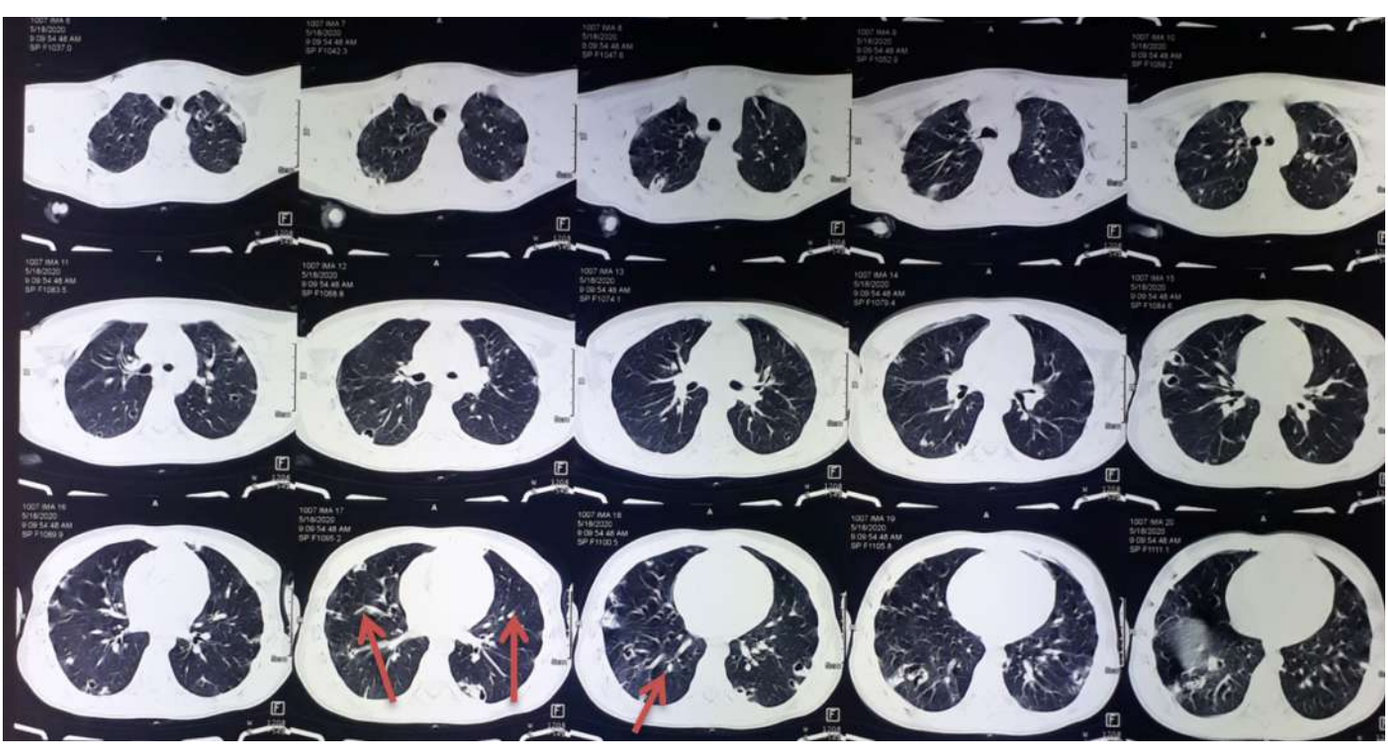


Figure 2 MRI B/L thighs i)Top Left axial STIR image ii) Rest all coronal STIR images showing multiple Hyperintense pyogenous collections (arrow mark pointing)

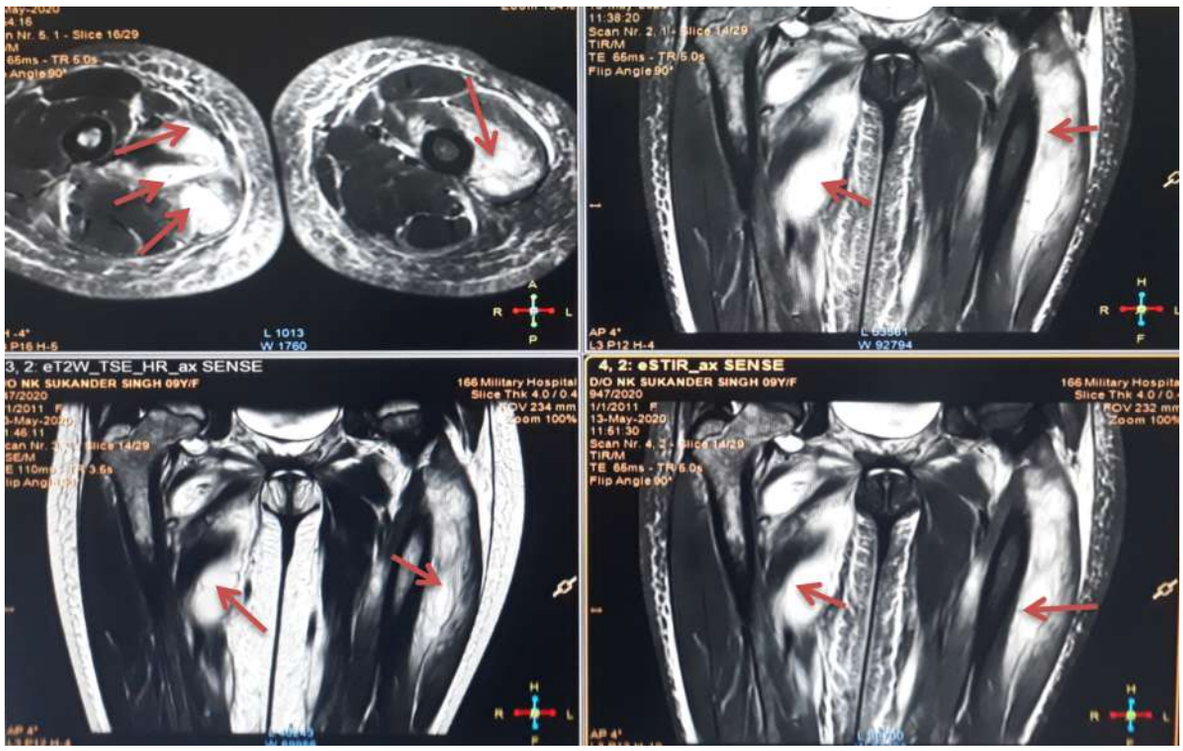
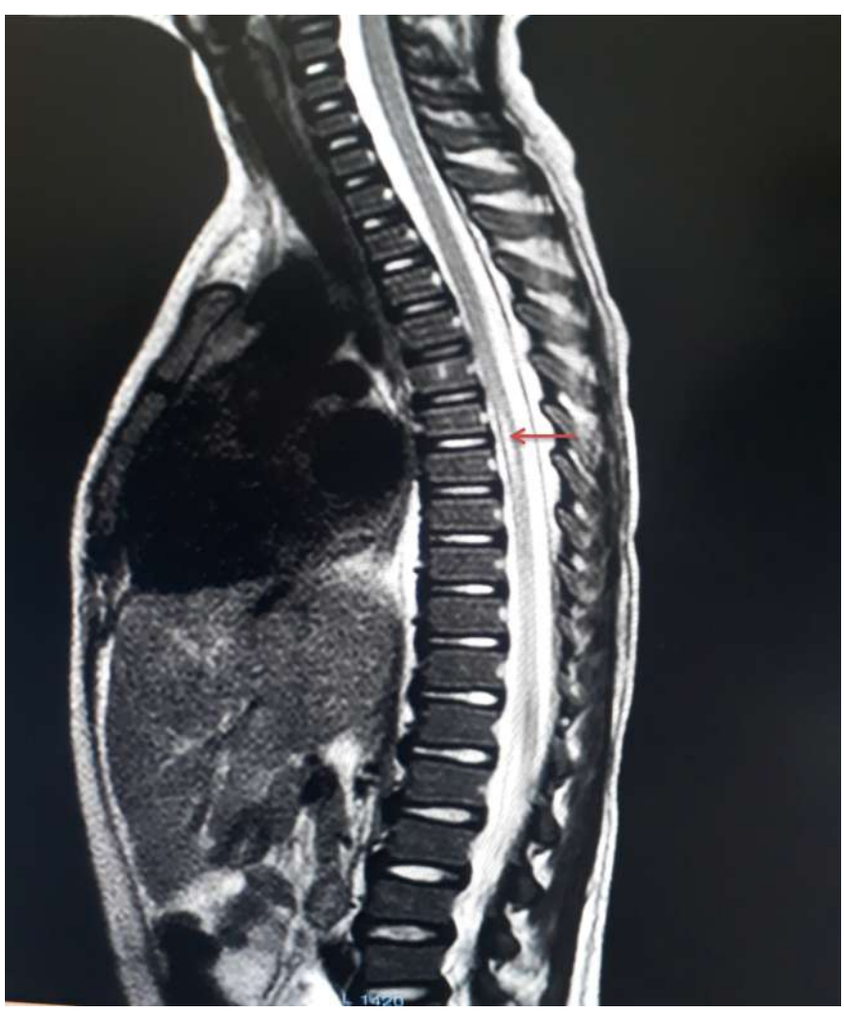


Figure 3 MRI Spine saggital T2W view showing hydrosyringomyelia D6-D8 spine (arrow mark pointing)



Discussion

Tropical pyomyositis is classically encountered in tropical countries as a pyogenic infection of skeletal muscles with a male to female preponderance of 2:1. [3,4]

The clinical and laboratory evaluation of our patient revealed that she had presented to the hospital in stage 1 of the disease, which was characterised by fever, body ache and diffuse pain in bilateral lower limbs. Pyomyositis was not kept as a differential because patient was a female child, hailing from a temperate region in the northern part of India, without any history of trauma, immunocompromised status or localising signs. The patient quickly progressed to stage 2 of the disease characterised by high grade fever, local muscle tenderness with leukocytosis and raised C-reactive protien. Patient was started on antibiotics. The physical examination and USG did not reveal any localised pus collection. Patient then progressed to stage 3 of the disease characterised by signs of sepsis and systemic involvement (bilateral lobar pneumonia with respiratory distress). The management was intensified with evaluation for COVID-19, blood culture and sensitivity, addition of specific antibiotics, oxygen supplementation and other supportive measures. MRI revealed multifocal pyogenous collections in both thighs which is contrary to the generally unifocal collections in immunocompetent children.[5]

The most common etilogical agent in pyomyositis is Staphylococcal aureus, however, community acquired MRSA is being increasingly isolated around the world including India as with this patient. [6] Staphylococcal aureus has multiple virulence factors which enables it to colonize, evade immune response, cause tissue injury and disseminate. [7] There is a wide spectrum of S. aureus diseases ranging from local skin suppuration; systemic dissemination such as sepsis, pneumonia, pyomyositis, osteomyelitis, endocarditis; to exotoxin induced toxic shock syndrome & scalded skin syndrome. Staph pneumonia is a necrotising pneumonitis which can be localized or diffused and spread by hematogenous septic emboli.[8] The disease progression in our patient was particularly severe with rapid deterioration in clinical condition despite on specific and sensitive antibiotic therapy. This is consistent with Community-acquired MRSA which was isolated from the patient. Recently, Panton-valentine leucocydin(PVL), a virulence protein secreted by S. aureus has garnered much attention due to its ability to attach to the phospholipids in the leucocyte cell membrane leading to increased permeability and cell death. PVL producing MRSA strains are now increasingly being reported globally and are associated with more severe and invasive disease mainly in young immunocompetent individuals which could be the case in our patient.[9,10] However, we could not test for the PVL because of lack of facilities.

Another peculiar feature of our case is the possible association of S. aureus sepsis with Guillain-Barré Syndrome (GBS). Nerve conduction velocity (NCV) study was not available at our centre, but the clinical profile of hyporeflexia with acute flaccid weakness, albumino-cytological dissociation in CSF study and good response to IVIG favoured the diagnosis of GBS.

GBS is an autoimmune postinfectious polyneuropathy which usually follows GI or respiratory illnesses, or rarely vaccination. The proposed mechanism is autoimmune neural injury to myelin gangliosides following molecular mimickery.[11] There have been case reports of GBS following staphylococcal endocarditis [12] and pyomyositis [13] in adult population. The most common complications following paediatric pyomyositis are arthritis, venous thrombosis, osteomyelitis and pneumonia. Sepsis, pericarditis and meningitis are rare. However, despite extensive research, we could not come across syndrome of transient weakness involving lower exterimities with hyporeflexia and CSF study suggestive of GBS following staph sepsis in paediatric population. By this case report, we propose that GBS may follow staphylococcal sepsis even after the septic focus is eliminated. Hence, it is imperative that an uncommon complication like GBS must be kept in mind while managing a patient of staph pyomyositis.

Conclusion

Pyomyositis is a relatively rare pyogenic infection of skeletal muscles in children. It presents with vague non-specific symptoms often leading to delay in diagnosis. Our patient was a young immunocompetent child without any risk factors hailing from a temperate zone in the northern part of India. Patient had CA-MRSA pyomyositis which presented with fever and bodyache without any localizing signs and developed respiratory distress requiring CPAP within 48 hrs. She subsequently developed sepsis despite intensive antibiotic therapy and surgical drainage of the septic focii. Furthermore, patient developed features suggestive of GBS which is an extremely rare complication as per existing literature. Thus, a very high index of suspicion with swift management of infection using targeted antibiotic therapy and close monitoring for unexpected complications is highly recommended.

Conflict of Interest

None.

Contribution.

All authors contributed in all phases of writing and approved the final version.

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