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Primary pyomyositis of the forearm in a non-immunocompromised boy

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Abstract
We present a previously healthy 5-year-old boy with primary pyomyositis of the right forearm due to methicillin-susceptible *Staphylococcus aureus*. He had no apparent portal of entry. He was cured after surgical drainage of the affected forearm along with a three-week course of sequential intravenous and oral antibiotics. Pyomyositis is a rare but serious bacterial infection of striated muscle that should be part of the differential diagnosis of any child with fever, localized extremity pain, and edema. It can advance to abscess formation in the affected muscle and have serious short- and long-term consequences. The vast majority of cases in both tropical and temperate regions are caused by *S. aureus*. Hence, empirical anti-staphylococcal therapy pending culture results is appropriate. MRI is the best imaging modality for diagnosis, while common laboratory tests like hemogram, ESR and CRP are useful for monitoring response to therapy. Early diagnosis, surgical drainage and lengthy treatment with appropriate antibiotics lead to clinical cure in most cases.

Keywords: Children, pyomyositis, *Staphylococcus aureus*, panton valentine leukocidin

Introduction
Pyomyositis (PM) is a serious bacterial infection of striated muscle that usually presents with low-grade fever, localized pain, and edema that can advance to abscess formation in the affected muscle. If inadequately treated, sepsis can occur [1]. Primary PM of the forearm is extremely rare, especially in immunocompetent individuals representing just 1.3% of PM cases in the literature [2].
We present a previously healthy 5-year-old boy with primary PM of the right forearm due to methicillin-susceptible *Staphylococcus aureus* (MSSA). Although he reported a minor injury of his forearm prior to presentation, he had no apparent portal of entry. He was cured after surgical drainage of the affected forearm along with a three-week course of sequential intravenous and oral antibiotics.

Case Report
A 5-year-old previously healthy boy presented to the emergency room of our hospital due to a low-grade fever up to 38°C, along with edema and pain of the right forearm over the last 5 days. His parents reported a minor injury of his right forearm during wrestling with his older brother one day prior to him complaining of pain. Physical examination on admission demonstrated edema, mild erythema and heat of the affected region that had painful and severely restricted range of movement (Fig 1).
Laboratory evaluation on admission revealed leukocytes 21,680/μl (74% neutrophils, 12% lymphocytes, 14% monocytes) with normal hemoglobin, hematocrit and platelets, serum C-reactive protein (CRP) 15.53mg/dl (reference values 0-0.8) and erythrocyte sedimentation rate (ESR) 110mm/hour. Plain X-rays of the forearm showed no fracture but confirmed the presence of localized edema. After a blood culture was obtained, the patient was started on intravenous ceftriaxone and clindamycin. An urgent ultrasonogram of the area showed localized collection of pus (Fig 2a), while an emergency Magnetic Resonance Imaging (MRI) scan confirmed our clinical suspicion (Fig 2b). Painful extremity edema is associated with fracture, muscle strain and/or hematoma. Fracture was ruled out by plain radiographs. Muscle strain and hematoma are not typically associated with fever or a three-digit ESR. Bursitis, cellulitis, and thrombophlebitis do not severely restrict the range of movement of

the affected extremity, while juvenile and septic arthritis are associated with joint inflammation, which was not the case in our patient, since the affected area was in proximity but not over a joint. Soft-tissue and bone sarcomas like Ewing's usually have a long history of symptoms until presentation for medical evaluation, which was not the case in our patient, who developed symptoms and signs over a 5-day period. Hence, osteomyelitis and PM were the two most likely diagnoses considered, although the presence of substantial soft tissue edema made us consider PM as the most likely diagnosis from the start.

The patient was taken to the operating room for exploration and drainage of the affected forearm. An extended volar incision of the forearm was performed, beginning from the elbow crease proximally and extending to the volar side of the wrist distally. Proximally, the interval between the brachioradialis and pronator teres was reached. After a diligent blunt dissection, a large amount of pus was drained (Fig 3a). Cultures of the pus were collected in order to identify the causative microorganism. Debridement, drainage, and copious irrigation with normal saline were performed, along with exploratory drilling of the proximal radius that did not detect pus inside the bone. Wound closure was managed with the dermal apposition technique, using skin clips and a vessel loop for gradual wound closure (Fig 3b). Culture of the pus revealed MSSA, while a molecular genetic identification method (GenoType MRSA-Hain Lifescience, Nehren, Germany) showed that the isolated strain did not harbor the resistance mediating genes *mecA* and *mecC* and did not produce the Panton Valentine Leukocidin (PVL); hence postoperative treatment was modified to intravenous cloxacillin at 100mg/kg/day divided in four daily doses. The blood culture that was obtained prior to surgery was negative. The patient received intravenous cloxacillin for two weeks.

During his hospitalization, he remained afebrile, and the fingers had a full range of movement. On the fifth post-operative day, serum CRP dropped to 1.66mg/dl. A full blood count on the eight postoperative day showed leukocytes 9,370/μl with neutrophils 42%, lymphocytes 45%, and monocytes 10%, while CRP dropped further to 0.22mg/dl. Two weeks after surgery, the patient was sent home with oral cefprozil for one more week, since no oral antistaphylococcal penicillin is available in Greece. The patient was compliant with the oral therapy. Six months after the described events, he is doing well (Fig 3c).

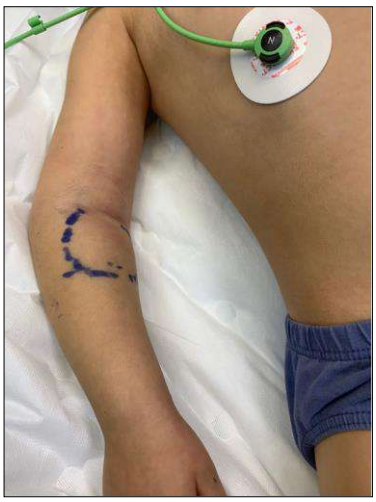

Fig 1: The patient's forearm at presentation. Note the edema and erythema close to the elbow that are demarcated with blue ink

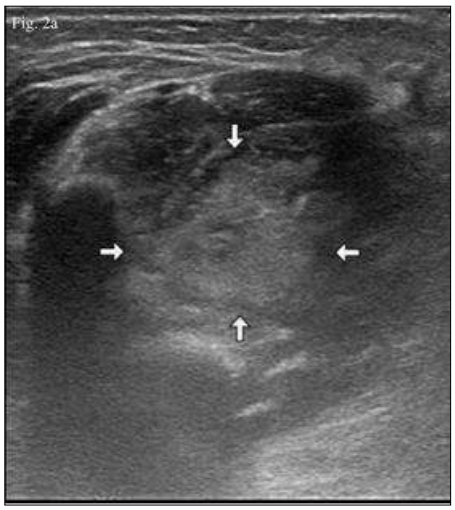

Fig 2a: Transverse ultrasound scan of the anterior aspect of the right proximal forearm shows an intramuscular hyperechoic fluid collection (arrows)

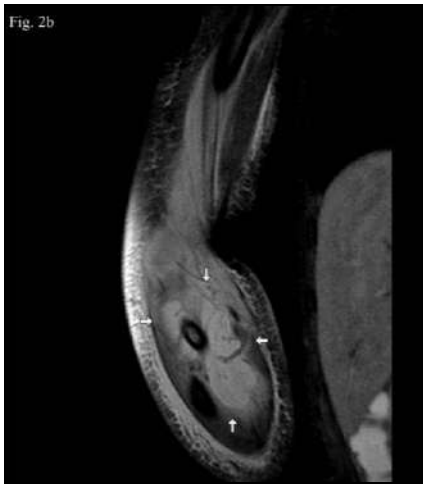

Fig 2b: Coronal fat-suppressed proton-density weighted MR image shows pyomyositis of the anterior compartment of the right proximal forearm, with an intramuscular abscess (arrows)

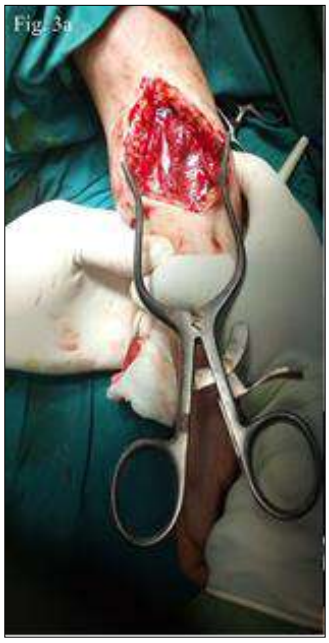

Fig 3a: The patient's forearm during surgery. Note the abundant pus in the operating field

Fig 3b: Photograph of the forearm showing the dermal apposition technique, using skin clips and a vessel loop for gradual wound closure

Fig 3c: Photograph of the patient's forearm approximately six months after surgery. Note the well-healed surgical scar

Discussion

Primary PM is a subacute, suppurative, deep bacterial infection of skeletal muscles, with no apparent spread from contiguous structures [2]. It was first described by Julius Scriba in Japan and has been observed mainly in tropical climates. However, over the last decades, PM is increasingly recognized in temperate climates as well [3, 4]. It is responsible for 1 per 3,000 pediatric admissions in non-tropical climates [5]. The most frequently affected muscles are in the thigh and gluteal region. Consequently, the incidence of PM in the lower extremities is approximately four times higher than in the upper extremities [6-8].

Primary PM of the forearm, like in our case, is extremely rare, especially in immunocompetent patients [6]. Only 1.3% of PM cases occurred in the forearm in a review of 676 patients performed by Bickels *et al.* [2]. Furthermore, in an older review of 112 patients, the forearm was not affected in any of the described cases [9].

Three clinical stages of PM have been described. The invasive stage, in which muscle pain, edema and low-grade fever occur. The suppurative stage characterized by higher fever and severe muscle tenderness. Eventually, if the patient remains untreated, the last stage follows, in which the patient becomes septic [9].

The pathophysiology of PM is thought to involve two events. First, muscle injury occurs, either acute or due to overuse followed by bacteremia [10]. Skeletal muscles are considered resistant to bacteria. However, trauma alters the muscle structure, so that bacteria from a subsequent, unrelated bacteremic episode implant to the traumatized tissue [2]. Animal studies have shown that intravenous injection of sublethal doses of *S. aureus* failed to cause PM, unless the muscles were injured by electric shock, ischemia or pinching [11]. Despite that, most patients with PM including our child do not have an obvious penetrating injury or a clear portal of entry [10].

The vast majority of PM cases in both tropical and temperate regions are caused by *S. aureus*, with *Streptococcus pyogenes* a distant second pathogen [12]. An analysis of 31 cases by Ameh showed that *S. aureus* was cultured in 75% of the patients [13], while in a study of 44 cases, 58% of the clinical cultures isolated *S. aureus*, of which 16 were community-acquired MRSA [14]. *Streptococcus pneumoniae* and Gram-negative organisms such as *Escherichia coli*, *Klebsiella* spp. and *Pseudomonas* spp. are additional pathogens [10]. *Mycobacterium tuberculosis*, *Mycobacterium avium* complex, *Neisseria gonorrhoeae*, *Bacteroides fragilis*, *Fusobacterium* spp., and fungi have rarely been identified as causative pathogens in a few cases of PM, mostly in immunocompromised patients, mainly those with hematologic malignancies [15, 16].

The distribution of MSSA and methicillin-resistant *S. aureus* (MRSA) cases varies according to the individual patient susceptibility and geographic location [17]. The increasing number of PM cases is likely linked to the rise in the prevalence of community-acquired MRSA [14]. The PVL toxin produced predominantly by MRSA clinical isolates, but occasionally by MSSA ones too is well-known to increase the invasive potential of *S. aureus*, predisposing to abscess formation, and deep-seated infections [18, 19]. PVL-producing clinical strains of *S. aureus* can cause fatal infections in children that appear acutely, deteriorate quickly, and have multiple short- and long-term consequences [18]. In the study by Pannaraj *et al.*, 81% of community-acquired MRSA isolates and 44% of community-acquired MSSA isolates caried the PVL genes [14]. The isolated clinical strain in our patient did not produce PVL.

Since the clinical and laboratory investigations of PM resemble many other clinical conditions, proper imaging studies are important for diagnosis. MRI is the preferred imaging modality due to its inherently high spatial and contrast resolution that provides exquisite anatomic information regarding the extent and degree of participation of the involved soft tissues and the underlying bone [19-21]. It clearly demonstrates abscess formation, especially after intravenous gadolinium administration [2]. For urgent bedside imaging, ultrasonography is particularly useful. Typical findings on ultrasonography are a bulky muscle with abnormal echotexture and hypoechoic focal lesions along with internal debris and air bubbles. Intramuscular

hyperechoic fluid collections due to abscess formation can also be seen [2].

Laboratory findings of PM are non-specific. Leukocytosis with a left shift is usual, like in any bacterial infection [2]. Blood inflammatory markers are helpful for diagnosis and especially during follow-up after definite surgery. CRP and ESR are both elevated. CRP is an indicator of disease's severity and is used to monitor response to surgical and medical treatment. Despite the localized muscle destruction, serum creatine phosphokinase is usually normal. Finally, blood cultures are usually negative, especially early in the course of disease.

Pus cultures obtained during surgical drainage reveal the pathogenic organism in most cases [15]. Even though diagnostic delays are frequent in PM, early diagnosis and treatment are crucial to avoid morbidity and mortality from sepsis. Empirical antibiotics alone without open or percutaneous drainage can cure some cases of stage one disease [7, 16, 22]. In these cases, it is prudent to administer a broad-spectrum antibiotic that covers both *S. Aureus* and Gram-negative organisms, especially in immunocompromised individuals [16]. When the pathogen is identified by culture, antibiotic treatment should be adjusted accordingly, like in our case. The optimal duration of treatment depends on the patient's immune status, the number of abscesses, the surgical intervention, and importantly, the clinical course [16]. The duration of antibiotic therapy should likely be no less than two to three weeks, while the timing of switch from intravenous to oral antibiotic treatment is based on clinical and laboratory improvement [23].

Untreated or inadequately treated PM can cause serious complications. Muscle necrosis, direct extension into an adjacent joint or bone or hematogenous spread causing secondary osteomyelitis and/or septic arthritis. Compartment syndrome is a rare, but feared complication [1]. The acute phase response of the bacterial infection is a major cause of systemic complications such as coagulopathy, venous thromboembolism, systemic inflammatory response syndrome, multiorgan failure and even death. Long-term complications of PM include irreversible muscle-scarring with residual muscle weakness and functional impairment. Also, delayed or inappropriate treatment may lead to avascular necrosis, pathologic fractures, growth arrest and limb amputation.

In conclusion, although PM is a relatively rare infection, it should be part of the differential diagnosis of any child with fever, localized extremity pain, and edema. MRI is the imaging modality of choice for diagnosis, while common laboratory tests like hemogram, ESR and CRP are useful for monitoring response to therapy. Early diagnosis, surgical drainage and lengthy treatment with appropriate antibiotics lead to clinical cure in most cases.

Conflicts of interest

The authors have no conflicts of interest regarding this report.

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References

1. Shedek BK, Nilles EJ. Community-associated methicillin-resistant Staphylococcus aureus pyomyositis complicated by compartment syndrome in an immunocompetent young woman. Am J Emerg Med 2008;26(6):737.e3-4.
2. Bickels J, Ben-Sira L, Kessler A, Wientroub S. Primary pyomyositis. J Bone Joint Surg Am 2002;84(12):2277-86.
3. Lovejoy JF, Alexander K, Dinan D, Drehner D, Khan-Assad N, Lacerda IRA. Team Approach: Pyomyositis. JBJS Rev 2017;5(6):e4.
4. Mouhtadi ME, Fakiri KE, Rada N, Draiss G, Mouaffak Y, Younous S *et al*. Toxic shock syndrome and pyomyositis: about an unusual case. Pan Afr Med J 2020;36:63.
5. Gubbay AJ, Isaacs D. Pyomyositis in children. Pediatr Infect Dis J 2000;19(10):1009-12; quiz 1013.
6. McRae M, Sharma S. Forearm pyomyositis in a breast cancer patient on chemotherapy. J Plast Reconstr Aesthetic Surg JPRAS 2010;63(10):e737-739.
7. Olson DP, Soares S, Kanade SV. Community-Acquired MRSA Pyomyositis: Case Report and Review of the Literature. J Trop Med 2011;2011:1-4.
8. Howman-Giles R, McCauley D, Brown J. Multifocal pyomyositis. Diagnosis on technetium-99m MDP bone scan. Clin Nucl Med 1984;9(3):149-51.
9. Chiedozi LC. Pyomyositis. Review of 205 cases in 112 patients. Am J Surg 1979;137(2):255-9.
10. Kamal T, Hall M, Moharam A, Sharr M, Walczak J. Gluteal pyomyositis in a non-tropical region as a rare cause of sciatic nerve compression: a case report. J Med Case Reports 2008;2(1):204.
11. Wong LFA, Mullers S, McGuinness E, Meaney J, O'Connell MP, Fitzpatrick C. Piriformis pyomyositis, an unusual presentation of leg pain post partum – case report and review of literature. J Matern Fetal Neonatal Med 2012;25(8):1505-7.
12. Zadroga RJ, Zylla D, Cawcutt K, Musher DM, Gupta P, Kuskowski M *et al*. Pneumococcal pyomyositis: report of 2 cases and review of the literature. Clin Infect Dis Off Publ Infect Dis Soc Am 2012;55(3):e12-7.
13. Ameh EA. Pyomyositis in children: analysis of 31 cases. Ann Trop Paediatr 1999;19(3):263-5.
14. Pannaraj PS, Hulten KG, Gonzalez BE, Mason Jr. EO, Kaplan SL. Infective Pyomyositis and Myositis in Children in the Era of Community- Acquired, Methicillin- Resistant *Staphylococcus aureus* Infection. Clin Infect Dis 2006;43(8):953-60.
15. Mitsionis GI, Manoudis GN, Lykissas MG, Sionti I, Motsis E, Georgoulis AD *et al*. Pyomyositis in children: early diagnosis and treatment. J Pediatr Surg 2009;44(11):2173-8.
16. AL-Sayyed B, Bayomy M, Rammal B, Mannaa M. Visual diagnosis: 11-year-old boy with leukemia, fever, elbow, and leg pain. Pediatr Rev 2015;36(1):e1-4.
17. Kern L, Rassbach C, Ottolini M. Streptococcal pyomyositis of the psoas: case reports and review. Pediatr Emerg Care 2006;22(4):250-3.
18. Sheikh HQ, Aqil A, Kirby A, Hossain FS. Panton-Valentine leukocidin osteomyelitis in children: a growing threat. Br J Hosp Med Lond Engl 2015;76(1):18-24.

19. Benvenuti M, An T, Amaro E, Lovejoy S, Mencio G, Martus J *et al*. Double-Edged Sword: Musculoskeletal Infection Provoked Acute Phase Response in Children. Orthop Clin North Am 2017;48(2):181-97.
20. Elzohairy MM. Primary pyomyositis in children. Orthop Traumatol Surg Res OTSR 2018;104(3):397-403.
21. Andrew JG, Czyz WM. Pyomyositis presenting as septic arthritis. A report of 2 cases. Acta Orthop Scand 1988;59(5):587-8.
22. Miller NJK, Duncan RDD, Huntley JS. The conservative management of primary pyomyositis abscess in children: case series and review of the literature. Scott Med J 2011;56(3):i-181.
23. McMullan BJ, Andresen D, Blyth CC, Avent ML, Bowen AC, Britton PN *et al*. Antibiotic duration and timing of the switch from intravenous to oral route for bacterial infections in children: systematic review and guidelines. Lancet Infect Dis 2016;16(8):e139-52.