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Original article

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**Pyomyositis: case series of 14 patients to study the significance of early diagnosis and management**

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Abstract

Introduction Pyomyositis denotes primary pyogenic infection of skeletal muscle. It is predominantly a disease of tropical countries. It usually involves the largest muscle groups around the pelvic girdle and lower extremities. Primary reasons for delay in diagnosis are its low incidence and vague presentation [7]. This delay can result in complications such as extension into and destruction of an adjacent joint, sepsis and, even death. Our study is aimed to highlight the extent and sequence of treatment protocol required for good management of these patients. **Methods** We retrospectively analyzed our experience with a series of 14 pediatric patients with primary pyomyositis who were treated and followed up. There were five girls and nine boys. All 14 patients underwent plain radiographs, USG and MRI of the affected area followed by surgical drainage and a course of antibiotics. Patients were followed up with weekly CRP. **Results** Six out of 14 (42.9 %) patients had a history of mild trauma. Iliopsoas muscle was involved in 4 patients, 3 cases in which the gluteals or quadriceps were involved, 2 cases with obturator muscle involvement and 2 cases in which adductors were the infection site. All 14 patients were treated surgically. **Conclusion** Our study shows that early diagnosis, complete drainage of the purulent material and the use of appropriate antibiotic therapy are the key determinants of successful treatment that lead to complete resolution in the majority of cases.

Keywords: CRP, ESR, MSSA, MRSA, Pyomyositis

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New Knowledge Provided By Study: Our study highlights the importance of early diagnosis and management of pyomyositis. Our study also highlights the importance of taking complete history and examination of patients apart from all the radiological investigations which are usually done.

Implications For Clinical Practice or Policy: Our study indicates the importance of early diagnosis and management which would prevent the development of complications like septic hip, septic knee, muscle scarring, injury to growth plate resulting in limb length discrepancies. Also, our study highlights the importance of management sequence in these cases and not administering reckless antibiotic usage which would prevent the spread of antibiotic resistance.

Научная статья

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Пиомиозит: серия случаев из 14 пациентов для изучения значения ранней диагностики и лечения

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Аннотация

Введение. Термин «пиомиозит» обозначает первичную гнойную инфекцию скелетных мышц. Это преимущественно болезнь тропических стран. Обычно он поражает самые крупные группы мышц тазового пояса и нижних конечностей. Основными причинами проблем с диагностикой являются низкая частота и неясная клиническая картина. Несвоевременное выявление патологии может привести к таким осложнениям как распространение инфекции, разрушение соседних суставов, сепсис и даже смерть. Наше исследование направлено на акцентирование внимания специалистов на данную нозологию и представление последовательного протокола, необходимого для достижения положительных исходов при лечении пациентов. **Материалы и методы.** Ретроспективно проанализирован опыт лечения и наблюдения 14 педиатрических пациентов с первичным пиомиозитом. Было пять девочек и девять мальчиков. Всем 14 пациентам были выполнены обзорные рентгенограммы, УЗИ и МРТ пораженного участка с последующим хирургическим дренированием и курсом антибиотикотерапии. Пациенты наблюдались еженедельно с определением уровня СРБ в сыворотке крови. **Результаты.** Шесть из 14 (42,9 %) пациентов имели в анамнезе легкую травму. У 4-х пациентов была поражена подвздошно-поясничная мышца, в 3-х случаях были вовлечены ягодичные и четырехглавые мышцы, в 2-х случаях – запиральная мышца и в 2-х – очагом инфекции были приводящие мышцы. Все 14 пациентов были прооперированы. **Заключение.** Наше исследование показывает, что ранняя диагностика, полное дренирование гнойного содержимого и применение адекватной антибактериальной терапии являются ключевыми факторами успешного лечения, обеспечивающего полное излечение в подавляющем большинстве случаев.

Ключевые слова: СРБ, СОЭ, MSSA, MRSA, пиомиозит

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INTRODUCTION

Primary pyomyositis also called pyogenic myositis is a rare, subacute, deep bacterial infection of skeletal muscles not associated with infection of adjacent skin,

bone or soft tissues [1, 2]. It is predominantly a disease of tropical countries, and hence is also referred to as tropical pyomyositis or myositis tropicans [2, 3].

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Etiology of pyomyositis is frequently classified as primary or secondary. Primary pyomyositis (PPM) is believed to be the result of hematogenous spread of an occult source [4]. Other predisposing factors that might be causative factors include trauma, malnutrition, viral and parasitic infections, bacteremia, immunodeficiency or chronic illness. Moreover, the diagnosis could be delayed if the affected muscle is deep and local signs are not apparent [5].

It usually involves the largest muscle groups around the pelvic girdle and lower extremities [1]. In tropical countries like India, the infection accounts for around 4 % of surgical hospital admissions [6]. Primary reasons for delay in diagnosis are its low incidence and vague presentation [7]. This delay can result in complications such as extension

into and destruction of an adjacent joint, sepsis and, even death [8, 9]. Although it can affect individuals of all ages, primary pyomyositis is most common in the first and second decades of life [1]. Primary pyomyositis has been only sporadically described in the literature. For these patients, an accurate prompt diagnosis and early treatment are critical. Drainage of the muscle abscess, followed by administration of appropriate antibiotics, remains the mainstay of treatment and usually leads to complete recovery [5]. We retrospectively analyzed our experience with a series of 14 pediatric patients with primary pyomyositis who were treated and followed up in our department. Our study was aimed to highlight the extent and sequence of treatment protocol required for good management of these patients.

METHODS

Our case series included 14 children with primary pyomyositis. Out of these 5 were girls and 9 boys with mean age of 2 years and 1 month in the age range of 4 months to 3 years. None of these patients had an underlying disease that might affect the immune system. In all, there were 4 patients in whom the iliopsoas muscle was involved, 3 cases of both the gluteal and quadriceps involvement, 2 cases of obturator muscle involvement and two cases with the adductors as the infection site. Since our department is a government facility, all test were done free of cost for the patient.

All patients belonged to lower socio-economic status. All patients presented with history of high grade fever, pain and swelling around the hip joint or thigh depending on the muscle group involved and inability to bear weight on the affected side due to pain. The duration of symptoms ranged between 5 and 10 days before children's hospitalization. Six out of 14 (42.9 %) children had a history of mild trauma before the onset of the disease, usually related to fall at home while playing. Eight out of 14 (57.1 %) patients had history of treatment at other facilities before coming to us. Out of these 8, three had a history of antibiotic intake before arriving at our hospital; the other 5 had taken only analgesics. Six out of 14 (42.9 %) patients had a history of massage. Complete blood counts, quantitative CRP (C-reactive protein), ESR (erythrocyte sedimentation rate) were sent immediately. All patients underwent plain radiographs of the affected area and none showed any bony involvement on admission following which USG was done to diagnose the condition and the area of collection. USG guided needle aspiration was done as a diagnostic tool in every patient, however collection could be aspirated only in nine out of 14 patients. In other 5 patients, nothing came on aspiration. Samples were sent for gram stain and culture

sensitivity and patients started broad-spectrum antibiotics. Gram-stain reports came within 2 hours on the basis of which we selected the antibiotic to be given. The report showed gram positive bacteria in 11 out of 14 patients on basis of which i/v amoxicillin and clavulanic acid combination was prescribed. Patients were then sent for MRI. MRI was carried out in all the patients as it clearly and most accurately demonstrated the diffuse muscle inflammation. All patients had marked elevation of acute phase reactants. ESR was between 40 and 120 mm/h and CRP between 84 and 251 mg/l at the time of admission. The TLC count ranged between 20000 and 34000/l. Following MRI, the patient was taken immediately to the operating theatre. Time difference between admission and surgery was < 1 day. All 14 patients were operated under general anaesthesia. Anterior approach was taken in 8 patients with collection around hip and hip arthrotomy was also done in them to look for hip joint involvement. The posterolateral approach to the thigh was chosen in 3 patients with quadriceps involvement. The posterior approach to the hip was used in patients with gluteal involvement. Intraoperative cultures were taken. Following surgical drainage, wounds were left open with daily dressing and packing of wound. Patients with surgical wounds around hip were taken up for secondary closure around 2 weeks after the first surgery while those around the thigh were allowed to heal by secondary intention. Following surgery patients continued injections of amoxicillin/clavulanic acid. Culture reports came 48 hours later following which culture specific antibiotics were started. Recovery was assessed based on weekly CRP reports. We have used CARE reporting guidelines for our case series. This paper adheres to these guidelines and is properly formatted.

RESULTS

The study included 14 patients, out of which 9 (64.3 %) were males and 5 (35.7 %) were females (Table 1). On examination, patients with infections around the hip had high grade fever, flexion contracture of the hip joint (positive Thomas test), and a significant local rise of temperature around the affected area with palpable external iliac lymph nodes. In patients with infection around the thigh, there was local redness, tenderness, local rise of temperature and palpable inguinal lymph nodes.

Six out of 14 (42.9 %) patients had a history of mild trauma before the onset of the disease, usually due to fall at home while playing (Table 2). However there was no serious injury.

Table 1

Distribution of patients according to gender

	Male	Female
Number of Patients	9	5

Table 2

Distribution of patients according to history of trauma

History of Trauma	No of Patients
Yes	6
No	8

MRI evaluated the muscle groups involved and it was found that there were 4 patients in whom the iliopsoas muscle was involved, 3 cases each in which the gluteals and quadriceps were involved, 2 cases with obturator muscle involvement and 2 cases in which adductors were the infection site (Table 3).

Table 3

Distribution of patients according to muscle groups

Muscle Groups	No of Patients
Iliopsoas	4
Quadriceps	3
Gluteus	3
Obturator muscles	2
Adductors	2

Of those operated, collections were successfully drained in eleven out of 14 (78.6 %) patients and among them 4 had collection in the hip joint as well which was most probably a secondary complication of a primary pyomyositis of the iliopsoas, adductors or obturator. Three out of 14 (21.4 %) patients did not have any collection. However, intraoperative pus samples were taken in all patients and sent for gram stain and culture sensitivity. In cultures, 7 patients had MSSA sensitive to amoxicillin clavulanic acid, 4 patients had MRSA sensitive to vancomycin and clindamycin while culture report of 3 patients came out to be sterile (Table 4).

Table 4

Distribution of patients according to organism found in culture report

Culture	No of Patients
MSSA	7
MRSA	4
Sterile	3

Patients with MSSA in culture report started i/v amoxicillin clavulanic acid combination. One patient with MRSA was on clindamycin and other 3 patients with MRSA started cloxacillin administration on the basis of the report. In the 3 patients where there was no collection, culture came out to be sterile, so in them broad spectrum antibiotics, i.e. amoxicillin/clavulanic acid combination was continued.

Nine out of 14 (64.3 %) patients operated responded well and did not have fever or had only mild fever

1–2 days post surgery. All other patients did show gradual and consistent clinical improvement with antibiotics if not immediate response. In all 14 patients, fever completely resolved within 1–2 weeks. All patients started weight-bearing walking from day 2 of surgery and one week later were told to do weight bearing as per pain tolerance. Mild ROM exercises were started from day 1 after the surgery and gradually ROM was increased as per pain tolerance. Daily dressings were done as all wounds were left open post surgery. Patients with surgical wounds around the hip were taken up for secondary closure around 2 weeks after the first surgery while those around the thigh were allowed to heal by secondary intention. CRP tests were sent on weekly intervals to monitor the recovery. Patients showed gradual but consistent clinical improvement as was assessed with declining CRP levels until the discharge from hospital without any functional impairment with gradual resolution of the limp, local tenderness and swelling, as well as a return to full range of motion at the adjacent joint. Patients were discharged on oral antibiotics once fever subsided and were kept on weekly follow-up with quantitative CRP reports. The infection resolved completely without sequela or significant functional impairment in all these 14 patients.

In one of the patients with involvement of adductors, MRI showed abnormal diffuse enhancement of the left hip adductors owing to diffuse inflammation (Fig. 1). She was discharged from hospital after 3 weeks of i.v. antibiotic treatment and a delayed closure of her surgical wounds; oral treatment with cloxacillin alone was administered for 3 more weeks. Upon discharge, all blood tests were within the normal range.

In another patient with the right side hip adductor involvement, pus was also drained from the hip joint. Two months later, during a routine follow-up visit at the outpatient clinic, a radiograph of her leg showed signs of involvement of the proximal femur (Fig. 2). However patient was able to walk without any pain.

Three patients had enlargement on axial sections of MRI of the gluteus muscles. Image of one of these patients is shown in Figure 3 with right gluteus involvement. Enlargement and change in echogenicity (hyperechogenicity) of the gluteus is demonstrated on the right, comparable to the left normal, hypoechonic muscle.

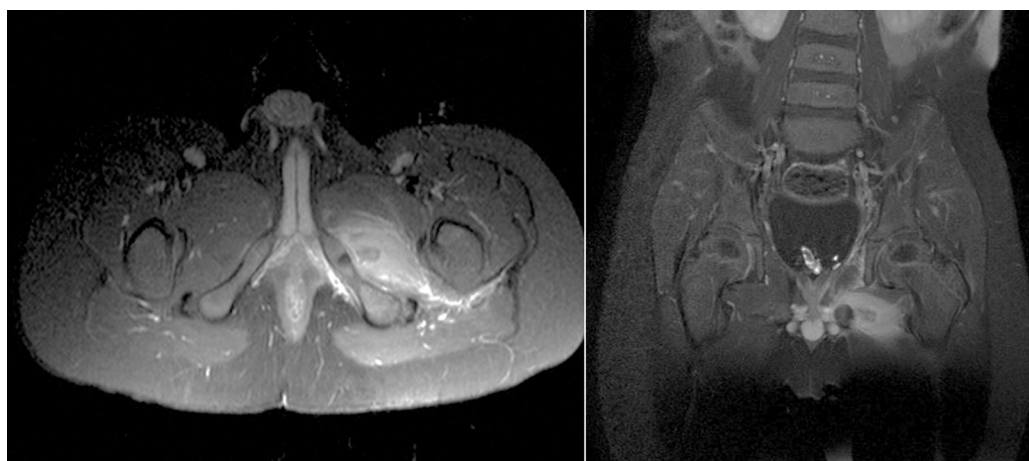


Fig. 1. Axial section of pelvis MRI showing left adductor involvement with inflammatory fluid surrounding the muscle



Fig. 2. X-ray showing bony involvement secondary to hip adductor involvement leading to septic hip



Fig. 3. Axial section of pelvis MRI: right gluteus medius showing early septations with inflammatory fluid surrounding the muscle

Three patients had enlargement on axial cuts of MRI of the quadriceps muscles. Image of one of these patients is shown in Figure 4.



Fig. 4. Shadow of the quadriceps muscle, which suggested the possible involvement of the muscle

Patients can be divided into stages depending on the severity of infection and treatment was started accordingly. In our study, 5 patients were in stage 2 and 9 were in stage 3, none were stage 1. Therefore all 14 patients were operated (Table 5).

Table 5

Distribution of patients according to stage at which patient presented

Stage of infection	No of Patients
Stage 1	0
Stage 2	5
Stage 3	9

DISCUSSION

PPM is a rare, subacute, deep bacterial infection of muscles. It is also called tropical myositis, infective myositis, pyogenic myositis, suppurative myositis, myositis purulenta tropica, epidemic abscess or bacterial pyomyositis. It commonly manifests as a local abscess, but can also present as a diffuse inflammatory or rapidly progressing myonecrotic process [8]. In tropical countries, peak incidence is at 2 to 5 years of age [10]. In our study, mean age was 2 years and 1 month. A male preponderance is found in almost all series, with a male to female ratio of around 2:1 to 3:1 usually reported [4, 5]. In our study also, male patients constituted 64.3 % of the cases (9/14) and the rest were females.

The etiology of primary pyomyositis remains unclear. The infection is believed to be a complication of transient bacteremia because it develops without an obvious penetrating injury or any other clear portal of entry in the vast majority of patients [1]. Trauma to the affected muscle resulting in alteration of the muscle structure has been proposed as a possible etiology [8, 9]. It has also been hypothesized that trauma alters local muscle tissue structure, thereby creating a locus of infection for implantation of bacteria from a subsequent, untreated bacteremic

episode [11]. In our study, 42.9 % patients had a history of minor trauma events before the acute onset of the infection, such as a fall at home while playing. Souid et al and Viani et al also reported around 41 % rate of a history of recent trauma to the hip in their respective studies [12, 13].

In our patients, clinical symptoms and signs included fever ranging between 100 and 103 degree F, local tenderness, swelling, erythema, local rise of temperature and pain around the infection site, limping due to pain and limited range of motion at the adjacent joint. Laboratory tests revealed marked elevation of the ESR (> 55 mm/h) and CRP (> 70 mg/l) levels, and elevated white blood cell count (> 16000/microL). Of these, we used CRP in monitoring the course of the disease and the response to the chosen antibiotic treatment as it is the most sensitive.

Imaging studies are of paramount importance in the diagnosis of pyomyositis. Harrington et al stated that although USG did not provide the diagnosis in the case they described because it had been performed early in the evolution of the condition, repeated US examinations would have detected the pus collections [14]. We found USG to be a useful and easy tool for initial diagnosis at the time of presentation enabling guided percutaneous needle

aspiration as well as for documenting the abscess formation and for repeated monitoring of the infection site to verify satisfactory progress until full recovery. USG revealed collections around the quadriceps muscle, iliopsoas muscle, gluteal muscle, adductors and obturator muscles. However, one disadvantage of USG which we felt was that it was unable to tell the magnitude of infection. Therefore, MRI was necessary.

MRI is the most useful imaging technique for the diagnosis of pyomyositis, as it most clearly demonstrates diffuse muscle inflammation and any subsequent abscess formation [11]. Yuh et al asserted that MRI is superior to CT scanning in diagnosing pyomyositis, and Mazur et al found MRI to be 97 % sensitive for acute musculoskeletal infections in children [15, 16]. Bickels et al regarded MRI as the investigation of choice for acute pyomyositis [8]. We agree with these authors so we also got MRI done in all of our patients which helped us make an early diagnosis.

The disease is considered to have three distinct stages. Choice of treatment depends on its stage at presentation. In stage 1, patient usually has insidious onset of dull, cramping, progressive pain associated with a low-grade fever, general malaise and muscle ache. In stage 2, muscle abscess formation is there. Overlying skin is swollen, erythematous and warm. In stage 3, along with stage 2 features there are systemic manifestations of sepsis [8, 17, 18]. Because of the vague clinical presentation, it is unlikely to be diagnosed during the initial stages. Also, if the affected muscle is deeply situated and local signs are not apparent, diagnosis might be delayed, which could result in extension into and destruction of an adjacent joint, sepsis and, occasionally death [14]. One of the reasons for primary iliopsoas pyomyositis being rare is that diagnosis is usually delayed because of the deep location of the muscle and because symptoms often resemble those of a primary infection in the hip [17]. During the early stage of infection, diffuse inflammatory changes can be effectively treated with antibiotics alone [19]. However if there occurs abscess formation, it would require drainage with large skin incision and wide exposure of the affected muscle before the initiation of antibiotic therapy [10]. Patients in our study were either in stage 2 or 3. Therefore in all 14 patients, infected site was surgically opened by wide incision, collection was drained, intraoperative culture samples were taken, and wound was thoroughly washed with normal saline and was left open. The wound was packed and daily dressings were done. Samples were sent for gram stain and culture sensitivity.

Causative organisms reported in the literature in pyomyositis are *S. aureus*, *Streptococcus* group A, *Escherichia coli* and *Enterococcus* [20, 21]. However, *S. aureus* is the most common causative agent, responsible for 50–95 % of pyomyositis cases in all age groups [10]. In our case series, 50 % (7/14) patients had MSSA, 28.6 % (4/14) had MRSA while in 21.4 % (3/14) cases cultures were sterile. This distribution of pathogens resembles

the findings previously reported in various studies [8, 14, 22]. Patients with MSSA in culture had intravenous administration of amoxicillin/clavulanic acid. One patient with MRSA received clindamycin and the other 3 patients with MRSA cloxacillin on the basis of the antibiotic sensitivity in the culture report. In 3 patients, where there was no collection, culture came out to be sterile, so in them broad spectrum antibiotics, i.e. amoxicillin/clavulanic acid combination, were continued. Causes for culture coming sterile could be insufficient sample or previous history of antibiotic intake. Thus when treating such patients, one should be careful to fully investigate the patient to find out the etiology rather than starting antibiotics recklessly as the latter would mask the real cause behind the pathology and also lead to spread of antibiotic resistance.

The recommended duration of treatment has not been well established, and has varied from 1 to 6 weeks based on clinical severity. In our cases, patients which were early presenters and responders were given antibiotics for a shorter duration compared to those which presented late or were showing slow response. Our tendency was to shift to oral antibiotics as early as possible. We continued i/v antibiotics till fever subsided and then patient was discharged on oral antibiotics. Oral antibiotics were also discontinued once ESR and CRP values were declining and were near normal. Average duration of antibiotic treatment in our study was around 5 weeks.

Pyomyositis usually involves the largest muscle groups located around the pelvic girdle and lower extremities [8]. Iliopsoas pyomyositis was long considered to be the most common form of this infection, but many of the cases were actually secondary infections that had developed as an extension from adjacent tuberculosis in the spine, the iliac lymph nodes or in patients suffering from inflammatory bowel disease [23, 24]. However, none of our patients had a history of tuberculosis or inflammatory bowel disease, yet the iliopsoas muscle was still the most common site of infection in our study (4 out of 14 children) which indicates some alternate pathology. Iliopsoas was followed by quadriceps and gluteus muscle (3 patients each) followed by hip adductors and obturator muscle (2 patients each).

Complications and long-term sequelae of PPM include osteomyelitis of adjacent bones, osteonecrosis, chondrolysis, complete head and neck resorption, muscle scarring, injury to growth plate resulting in limb length discrepancies, residual weakness and functional impairment due to fibrous ankylosis. Lack of awareness of the less common locations of infection may account for the delay in diagnosis, among other factors, which may lead to the above consequences [9].

In our case series, 3 patients with collection around the thigh had some changes in bone morphology such as periosteal thickening, focal osteopenia and sclerosis, but patients were able to walk without difficulty. Three had septic hip but were managed at the appropriate time that prevented any significant functional impairment.

CONCLUSION

Thus, our study shows that early diagnosis, complete drainage of the purulent material and the use of appropriate antibiotic therapy are the keys to

successful treatment that lead to complete resolution in the majority of the cases. Also due to limited availability of sophisticated imaging techniques on a routine basis,

especially in developing countries like India, a thorough history and physical examination, together with high clinical suspicion and awareness of this potentially severe condition, may be decisive for an accurate

diagnosis and management of primary pyomyositis. Providing an antibiotic therapy after a complete work up, we also can decrease the risk of development of antibiotic resistance.

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Patient perspective All patients received the best possible treatment as per the availability of resources in the hospital. All 14 patients included in the study recovered and were able to do their day to day activities without difficulty. All patients were completely satisfied with the treatment.

Informed consent Written informed consent was taken from parents of every patient included in the study. Also patients underwent all investigations with their parents fully aware of their advantages and disadvantages. Written consent was also taken before every procedure after full explanation of the steps of procedure and also the advantages and disadvantages of the procedure.