Primary pyomyositis in children: a retrospective analysis of 11 cases
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This study was undertaken to review our approach to diagnosis and treatment in a series of 11 patients (mean age 8.2 years) with primary pyomyositis, who had neither an underlying disease nor a compromised immune system. Nine of the children had positive blood cultures, \textit{Staphylococcus aureus} (eight) and \textit{Streptococcus group A} (one). The sites of infection were iliopsoas (four), obturator (two), hip adductors (two), levator scapula (one), thoracolumbar paraspinal (one) and gastrocnemius (one) muscles. Antibiotic treatment was initially intravenous, followed by oral administration. Of five patients with evidence of abscess formation, three underwent percutaneous drainage, whereas two required open surgical drainage. The infection resolved completely without any sequela in 10 children. One patient who developed acute compartment syndrome showed late signs of osteonecrosis of the tibial shaft segment. \textit{J Pediatr Orthop B} 16:153–159 © 2007 Lippincott Williams & Wilkins.

Keywords: conservative treatment, disease management, primary pyomyositis, surgical procedures

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Introduction
Primary pyomyositis (otherwise known as tropical myositis, infective myositis, pyogenic myositis or myositis purulenta tropica) is a rare, subacute, deep bacterial infection of skeletal muscle not associated with any contiguous infection of the skin, bone or soft tissues [1–8]. It usually involves the largest muscle groups around the pelvic girdle and lower extremities [2,3,9–18]. In tropical countries, the infection accounts for around 4% of hospital surgical admissions, but it is far less common in temperate climate regions [19], in which it is unlikely to be considered during the initial stages of differential diagnosis. In addition to its low incidence, the vague presentation of primary pyomyositis may also contribute to a delay in diagnosis [18], at times with subsequent complications such as compartment syndrome, extension into and destruction of an adjacent joint, sepsis and, even death [11,20]. Although it can affect individuals of all ages (with a slight male predominance), primary pyomyositis is most common in the first and second decades of life [2,3,5,21–24]. Patients with pyomyositis who are more than 30 years old, commonly have an underlying condition that impairs the immune system such as diabetes mellitus, malnutrition, AIDS, malignancies, chronic liver disease and drug abuse [11]. These patients are considered to have secondary pyomyositis and may have an increased risk of multifocal disease, an atypical presentation and minimal symptoms because of neutrophils dysfunction.

Primary pyomyositis has been only sporadically described in the literature. Taking in consideration the increasing population mobility owing to tourism and globalization, an increase in the number of affected adults and children attending medical facilities in nontropical countries can be expected. For these patients, an accurate prompt diagnosis and early treatment are critical. As most of its signs and symptoms are mainly musculoskeletal, it is worthwhile to highlight the information concerning this entity, including the most important clinical data for the use of orthopedic surgeons. For this purpose, we retrospectively analyzed our experience with a series of 11 pediatric patients with primary pyomyositis who were treated and followed up in our department.

Materials and methods
Patients
Between May 2000 and December 2004, we diagnosed and treated 11 children (five girls and six boys, mean age 8 years and 2 months, range 8 months – 14 years and 1 month) with primary pyomyositis. None of these patients had an underlying disease that might impair the immune system. In all, there were four patients in which the iliopsoas muscle was involved, two cases in which the obturator muscle was the infection site, two other cases in which the hip adductors were infected and one case each in which the thoracolumbar paraspinal muscles, the levator scapula and the gastrocnemius muscles were involved.
All pertinent data on the children's clinical signs and symptoms, the results of laboratory and bacteriology tests and the findings on imaging studies, as well as the type and outcome of treatment and invasive procedures, and the clinical course and final outcome were recorded and reviewed (Table 1).

**Clinical signs and symptoms**

Painful limping was the most common presenting symptom followed by fever of up to 40°C (range 38–40°C), general signs of sepsis, swollen extremity, flexion contraction of the hip joint (positive Thomas test), various degrees of limited range of motion at the adjacent joint, compartment syndrome, abdominal pain mimicking acute appendicitis and cervical pain with torticollis. The duration of symptoms ranged between 2 and 14 days before children's hospitalization. Four children had a history of mild trauma before the onset of the disease, usually related to excessive gymnastics, weight lifting, dancing or a fall injury.

**Laboratory findings**

All patients had marked elevation of acute phase reactants. Erythrocyte sedimentation rate between 26 and 120 mm/h and C-reactive protein (CRP) between 45 and 251 mg/l. The white blood cell count ranged between 3900 and 27 000/l, whereas the platelets count ranged between 60 000 and 304 000/l. Among these, we found the CRP to be the most accurate predictive indicator for the monitoring of the efficacy of the treatment. When CRP levels returned to normal, it clearly indicated the remission of the disease, with no residual sites of active bacterial infection.

**Bacteriology**

Initial blood cultures were positive in nine out of 11 patients: *Staphylococcus aureus* was detected in eight cases and *Streptococcus* group A in one. This distribution of pathogens resembles the findings previously reported [11,25,26]. Four patients also had cultures taken directly from the site of infection, three of which were positive with the same causative organism as the one that grew on the blood culture. One patient had a negative culture taken from an abscess in the iliopsoas after 4 days of appropriate antibiotic treatment. At admission, the two youngest children in our series had negative blood cultures: a 9-month-old boy with multifocal pyomyositis at both the right iliopsoas and gluteus muscles that was demonstrated on computed tomography (CT) scan and repeated ultrasonographic studies (US) (Fig. 1), and a 1 year and 11 months old boy with pyomyositis of the right hip adductor muscle that was proven on magnetic resonance imaging (MRI). Of note is that before their admission, none of our patients received any antibiotic treatment.

Repeated blood cultures remained positive for a mean duration of 6 days (range 5–8 days). The patient who had a Streptococcal infection is of special interest. This 5 years and 10 months old girl had a clinical presentation of a septic arthritis of the hip joint (case no. 2). US of the hips showed a marked effusion on the affected side, and because she had signs of sepsis, the patient was taken immediately to the operating theatre. A formal hip arthrotomy was performed via an anterior approach, and pus was drained from the joint space. Cultures taken from the pus were also positive with the same Streptococcal group A pathogen as evidenced in the blood cultures taken several hours earlier. She showed good clinical improvement after 2 days of appropriate postoperative intravenous (i.v.) antibiotic treatment with the combination of penicillin and clindamycin, but 8 days later, her temperature increased once again to 39.5°C. The patient once again showed general signs of sepsis with ileus involvement and was hemodynamically unstable, and was transferred to the pediatric intensive care unit for treatment.

<table>
<thead>
<tr>
<th>No.</th>
<th>Age (years)</th>
<th>Presenting symptom</th>
<th>Fever on admission (°C)</th>
<th>WBC count</th>
<th>ESR</th>
<th>CRP</th>
<th>Infection site</th>
<th>Pathogen</th>
<th>Culture source</th>
<th>Treatment</th>
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<tbody>
<tr>
<td>1</td>
<td>12.5</td>
<td>Limp + pain</td>
<td>39</td>
<td>19000</td>
<td>120</td>
<td>58</td>
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<td><em>S. aureus</em></td>
<td>Blood</td>
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<tr>
<td>2</td>
<td>5.9</td>
<td>Septic hip</td>
<td>40</td>
<td>27000</td>
<td>55</td>
<td>141</td>
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<td><em>S. aureus</em></td>
<td>Hip</td>
<td>Drainage</td>
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<td>Torticolis</td>
<td>38</td>
<td>4400</td>
<td>115</td>
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<td>Levator scapula</td>
<td><em>S. aureus</em></td>
<td>Blood</td>
<td>Antibiotics</td>
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<tr>
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<td>37</td>
<td>14300</td>
<td>60</td>
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<tr>
<td>5</td>
<td>13.6</td>
<td>Limp + pain + LBP</td>
<td>40</td>
<td>12400</td>
<td>106</td>
<td>251</td>
<td>Iliopsoas</td>
<td><em>S. aureus</em></td>
<td>Blood + abscess</td>
<td>Drainage</td>
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<tr>
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<td>Compartment syndrome of leg</td>
<td>39.8</td>
<td>17200</td>
<td>90</td>
<td>125</td>
<td>Gastrocnemius</td>
<td><em>S. aureus</em></td>
<td>Blood + muscle</td>
<td>Fasciectomy</td>
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<td>40</td>
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<tr>
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<td>34</td>
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<td>Blood</td>
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<td>Abdominal + back pain</td>
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<td>55</td>
<td>167</td>
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<td><em>S. aureus</em></td>
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<td>41</td>
<td>Hip adductors</td>
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</table>

CRP: C-reactive protein; ESR, erythrocyte sedimentation rate; *S. aureus*, *Staphylococcus aureus*; WBC, white blood cell; LBP, low back pain.
treatment and observation. She underwent an urgent CT scan of the abdomen and pelvis, which revealed signs of pyomyositis on the ipsilateral iliopectos muscle, including abscess formation. The patient underwent a second operation for aspiration and drainage of the abscess and subsequently showed a gradual but consistent clinical improvement until the discharge from hospital without any sequela or functional impairment.

**Imaging studies**

All 11 patients underwent plain radiographs of the affected area and none showed any bony involvement on admission. Three patients had enlargement of the shadow of the psoas muscle, which suggested the possible involvement of the muscle, two patients had effusion of the hip joint and one patient had marked enlargement of the calf muscles of her leg. They all also underwent US of the involved region. In one case (case no. 9), US was found especially helpful for the primary detection of an abscess formation, for enabling percutaneous drainage of the abscess and for ruling out bony involvement (lack of periosteal elevation). It was also valuable for the routine monitoring of all the patients during their hospitalization.

Eight patients underwent CT. The four cases who had pyomyositis of the iliopectos muscle underwent a CT scan of the abdomen and pelvis (Fig. 2), the two patients with an involved obturator muscles and the patient who had pyomyositis at the levator scapula muscle had scans of the affected regions (Fig. 3). The patient with pyomyositis of the paraspinal muscles (case no. 10) who was initially suspected as having acute appendicitis also had a CT scan of the abdomen.

Technetium scintigraphy was performed in seven children followed by gallium scans in two of them; evidence of increased soft tissue uptake helped to confirm that there was no involvement of adjacent bone. It should always be kept in mind that there is hypervascularization of the affected muscle, which might cause a secondary effect on the adjacent joint; in two of our patients with pyomyositis of the psoas muscle it showed an increased uptake of the sacroiliac joint that was misinterpreted as sacroileitis on the bone scan.

MRI was carried out in only four patients. It clearly and most accurately demonstrated the diffuse muscle inflammation (Fig. 4).

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**Fig. 1**

Axial ultrasonographic posterior view of the gluteus muscles. Enlargement and change in echogenicity (hyperechogenicity) of the gluteus is demonstrated on the right comparable to the left normal, hypoechoic muscle (between calipers). Note also a small lesion filled with hypodense fluid (arrow), finding consistent with a small abscess within the enlarged, inflamed muscle.

**Fig. 2**

Axial contrast enhanced computed tomography (a) and ultrasound (b) scans of the pelvis at the level of the iliopsoas muscles both demonstrating nonhomogenous enlargement of the left iliopsoas muscle (1.8 cm on left versus 1.1 cm on the right) consistent with a diffuse inflammatory process. Moreover, note the disappearance of the normal femoral nerve sheath on the left side in comparison with its normal appearance on the right.
Treatment
The initial treatment was i.v. antibiotics for 10–21 days, the administration period depending on the stage of the disease and the resultant clinical and laboratory improvement. We usually began empirically with a second-generation cephalosporin or with a combination of cloxacillin and aminoglycoside. It was necessary to change the treatment according to the sensitivity of the bacteriogram in three patients, and in one because of the lack of clinical improvement after 4 days of treatment with cephalosporin. Of these patients, three received a combination of clindamycin and rifampin, and a combination of penicillin and clindamycin was used in the fourth. The antibiotic treatment was then continued orally for an overall duration of 6 weeks. Conservative treatment protocol alone with i.v. antibiotics was effective in six children.

Invasive procedures
Three patients required percutaneous drainage of an established abscess: one was CT-guided and two others were US-guided. Two other patients required open surgical drainage: one had compartment syndrome of the leg on initial presentation and the other had the streptococcal infection described above.

Clinical course and outcome
Five patients showed marked improvement within 2–3 days from the start of antibiotic treatment, with gradual resolution of the limp, local tenderness and swelling, as well as a return to full range of motion at the adjacent joint and a decline of the laboratory markers of acute phase reactants. Four patients who failed to show any improvement within 72 h of i.v. antibiotic treatment were further evaluated with CT or US, which revealed the presence of an abscess in either the iliopsoas or the obturator muscles. The abscesses were immediately and successfully drained in three patients, and improvement in the patients’ condition followed shortly thereafter. One patient had some very small abscesses in the iliopsoas and gluteal muscles. Their size made it impossible to perform a percutaneous CT or US-guided drainage. As the blood cultures were negative, the child’s antibiotic treatment was changed empirically from a second-generation cephalosporin to a combination of clindamycin and rifampin. He showed a marked improvement within the following 48 h. Another patient (case no. 2) required open drainage of a septic hip joint that initially was thought to be the only pathology but later emerged as being a secondary complication of a primary pyomyositis of the iliopsoas muscle and sepsis. The infection resolved completely without sequel or functional impairment in all these 10 patients.

One girl (case no. 6) had signs of compartment syndrome of the leg at presentation, requiring emergency fasciotomy. Cultures taken both from her blood and from the site of operation grew S. aureus and she was treated with a combination of cloxacillin and gentamycin, to which the patient showed a very good response. She was discharged from hospital after 3 weeks of i.v. antibiotic treatment and a late primary closure of her surgical wounds; oral treatment with cloxacillin alone was administered for an additional 3 weeks. Upon discharge, all blood tests were within the normal range. Two months later, during a routine follow-up visit at the outpatient clinic, a radiograph of her leg showed signs suggestive of extensive

Fig. 3
Axial contrast-enhanced computed tomography scan of the cervical spine (C7) demonstrated non-homogeneous enlargement of the left erectospine muscle, consistent with inflammation (arrows). The central located hypodense area (arrow head) represents a more necrotic focus and abscess formation.

Fig. 4
Axial T1 MRI image with fat saturation showing abnormal, diffuse enhancement of the left hip adductors owing to diffuse inflammation (between arrows).
osteonecrosis of the tibia (Figs 5a and b). As osteomyelitis could be one of the late complications of pyomyositis, the patient underwent another operation and, this time, an effective debridement of all the necrotic bone was carried out, leaving a wide area of missing bone (Figs 5c and d). Histological studies of the specimens, taken during the operation, revealed necrotic bone with no signs of osteomyelitis. Postoperatively, a long leg cast was applied. The last radiographs showed that the area of bone loss is improving, but it has not returned to normal yet (Figs 5e and f).

**Discussion**

Primary infection of muscle is usually regarded as a tropical disease, and has been rare in temperate climates until recent times [3,27]. It is possible that the subtropical climate in our country, which is somewhat in between, may explain the number of patients reported here.

The etiology of primary pyomyositis remains unclear. Despite the frequency of bacteremia, infection in muscle is surprisingly rare and, indeed, it is difficult to produce experimentally [8]. 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 [3,22,28–30]. Trauma to the affected muscle resulting in alteration of the muscle structure has been proposed as a possible etiology [11,20]. It has also been hypothesized that trauma alters local muscle tissue structure, thereby creating a locus minoris for implantation of bacteria from a subsequent, untreated bacteremic episode [31]. Among the 11 patients in our current series, four sustained minor trauma events before the acute onset of the infection, such as a fall or strenuous exercise. This resembles the 41% rate of a history of recent trauma to the hip that was reported in previous studies [32,33].

The disease is considered to have three distinct stages, which represent a gradual progression from diffuse inflammation of the affected muscle (stage 1) to focal abscess formation (stage 2) and finally to a septic state (stage 3) [11]. In our group of patients, we saw the full spectrum of these three stages: there was one patient who was diagnosed when he was still in stage 1 of the disease (case no. 4), eight were in stage 2 (case no 1,3,5,7,8,9,10,11) and two others were in stage 3 (case nos 2,6).

The causative organism reported in the literature is usually *S. aureus*, but *Streptococcus* group A, *Escherichia coli* and *Enterococcus* have also been implicated [3,13]. The findings of our study support these data. All but two of our eleven patients had positive blood cultures, eight with *S. aureus* and one with group A streptococcal bacteremia. It is important to monitor the blood cultures.
as the bacteremia persists for a few days even when proper antibiotic treatment has been applied. It should be noted that the two children with negative blood cultures were the youngest in our series, one aged 8 months and the other 1 year and 11 months.

Pyomyositis usually involves the largest muscle groups located around the pelvic girdle and lower extremities [11], but it can affect muscles in other body regions as well [3], as we found in our series of exclusively pediatric patients. Iliopsoas pyomyositis was long considered to be the most common form of this infection, but many of these 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 [34,35]. 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 (four out of 11 children), followed by the obturator muscle (two patients), the hip adductors (two patients), whereas the affected area in the other three children was levator scapula, paraspinal and gastrocnemius, respectively. The distribution in our series of otherwise healthy children demonstrates once again that every starched muscle in the body can be involved, including those less frequently reported in regions such as spine or upper extremity or trunk [11,14,24]. Lack of awareness of the less common locations of infection may account for the delay in diagnosis, among other factors, sometimes with tragic consequences [20].

Our patients’ clinical symptoms and signs included fever (usually between 39 and 40°C), local tenderness and pain around the infection site, abdominal pain in the patient whose infection was located in the paraspinal muscles, limping (if the infection was around the hip joint) and limited range of motion at the adjacent joint. Laboratory tests revealed marked elevation of the erythrocyte sedimentation rate (> 55 mm/h) and CRP (> 60 mg/l) levels, and a white blood cell count within the normal range or moderately elevated. Of these, we feel that the CRP is the most sensitive and is clinically helpful in monitoring the course of the disease and the response to the chosen antibiotic treatment.

Yuh et al. [36] contended that MRI is superior to CT scanning in diagnosing pyomyositis, and Mazur et al. [37] found MRI to be 97% sensitive for acute musculoskeletal infections in children. Bickels et al. [11] regarded MRI as the investigation of choice for acute pyomyositis. We agree with these authors. When carried out, MRI was found to be the most sensitive diagnostic modality. Harrington et al. [26] stated that although US 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. We found US to be a useful tool for enabling guided percutaneous needle aspiration and drainage of abscess formations, as well as for documenting the abscess formation and for repeated monitoring of the infection site to verify satisfactory progress until full recovery.

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 vast majority of the cases [11], including our series of children with the exception of one patient who presented with pyomyositis of the gastrocnemius complicated by compartment syndrome of the leg. We suspect that the elevated compartment pressure together with the marked edema of the muscles adjacent to the tibia caused a significant reduction in the periosteal blood supply to the bone in that region, resulting in an area of osteonecrosis.

Considering the limited availability of sophisticated imaging techniques on a routine basis, we entirely agree with King et al. [38] that a thorough patient’s 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.

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References


