Tropical primary pyomyositis in children of the UK: an emerging medical challenge

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Abstract Pyomyositis is a commonly encountered condition in the tropics. It was not described in the UK until 1998. The reason for the increasing incidence is not understood. We sought to identify the experience gained of this condition within a UK paediatric tertiary referral unit. Retrospective review of cases of pyomyositis from our institution since 1998 was undertaken to identify demographics, presentation, diagnosis and management. Thirteen cases were identified. The obturator internus was most commonly affected (62%). *Staphylococcus aureus* was cultured in nine cases (69%). One diagnostic retroperitoneal exploration was performed and all cases were identified by computed tomography or magnetic resonance imaging. To our knowledge, this is the first UK series of pyomyositis, reflecting its increasing Western incidence. Early diagnosis and treatment with antibiotics is all that is needed in the majority of cases. A greater awareness of this emerging condition is necessary to prevent misdiagnosis and unnecessary surgical intervention by all surgeons.

Introduction

Primary pyomyositis is a primary infection of skeletal muscle [1, 4–7]. It is a diagnosis which is common in warmer climates—hence is also called ‘tropical pyomyositis’—and frequently has fatal consequences. It largely affects muscles around the hips and can mimic a range of acute paediatric disorders such as septic arthritis or appendicitis [15]. The infection was virtually unknown in the temperate climates of the UK until recently. Our case series provides evidence that this is a diagnosis which requires increasing awareness amongst paediatric physicians and surgeons alike—and one which must be considered amongst differential diagnoses of hip or abdominal pain.

Materials and methods

This study was conducted in our tertiary paediatric orthopaedic unit. All cases of primary muscle infection in children (16 years and under) between 1998 and 2007 were collected retrospectively using the hospital coding systems, and radiology and medical case records. Inclusion required that there be muscle oedema or swelling on magnetic resonance imaging (MRI) scan associated with appropriate symptoms of local pain and tenderness and signs of infection that responded to antibiotic treatment. Patients who had infection of muscle secondary to open trauma and/or primary osteomyelitis or previous other surgical infections were excluded.

Patients in the age group of 0–16 (mean age: 10 years) who had MRI of the trunk, any of the upper or lower limbs, pelvis and hip were identified. Coding classification terms like pyomyositis, muscle infection and muscle abscess were used in the search. There was significant overlap of these two

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methods except for one patient identified from the radiological database who was not reflected in the medical record search.

We reviewed the epidemiology. We also looked into the clinical presentation of each case, including symptoms before initial presentation, weight-bearing status, any history of predisposing trauma fever and past medical history. Laboratory results of white cell count (WCC), erythrocyte sedimentation rate (ESR), C-reactive protein (CRP) and blood cultures were noted. Imaging procedures like X-rays, ultrasonography (US), bone scintigraphy, computed tomography (CT) and MRI were reviewed. All children were followed up to six months and discharged.

Results

A total of 13 patients (seven boys and six girls) with a diagnosis of primary pyomyositis were identified. The age ranged between four and 15 years. There was no clear seasonal distribution, but there was a trend towards the spring and summer months (see Table 1).

All subjects presented with either a limp or inability to bear weight. Three patients had a history of symptoms in the affected area following closed trauma after a football injury, a trampoline injury and a fall, respectively.

All but one of the subjects presented within three to four days of the onset of symptoms. The only one that presented late (after three weeks) had idiopathic juvenile arthritis and thought he may have bruised his hip after jumping on the trampoline. All other patients had no significant past medical history.

At presentation all 13 had a raised temperature (range: 37.8–40°C) and CRP (range: 71.6–436). Leucocytosis was noted in nine patients. Four had normal WCC. Unfortunately ESR (range: 0–10 mm) was not measured in four patients and was elevated in the rest. A provisional diagnosis of septic arthritis was made in the majority of cases until US scan showed no effusion in the suspected joint. Two patients were initially thought to have appendicitis. One underwent a negative diagnostic laparotomy and pyomyositis was subsequently diagnosed by MRI. The second underwent laparotomy which identified a normal appendix, and the third had a right adductor muscle abscess identified which was incised and drained.

Once the clinical diagnosis was made most patients responded to antibiotic therapy and no further intervention was necessary. Organisms were identified in ten (77%) patients following blood culture. *Staphylococcus aureus* was the most common organism isolated in nine patients (69 %). One had *Streptococcus pyogenes*.

As all patients presented with either a limp or inability to bear weight, X-rays of the pelvis were obtained in every case. Bony radiographs were normal in all patients, although

initially a suggestion of an avulsion fracture of the ischial tuberosity secondary to jumping on the trampoline was considered in one patient. Another patient was thought to have modest subcutaneous oedema, but in retrospect this was normal.

All subjects where septic arthritis of the hip was the initial diagnosis underwent US assessment of the hip joint. When this proved to be normal with no evidence of joint effusion, septic arthritis was considered unlikely and a search for an alternative source of the symptoms was commenced. Ten subjects underwent US of the lower abdomen and pelvis; one had some evidence of inconclusive groin swelling, another with minimal free intraperitoneal fluid and the third child with the history of trampoline trauma showed asymmetry of soft tissues overlying the iliac crest. Normal study was seen in the rest. One patient underwent a CT of the pelvis. The CT identified typical enlargement of the piriformis muscle with focal low attenuation area and peripheral enhancement post-contrast [2]. No patient had bone scintigraphy.

Eventually all 13 patients underwent MRI of the pelvis and hip region in order to identify the source of symptoms. MRI was able to make the diagnosis in every case. Oedema and swelling of the affected muscles was easily identified. A high signal was also seen in the surrounding bony structures in five patients (40%). The pelvic girdle muscles were the primary site of infection in most patients. The muscles involved were obturator internus (8/13=54%), obturator externus and adductors (23%), iliopsoas, gluteus medius and gluteus minimus. Abdominal muscles such as external and internal oblique along with transverse abdominis were the other muscles affected. There were five patients with multiple pelvic girdle muscle involvement. In one patient the muscles of the anterior compartment of the shin including tibialis anterior were affected.

As soon as the diagnosis was made, intravenous (IV) cefuroxime was started. In one patient antibiotic treatment was delayed until urgent surgery to decompress an abscess in the adductors had been performed and material for culture obtained. Compartment release of the leg was performed in the patient with tibialis anterior involvement and in one diagnostic laparotomy had to be done.

All patients improved promptly after antibiotic treatment was commenced. The IV antibiotic was continued for five days (mean) and was switched over to oral cephalosporins once the patient showed signs of improvement. The oral antibiotic was continued for a minimum of three weeks and a maximum of six weeks. Complete recovery was noted by three months after commencement of treatment and was confirmed both clinically and a repeat of the MRI scan. Patients were followed up initially once in six weeks and thereafter once in three months. There were no post-treatment sequelae in any of our patients who were followed up for 15 months.

Table 1 Demographics of the 13 patients

Patient	1	2	3	4	5	6	7	8	9	10	11	12	13
Age	11	13	6	15	11	11	6	6	7	13	15	4	4
Month	Aug 07	Sep 07	Apr 06	Sep 01	Apr 05	May 05	Oct 07	Sep 07	Dec 98	May 02	Jul 05	Feb 99	Aug 07
Days before presentation	7	4	2	3	2	3	2	3	4	7	21	3	3
Weight-bearing	Unable	Unable	Limp	Limp	Unable	Limp	Unable	Limp	Unable	Limp	Limp	Unable	Limp
Trauma	Nil	Football	No	No	No	No	No	No	No	Fall	Trampoline	No	No
Past medical history	Nil	Nil	Asthma	Nil	Nil	Nil	Nil	Nil	Nil	Nil	Juvenile arthritis	Nil	Nil
Temperature	39	40	37.8	38.2	36.2	38.1	37.2	37.6	39	38.1	39	40	38
WCC	12.29	2.6	10.19	12.6	8.73	6.93	7.88	12.1	11.9	8.9	21.8	24.24	17.92
ESR	61	17	19	25	Not done	Not done	Not done	90	Not done	Not done	149	18.77	Not done
CRP	124.3	167	80.5	178	71.6	91.3	108	253	143	117	201	436	281
X-ray	NAD	NAD	NAD	NAD	NAD	NAD	NAD	NAD	NAD	NAD	? Avulsion of ischial tuberosity	? Subcutaneous oedema	NAD
CT	Piriformis pyomyositis	No	No	No	No	No	No	No	No	No	No	No	No
US scan	NAD	NAD	NAD	No	Groin swelling	No	NAD	NAD	Minimal free peritoneal fluid	Asymmetry of soft tissues overlying left iliac crest	No	NAD	NAD
MRI	Obturator internus	Obturator internus	Obturator internus	Iliopsoas	Adductor abscess	Obturator internus	Adductor	Obturator internus & adductor	Obturator internus	External & internal oblique, transverse abdominis, gluteus medius & minimus	Obturator internus & externus	? Necrosis of muscle in leg	Obturator internus & externus
Blood cultures	Staph A	Staph A	Staph A	Staph A	Not done	Not done	Staph A	Staph A	Staph A	Not done	Staph A	Staph A	Strep
Provisional diagnosis	Septic arthritis	Septic arthritis	Appendicitis	Ischiorectal abscess	Tuberculosis	Septic arthritis	Appendicitis	Septic arthritis	Appendicitis	Abscess	Septic arthritis	Abscess	Septic arthritis
Other cultures	Nil	Nil	Nil	Nil	Staph A	Nil	Nil	Nil	Nil	Nil	Nil	Pseudomonas	Nil
Surgery	No	No	No	No	Incision and Drainage	No	No	No	Diagnostic laparotomy	No	No	Compartment release & débridement	No
Antibiotics	Cef	Cef	Cef	Cef	Cef	Cef	Cef	Cef	Co-amoxiclav	Cef	Cef & clindamycin	Cef & flucloxacillin	Cef
Duration of antibiotics	3 weeks	3 weeks	6 weeks	Not recorded	6 weeks	Not recorded	6 weeks	3 weeks	6 weeks	5 weeks	4 weeks	6 weeks	5 weeks

NAD nothing abnormal diagnosed, *Staph A Staphylococcus aureus*, *Cef* cefuroxime

Discussion

Primary pyomyositis (PP) is an acute, primary deep skeletal muscle infection seen in the tropical population especially in parts of Africa and the South Pacific [1, 4–7]. In most patients it is probably due to underlying transient bacteraemia and is increasingly being reported in the temperate climates [2, 8–10]. It is variously called tropical myositis, bacterial myositis, myositis purulenta tropica, bacterial myositis, suppurative myositis or epidemic abscess [1]. It is reported to be rare in the USA and other temperate climates [4–7]. The incidence of the condition in the UK, both past and present, is difficult to determine given the dearth of reports of the condition. Until the late 1990s the condition had not been reported in the UK. We report the largest series in children to date. There is poor understanding of the exact aetiology. PP can occur in any age group although it is common in the first and second decade of life. Children seldom have pre-existing conditions in comparison to adults [2–8, 12–16]. Only two patients in our series had any pre-existing medical problems although not directly linked. Association of trauma at the time of bacteraemia has been postulated for the increased risk of developing PP. This may explain its high incidence in men and in the tropics. Bickles et al. in their review of 676 patients reported trauma in less than 5% of patients.

Primary pyomyositis has three discrete stages of presentation. The initial diffuse inflammation or invasive stage is where the organism enters the muscle and produces insidious symptoms like aches/pains with or without fever. It is during the second stage of focal abscess formation (purulent stage or suppurative stage) when most patients present with local and systemic manifestations. A soft tissue swelling may be palpated with overlying skin being normal or warm and erythematous. All children in our series either presented with a limp or non-weight-bearing status and fever. On examination, they will be exquisitely tender in the affected hip joints. When the purulent stage is not treated, they move on to the final, also called the toxic shock stage. The characteristic of this stage is pain and more importantly will manifest with systemic signs of sepsis. Pyomyositis can be easily confused with septic arthritis, osteomyelitis, Perthes' disease, arthritis, discitis, appendicitis, deep vein thrombosis, synovitis, perinephric abscess or soft tissue sarcomas [15]. Unifocal presentation is common, but literature evidence suggests it can be multifocal in 15–43% of cases [4, 8, 9, 11]. Pyomyositis typically affects muscle groups in the pelvis: obturator internus, externus, adductors, gluteus maximus, minimus, medius, piriformis and iliopsoas. When the iliopsoas is affected, there will be a discrete presenting symptom of pain in the retroperitoneal muscle location. The limb affected will be held in thigh flexion and external rotation.

The interpretation of laboratory investigations will rarely aid in the diagnosis of PP. Children may have normal or elevated WCC. ESR and the CRP will be invariably raised. Serum levels of muscle enzyme are usually normal [17, 18]. *Staphylococcus aureus* is the most common organism isolated in all the studies conducted in both temperate and tropical climates. B haemolytic group A streptococcus is the next commonest organism (*Streptococcus pyogenes*) followed by *Escherichia coli*. In our series, it was *Staphylococcus aureus* (61.5%) followed by *Streptococcus pyogenes*. Pantone-Valentine leukocidin (PVL) is a bacterial exotoxin secreted by staphylococcus strains that are either methicillin sensitive or methicillin resistant. This toxin is known to cause leucocyte destruction and tissue necrosis in children. We did not identify the PVL status of *Staphylococcus aureus* isolates in any of our patients. There is a suggestion in the literature to request PVL status of all *Staphylococcus aureus* isolates in suspected musculoskeletal sepsis [3].

Imaging studies are of supreme value in diagnosing PP. Plain radiographs are seldom useful in the diagnosis but appropriate to exclude any suspected bony pathology. Soft tissue swelling or abscess may be identified [1, 17]. US can be useful in the diagnosis of PP. It may be used as the first line since it is non-invasive, easily obtainable and inexpensive. Typically US excludes septic arthritis from the differential diagnosis. However, affected muscles are sometimes identified and show an abnormal echotexture with hypoechoic focal lesion with a bulky muscle mass. When US is inconclusive, MRI is preferred to CT as it is able to identify soft tissue abnormalities. Bone scintigraphy lacks sensitivity to identify pyomyositis but can show any associated osteomyelitis. MRI classically will demonstrate diffuse muscle fluid collection with a rim enhancement if an abnormal abscess is present. MRI may show inflammation in the adjacent bone, but these variations could be secondary to the muscle inflammation. Pyomyositis was diagnosed in 12 patients by MRI scan and four among them also had increased signals in the neighbouring bone. Multifocal pyomyositis was identified in four patients.

The treatment of choice is IV antibiotic in PP. In the presence of an abscess, antibiotic commencement must follow incision and drainage (I and D) of the abscess. An exception to this rule would be when the patient presents in a state of septic shock. With the exception of one child, none required I and D, which suggests children usually present in the early stage of pyomyositis. With the advancement of interventional radiological procedures, I and D can be achieved percutaneously under US or CT guidance. There is a debate on the choice of antibiotic to be used. Bickles et al. recommend cloxacillin with the addition of aminoglycoside if patients are immunocompromised or in a state of sepsis [1]. Zimelman et al. recommend clindamycin with or without penicillin as superior to B lactamase antibiotic alone

[18]. We treated all our patients with IV dose of cefuroxime appropriate for the age and weight three times daily until the child showed initial signs of clinical improvement. The IV was then changed to oral cephalosporins which were continued for a total of six weeks.

Conclusion

Primary pyomyositis is increasingly common among children in temperate climates. The aetiology is still unclear. It can affect any group of muscles in the body. Commonly involved groups are around the pelvis and lower limbs. The child usually presents with a limp or inability to bear weight on the affected limb with fever and features of being unwell. Early diagnosis and management are crucial. The investigation of choice is MRI scan. CT scan may be used as a guide for percutaneous drainage of an abscess. Appropriate antibiotic treatment should be instituted at the earliest opportunity. Potential complications are secondary to delayed presentation and diagnosis of this new challenging clinical condition. The vast majority of patients show excellent and complete recovery with no long-term complications. A high level of awareness and suspicion of this condition is warranted from all paediatric clinicians.

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