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Primary obturator internus and obturator externus pyomyositis

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Background: Pyomyositis is a rare condition in immune competent patients and is usually seen in tropical countries.

Pyomyositis of obturator muscles in particular is an extremely rare condition, which causes hip pain and mim-

ics septic arthritis.

Case Report: This is a case report of a 9-year-old boy without an underlying disease or a compromised immune system, who

presented with knee pain that progressed to hip pain and inability to bear weight. He was diagnosed initially with septic arthritis of the hip and underwent unnecessary hip exploration surgery. Magnetic resonance imaging scan was performed postoperatively and showed pyomyositis of obturator internus and obturator exter-

nus muscles. He was managed medically and had a good outcome.

Conclusions: A greater awareness of this emergency condition is necessary to prevent misdiagnosis, unnecessary surgical

intervention, and to avoid the devastating possible complications of delayed diagnosis.

Key words: obturator externus • obturator internus • obturator pyomyositis • primary pyomyositis •

septic arthritis

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Background

Pyomyositis is an acute infection of striated muscle that manifests as a localized abscess or an aggressive myonecrotic process [1]. It can be primary or secondary to neighboring or remote infection. Primary pyomyositis is a rare condition and usually affects children and adolescents, with a majority of cases involving large skeletal muscles of the lower limbs that may rarely affect the peristatic muscles [2]. It is common in many parts of Africa and the South Pacific; hence, it earned the name tropical pyomyositis [1]. It occurs more frequently in immune compromised patients, or it can be related to factors affecting the muscle itself (strenuous exercise, direct muscle trauma, viral or parasitic myositis) [3,4]. Intra-abdominal pathology, procedures, or pelvic osteomyelitis can cause pyomyositis [5]. Pyomyositis is rare and it is even difficult to produce experimentally [6]. Its rarity comes from the high level of resistance of striated muscle to episodes of bacteremia [7]. It has been suggested that pyomyositis begins when a hematoma in a muscle from recent trauma is colonized during an episode of bacteremia [3]. The most common causative organism (70-90%) is Staphylococcus aureus. Other causative organisms include Streptococcus pyogenes, Neisseria gonorrhoeae, Enterococcus faecalis, Escherichia coli, Salmonella enteritidis and Mycobacterium tuberculosis [1,4,7].

The incidence of pyomyositis is on the increase world-wide among children and adults, but it is not clear whether this increase is real, or reflects the use of modern imaging techniques like magnetic resonance imaging (MRI) [3,8]. Pyomyositis of the obturator internus (OI) and the obturator externus (OE) in particular is an extremely rare condition that can affect healthy youth (mean age: 9.5 years); however, no age group is exempted. The condition mimics other more common conditions of this area as septic arthritis and transient synovitis of the hip [1]. Moreover, reactive hip effusion may develop in cases of pyomyositis of nearby muscles such as the OI and OE, which complicate the diagnosis process and further confuses the clinical picture [5]. The infection is subfascial; the usual soft tissue inflammatory signs are not apparent in the early stages (like the case in hand) and thus the correct diagnosis might not be considered in the working diagnosis list [9]. Although infection of the obturator muscles is exceptionally rare, it is possibly more frequent than assumed. It may be that non-diagnosed cases were previously treated empirically with antibiotics without a firm diagnosis [3,8].

The OI and OE are attached to the structures around the obturator foramen and their tendons pass across the capsule of the hip joint posteroinferiorly, and insert onto the medial surface of the greater trochanter. The action of the OI as an external rotator of the extended thigh and an abductor of the flexed thigh would explain the painful range of motion in patients with pyomyositis of the OI [5]. OI and OE abscess may

cause pressure on the obturator or sciatic nerve, leading to radiating pain of the thigh, knee, or leg [2].

The location of the obturator muscles makes clinical diagnosis difficult and requires exhaustive repeated clinical examination and investigation with a high level of suspicion [10]. We report this case to draw the attention to this rare condition and to emphasize the importance of its early diagnosis to avoid unnecessary surgical intervention and the potentially severe sequelae.

Case Report

A 9-year-old boy was presented to our emergency department complaining of left hip pain, fever, malaise, and inability to bear weight. The pain was described as "dull" and was aggravated by movement and only partially relieved by rest and analgesics. The condition started 14 days prior to presenting at our department with left knee pain and limping. The fever was intermittent. There was a history of sore throat 5 days before the knee pain, for which the patient received an incomplete course of antibiotics. Ten days later, the knee pain progressed to an increasing pain that started in the left groin, with inability to bear weight. At this stage of the disease the patient presented to our hospital. The patient denied any history of trauma.

On examination the patient was unwell, with a temperature of 39.5°C. No other focus of infection was identified on examination. His left hip was held in flexion, abduction, and external rotation. His hip and thigh were tender, but there was no erythema or swelling. Active movements were more painful than passive movements. The painful limitation of all ranges of motion suggested an acute hip problem.

Laboratory investigations at the time of presentation showed evidence of acute infectious process. Leukocytic count was 22 300/mm³, erythrocyte sedimentation rate (ESR) was 135 mm/h, and C-reactive protein was 4.6 mg/dl. The patient's blood culture grew *Staphylococcus aureus* sensitive to oxacillin, cefoxitin, and gentamicin.

The plain radiograph of the hip and pelvis revealed normal contour, joint space, and alignment, with no soft-tissue abnormalities, but the left hip was in abduction, flexion, and external rotation (Figure 1). Ultrasound of both hips revealed mild left hip effusion.

The clinical picture and laboratory investigations were strongly suggestive of septic arthritis of the hip, and the patient had signs of sepsis that needed urgent intervention. An informed consent for urgent arthrotomy and drainage was obtained after discussion with the family.



Figure 1. AP radiograph of the pelvis at presentation showing the left hip joint in external rotation, abduction and mild flexion.

Operative findings

Under general anesthesia and with the help of an image intensifier, aspiration of the hip was performed, which showed blood-stained fluid that was sent for gram staining, culture, and sensitivity testing. The decision was made to proceed with open drainage of the hip through an anterior approach. Exploration of the left hip revealed no pus and the synovial membrane was normal. Samples of synovial fluid and synovium were taken for gram staining, culture, sensitivity testing, and for histopathological examination; the results were normal.

Postoperative course

Antibiotics were started in the operating room – intravenous cefoxitin 100 mg/kg /day in 4 divided doses and cloxacillin 100 mg/kg/day in 4 divided doses – after taking the necessary samples. Skin traction was applied to the left hip.

MRI (Figures 2 and 3) was requested to rule out osteomyelitis of the proximal femur, pelvic collection, or sacroiliitis. It revealed lobulated collection, of low attenuation, with enhancing rim at the left side of the pelvis involving the left OI and extending to the left OE muscles, as well as small abscesses seen in the left vastus intermedius and vastus lateralis muscles. MRI findings confirmed the diagnosis of pyomyositis of the left OI extending to the OE.

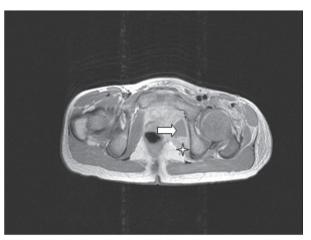


Figure 2. T1 weighted image MRI, with IV gadolinium of the pelvis at the level of the greater trochanter showing edematous changes in the obturator internus muscle (star) along with central necrosis (arrow) with ring enhancement suggestive of pyomyositis



Figure 3. T1 weighted image MRI, with IV gadolinium of the pelvis at the level of the lesser trochanter showing edematous changes in the obturator externus muscle (star) along with central necrosis (arrow) suggestive of pyomyositis.

Treatment was continued in the form of antibiotics and skin traction. The patient showed steady improvement and his temperature returned to normal within 48 hours, with gradual improvement of the hip pain. After 3 days the patient regained painless movement of the left hip and he was walking with a slight limp. The patient was discharged from the hospital in good general condition, with full weight-bearing and without limping after completing a 1-week course of intravenous antibiotics. When C-reactive protein returned to normal, oral cloxacillin, 500 mg every 6 hours was started and continued for 2 weeks. The patient was followed in our outpatient clinic for 2 years without any complaints. At the final followup the patient was fully active and had full range of motion.

Discussion

Most patients with OI pyomyositis present with fever, pain in the hip, thigh or abdomen, limping, and inability to bear weight [3]. Thigh pain is a rare symptom that has been described in cases of infection of the OI [4,11]. Our patient had initial knee pain (to the best of our knowledge, a presentation of pyomyositis of the OI and OE has never before been reported in the available English literature) because the posterior division of the obturator nerve pierces the OE muscle, which was affected in our case, and because the terminal branch of the obturator nerve supplies the knee joint [12].

Septic arthritis of the hip should always be excluded and proximal femoral osteomyelitis, pelvic osteomyelitis, or psoas abscess should also be considered in the working diagnosis [5]. Pyomyositis can also mimic a malignant tumor, thrombophlebitis, muscle rupture, or obturator hernia [13,14].

Children who present with symptoms and signs suggesting hip effusion or musculoskeletal sepsis in the region of the hip should have an ultrasound of the affected hip. A negative ultrasound in the presence of signs of sepsis is an indication for MRI [4,15]. MRI is an increasingly valuable tool for evaluating musculoskeletal infection, with reporting sensitivity ranging from 88% to 100%, specificity from 75% to 100%, and a positive predictive value of 100% [3,16]. It allows visualization in several planes and can reveal diffuse inflammation in early stages of the infection [7]. Trusen et al. recommended that MRI should be the imaging method of choice for the pelvis whenever possible [17]. Similar to our case, the diagnosis of all reported cases of OI and OE pyomyositis was confirmed using CT and/or MRI.

Kan et al. studied 130 children with suspected musculoskeletal infection; 34 patients (study group) underwent an MRI after diagnostic or therapeutic intervention and 96 patients (control group) had an MRI prior to any procedure [18]. The results showed that about 60% of patients had neither septic arthritis

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nor osteomyelitis, suggesting that the majority of patients in the study group had a diagnostic or surgical procedure that could have been avoided with early MRI evaluation. This was also true in our patient, because if MRI had been done before the surgical intervention, the surgery would have been avoided.

Karmazyn et al., in their review of patients with non-traumatic acute hip pain, ESR of more than 30 mm/h, and no evidence of septic hip suggested that pelvic MRI should be performed to rule out pelvic osteomyelitis or pyomyositis [15].

Medical management with 3 to 6 weeks of antibiotic therapy is usually successful in treating OI and OE pyomyositis [1]. Failure of the patient to improve with adequate and appropriate antibiotic treatment is an indication for intervention in the form of ultrasound- or CT-guided percutaneous drainage, which can be helpful diagnostically and therapeutically [1,3,10,19]. If this is inadequate, drainage should be performed with open surgery [1,20,21]. With the right treatment, pyomyositis resolves without sequelae, as in the case presented in this study [3,22]. However, if not treated properly, it can relapse or progress towards acute compartmental syndrome, solid-organ impairment, and even septic shock and death [21,23].

Conclusions

In conclusion, in children who present with symptoms of hip effusion or musculoskeletal sepsis in the hip region, septic arthritis of the hip should always be excluded and pyomyositis should also be considered in the working diagnosis. MRI is a very important diagnostic tool to identify the cause of hip pain in febrile patients. Most cases of OI and OE pyomyositis respond to conservative management.

List of abbreviations

OI – Obturator Internus; **OE** – Obturator Externus; **MRI** – Magnetic resonance imaging; **CT** – Computerized tomography.

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