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Case report/Kazuistyka

# Pyomyositis in the course of acute lymphoblastic leukemia

## *Ropne zapalenie mięśni szkieletowych w przebiegu ostrej białaczki limfoblastycznej*

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ABSTRACT

Pyomyositis is a term used to asses pyogenic infection of the skeletal muscle and develops as the result of bacteriemia and occurs most commonly in patients with various immunosuppressive diseases. Pyomyositis should be considered in the differential diagnosis in patients complaining of intensive muscle pain and fever. The usual causative organism is *Staphylococcus aureus*. We present a 3-year-old boy with acute lymphoblastic leukemia (ALL) and pyomyositis caused by Pseudomonas aeruginosa diagnosed in the course of induction therapy. The diagnosis of pyomyositis in the crural muscles of both legs was given based on imaging examinations: ultrasonography and magnetic resonance.



## Introduction

Pyomyositis is a term used to asses pyogenic infection of the skeletal muscle. Pyomyositis develops as the result of bacteriemia and occurs most commonly in patients with various immunosuppressive diseases. The usual causative organism is *Staphylococcus aureus*. Other pathogens that may cause pyomyositis are coagulase-negative *Staphylococcus species*, various streptococcal species, Gram-negative bacteria and fungi. Pyomyositis should be considered in the differential diagnosis in patients complaining of intensive muscle pain and fever. Routine laboratory investigations are nonspecific, and the diagnosis rests on imaging modalities [1–6].

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Magnetic resonance scan is the most useful investigation in the diagnosis and can pick up early changes in the muscle [7, 8]. Treatment involves appropriate antibiotic therapy with or without drainage, according to local conditions [1–6]. We present 3 years old boy with acute lymphoblastic leukemia and pyomyositis caused by *Pseudomonas aeruginosa* diagnosed in the course of induction therapy.

## Case report

A 3-year-old boy diagnosed with acute lymphoblastic leukaemia (ALL), pre B type, SRG (standard risk group) treated in accordance with ALLIC BFM 2002 Protocol (Acute Lymphoblastic Leukemia Inter-Continental Berlin-Frankfurt-Munster). We observed a good response to the treatment. Because of the observed fever and diarrhea in the course of neutropenia, the chemotherapy was halted on the 18th day of induction therapy. The cultures of blood and stools were negative. The antibiotic cefepim was administered and was continued until the 10th day. On the 3rd day after the completion of the antibiotic treatment the child complained of intense pain in the lower limbs, especially in the crura. In the physical examination there was only a pain in the crura during palpation. Laboratory tests revealed a high value of ESR (erythrocytes sedimentation rate; 180 mm/h), C-reactive protein (CRP; 177 mg/l) and creatine kinase (CK; 788.0 U/l). Tests for the presence of myoglobin in urine came back negative which excluded rhabdomyolysis. Typical bacteriological tests, including blood culture came back negative. On the 6th day after the patient's notification of pain an edema and redness of the calves appeared. Due to the pain the child was not able to walk despite the administration of analgesics. The ultrasonography (USG) examination revealed significant extension of contours and intensified echogenicity of the subcutaneous tissue and crural muscles which suggested an inflammatory process. A broad-spectrum antibiotic therapy (imipenem, teicoplanin) and an antifungal therapy (amphotericin B) were introduced.

After a 10-day therapy the edema, redness of crura and pain on palpation reduced. Parameters of the inflammatory status improved significantly (ESR 54 mm/h, CRP 10.1 mg/l) and the controlled level of CK was normal (72 U/l). It was decided to resume the induction chemotherapy while continuing the antibiotic therapy. 3 weeks after the occurrence of the ailments the pain in the crura intensified again and a physical examination revealed an increased warmth and edema. Another USG examination showed a two-sided elevated echogenicity and fluid between the crural muscles. A magnetic resonance (MRI) revealed two excessive fluid areas of 2.2 × 4.3 cm and 2.4 × 5.7 cm in the area of the left crural muscles. Similar lesions of 4.1 × 2.3 cm and 4.3 × 1.4 cm were observed in the right crus (Fig. 1, Fig. 2). The lesions observed in the crura were consistent with pyomyositis. The fluid areas were drained and pus was extruded. The culture revealed *P. aeruginosa*. The crural pains retreated completely upon the incision and drainage of the lesions. A 14-day antibiotic therapy compliant with the antibiogram (tazobactam/piperacyllin) was introduced and the drainage of the crural soft tissues was maintained

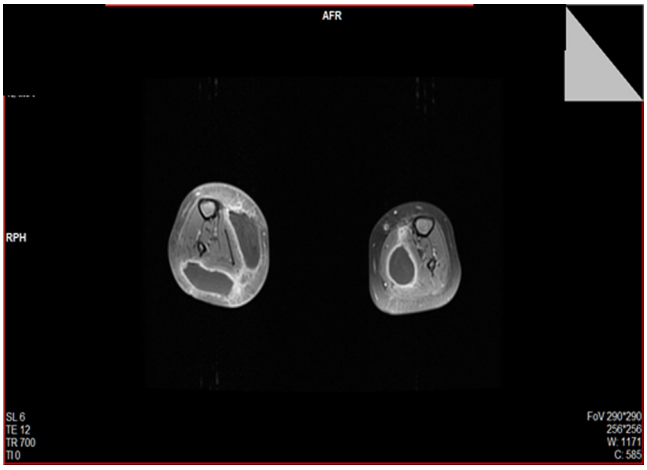


**Fig. 1 – The MRI images – pyomyositis areas in the crural muscles, cross-section**
***Ryc. 1 – Obraz MRI – ropne zapalenie mięśni podudzi, przekrój***

for 7 days. The clinical condition allowed for a continuation of chemotherapy. The follow-up USG of the crura performed on the 7th day after the surgery did not show fluid reservoirs. On the 3rd day after the completion of the antibiotic therapy a massive pneumonia was diagnosed. The child rapidly developed respiratory insufficiency and required treatment at the Intensive Care Unit. After 4 days the child was admitted to our department in an improving condition. The antibiotic therapy continued until the 14th day. The patient underwent a rehabilitation of the lower extremities. The clinical condition of the child improved completely which enabled the completion of the chemotherapy. The boy feels well and the motor activity of his lower extremities is normal.

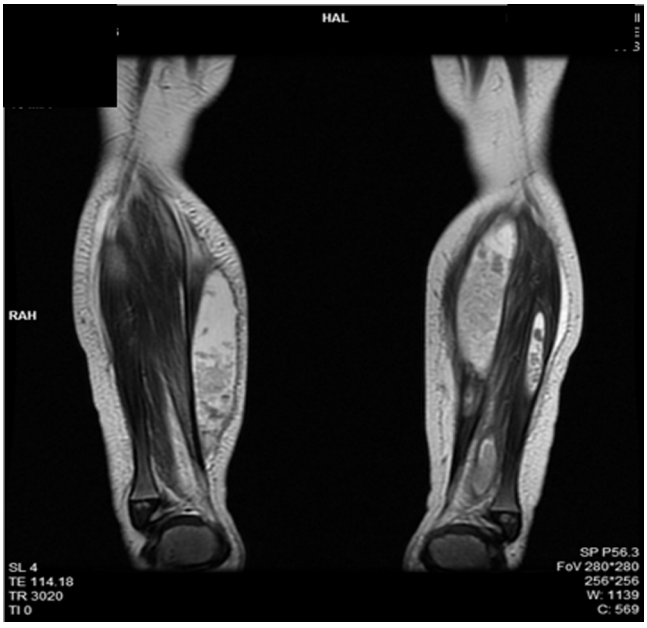


**Fig. 2 – The MRI Images – pyomyositis areas in the crural muscles**
***Ryc. 2 – Obraz MRI – ropne zapalenie mięśni podudzi***

## Discussion

Pyomyositis is an endemic disease commonly diagnosed in tropical regions; hence the name ''tropical pyomyositis''. Reports of cases from temperate climates are seldom and concentrate on immunocompromised patients or patients with underlying chronic disease [1–6, 9–11]. Pyomyositis has three stages, from diffuse inflammation to focal abscess formation and to a septic state. It begins with the insidious onset of progressive pain with low grade fever and muscle ache. In second stage muscle abscess formations are organized and in the next stage the manifestations of sepsis may be observed [1–3]. Due to non-specific symptoms the diagnosis is rarely given in the early stage of the disease. The differential diagnosis should include above others venous thrombosis, osteomyelitis, intramuscular haematoma and rhabdomyolysis and in the case of a situation when the process in localised in the iliopsoas muscle, also appendicitis [1–6]. In the case of patients with leukaemia, treated with cytostatic drugs, as with our patient, initial symptoms suggested drug-induced toxicity above all after steroids and vincristine [9–11]. The laboratory tests results are non-specific for the course of pyomyositis, which may indicate moderate leucocytosis with a left shift, elevated ESR and normal muscle enzyme levels [1–6]. USG examinations may reveal pyomyositis, but if the USG picture is inconclusive MRI is the most useful imaging technique for diagnosis. Because of its availability, a USG examination remains very helpful in monitoring those patients [7, 8]. The treatment depends on the stage of the disease in which it was diagnosed. An early stage of pyomyositis, the so called diffuse inflammatory infiltration of the muscle is treated exclusively with antibiotics, involving antibiotics recommended for the treatment of infections caused by *S. aureus*, such as vancomycin or teicoplanin. In the case of an abscess, apart from antibiotic therapy, it is also important to incise and drain it. According to the authors of publication on this pyomyositis, therapeutic success is conditional on early diagnosis, the use of a relevant antibiotic and surgical intervention [1–6, 9–11]. What probably could have contributed to a more complicated course of pyomyositis in the presented case, was his underlying disease – leukaemia. As a result of both the disease and the applied chemotherapy, significant immunological deficits are observed, which in the relevant literature is considered as a significant risk factor. Another important risk factor for a severe course of the disease was is an infection caused by the aggressive Pseudomonas aeruginosa pathogen [9–11]. A similar clinical course of pyomyositis in a 5 year-old boy but of a typical *S. aureus* etiology was presented by Taksande et al. [1].

Pyomyositis is sporadically diagnosed in children which is confirmed by only a few publications on this matter. Publications on pyomyositis concerning oncological children, including those with leukaemia are sparse. A considerably greater number of publications concern adult patients. Falagas et al. in their study presenting a case of an adult woman with pyomyositis of the iliopsoas muscle in the course of the Hodgkin's lymphoma reviewed the literature on the subject of the occurrence of this disease in patients with haematological malignancies and analysed 44 cases including only 4 children [12]. Blat et al. presented the cases of 2 boys with acute lymphoblastic leukaemia, who in the course of remission-inducing therapy, during neutropenia, developed multifocal pyomyositis, similarly to our patient [9]. Also Corden et al. described in their publication cases of 2 boys with ALL and pyomyositis diagnosed during the induction of the remission [10]. Kao et al. reported on a case of a 10-year-old girl treated for ALL in whom pyomyositis localised in the thoracic muscles and thigh was diagnosed during the remission-inducing therapy. The antibiotic therapy (vancomycin) corresponding with the results of the culture of the pus content (*S. aureus*) and surgical intervention were completely effective [10]. According to the available literature, so far 5 cases of children with acute lymphoblastic leukaemia and pyomyositis during the induction therapy have been published. Our case report seems to be the 6th one following a similar clinical suit [9–11]. In all the patients the course of the disease in its initial stage was non-characteristic and suggested drug-induced toxicity. In the cases of children with all published to date, *S. aureus* was in each case the cause of pyomyositis. The pathogen responsible for the disease in our case was *P. aeruginosa*, an etiological factor so far not published in the group of cases of children with ALL and pyomyositis. These bacteria may be the reason for the more complicated course of pyomyositis in our patient.

To summarize, in children with acute lymphoblastic leukaemia, local muscle pains during the induction therapy require in the differential diagnosis the consideration of pyomyositis and instant imaging examination (MRI) of the affected area. The use of a broad-spectrum antibiotic therapy with possible surgical intervention is also of a crucial importance.

## Authors' contributions/Wkład autorów

GS-M – study design, data collection and interpretation, manuscript preparation. TK, HW – study design. AM-M – data collection and interpretation, manuscript preparation, literature search. KM – literature search.

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## Conflict of interest/Konflikt interesu

None declared.

## Ethics/Etyka

The work described in this article have been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; EU Directive 2010/63/EU for animal experiments; Uniform Requirements for manuscripts submitted to Biomedical journals.

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