
Rotator Cuff Pyomyositis in Sickler Pediatric Caused by Salmonella Species: A Case Report

Mohammed W. Tolah^{1*}, Amgad Affi¹, Housen K. Albalwi¹, Ayman Zein¹ and Musab A. Hijan²

¹Orthopedic Surgery, King Salman Armed Forces Hospital, Tabuk, Saudi Arabia

²Orthopedic Surgery, King Salman Armed Forces Hospital, Northwestern Region, Tabuk, Saudi Arabia

***Corresponding author:** Mohammed W. Tolah, Orthopedic Surgery, King Salman Armed Forces Hospital, Tabuk, Saudi Arabia. E-mail: mw.tolah@hotmail.com

Received: October 02, 2024; **Accepted:** October 26, 2024; **Published:** November 15, 2024

Abstract

This case report presents the case of a 1-year-old female with sickle cell anemia who developed pyomyositis due to Infection with Salmonella species. The clinical presentation of the patient included jaundice, high fever, and pain in the left shoulder, which progressed to redness, swelling, and limitations in movement. An MRI scan was conducted on the patient, revealing fluid loculations in the shoulder muscles. A surgery was performed on the patient. Post-operative care included antibiotics, leading to significant clinical improvement. This report highlights the importance of early diagnosis and the appropriate management of pyomyositis in patients with underlying conditions, such as sickle cell anemia.

Keywords: Intramuscular abscess; Salmonella species; Sickle cell anemia; Rotator cuff pyomyositis

Introduction

Pyomyositis is a rare and treatable infection caused by bacteria. It affects skeletal muscles. Pyomyositis usually leads to an abscess, a swollen area containing pus in the muscles [1]. The infection commonly occurs in the large muscle group of the legs but can also occur in other muscles. Most cases of pyomyositis are caused by Staphylococcus aureus. The disease can affect people of any age, with about 35% of all cases occurring in children [2]. Pyomyositis usually results from the hematogenous spread of bacteria. The incidence of pyomyositis is higher in patients who have underlying medical conditions, such as human immunodeficiency virus (HIV), sickle cell anemia (SCA), malignancy, and diabetes mellitus. Infections by Salmonella spp. have been reported in these patients [3]. Intramuscular abscesses of the rotator cuff, including those that involve the pericapsular space, are uncommon. When they occur, their clinical manifestations resemble those of shoulder septic arthritis, a significant cause of diagnosis and treatment delays because such cases are rare [4]. Sickle cell disease is an autosomal recessive hereditary disorder of the hemoglobin. It is characterized by a mutation in the sixth codon on the hemoglobin beta (β) chain. Individuals with sickle cell disease often face different infections, especially as children [5]. These infections can include osteomyelitis, septic arthritis, and pyomyositis on rare occasions.

This report describes the case of a pediatric patient with sickle cell anemia who developed pyomyositis, which led to a periscapular intramuscular abscess. The patient was managed through surgical irrigation and debridement. An intraoperative culture showed that an infection with *Salmonella* spp was the cause of the patient's pyomyositis. Antibiotic treatment significantly improved the patient's clinical condition. Follow-up revealed an excellent outcome with no further complications.

Case Presentation

Patient Information

A one-year-old female, diagnosed with sickle cell anemia (SCA) at nine months, presented to the emergency department with a high fever and jaundice that had lasted for three days. The fever was intermittent, and management with paracetamol relieved it. The recorded temperature was 38°C. The jaundice had worsened progressively for three days, and the patient complained of a limitation in the movement of the upper left extremity with tenderness. These symptoms started suddenly without any history of trauma. The patient also demonstrated decreased levels of physical activity and poor oral intake.

Medical History

The patient had a medical history of sickle cell anemia, which was diagnosed at nine months of age. She is currently on folic acid medication. The patient has no previous history of hospital admissions or surgeries. According to the family history, the patient's two parents are carriers of sickle cell anemia, and two of her six siblings are also carriers. The patient has no known allergies.

Physical Examination

Upon physical examination, the patient was found to be stable, alert, and oriented. The results of the physical examination are presented in Table 1 below.

Table 1: The results of the physical examination of the patient.

Parameter	Results
Vital Signs	
Heart Rate	118 beats per minute.
Respiratory Rate	22 breaths per minute.
Blood Pressure	113/66 mmHg.
Oxygen Saturation	97% on room air.
Temperature	38°C.
General Appearance	Ill, jaundiced, pallor, not in distress.
Chest Examination	Clear lung fields, good bilateral entry of air.
Cardiovascular Examination	Audible S1 and S2, no additional sounds or murmurs.
Abdominal Examination	Soft and lax with no splenomegaly.
Neurological Examination	Glasgow Coma Scale (GCS): 15/15, unremarkable CNS examination
Left Shoulder Examination	No visible redness, hotness, or swelling. Swelling at the posterior part limits the passive and active range of motion, especially in external rotation and abduction. The distal neurovascular status was intact.
Other findings	The patient has no respiratory distress, rash, or enlarged lymph nodes. Other examinations reveal no remarkable findings.

Laboratory Investigations

Table 2: The laboratory investigations yielded the results.

Test	Results
White Blood Cell (WBC) Count	10.50 cells per mm ³
Hemoglobin (Hgb)	6.7 grams per deliliter (g/dL)
Platelet Count	214 k/ μ L
Reticulocyte Count	3%
C-reactive protein (CRP)	22 mg/dL
Erythrocyte Sedimentation Rate (ESR)	99 mm/hr

Initial Management

The patient was admitted to a pediatric ward with a working sickle cell anemia diagnosis complicated by fever, a possible osteoarticular infection, and a potential sickle cell crisis. The patient received intravenous cefoxamine and a blood transfusion to address low hemoglobin levels. A blood culture revealed the patient had a Salmonella infection.

The patient's general condition improved on the second day of admission. However, the left shoulder condition worsened, showing redness, swelling, and hotness, with a limitation of movement. There was also mild swelling, movement limitation, hotness, and redness on the left thumb. Besides, the patient had an intact distal neurovascular status on the left upper extremity. Orthopedic consultation was sought to address these developments.

Imaging Studies

An x-ray of the left shoulder showed unremarkable results, with mild soft tissue swelling. It is shown in Figure 1 AP and lateral Scapular Y View in Figure 2 below.

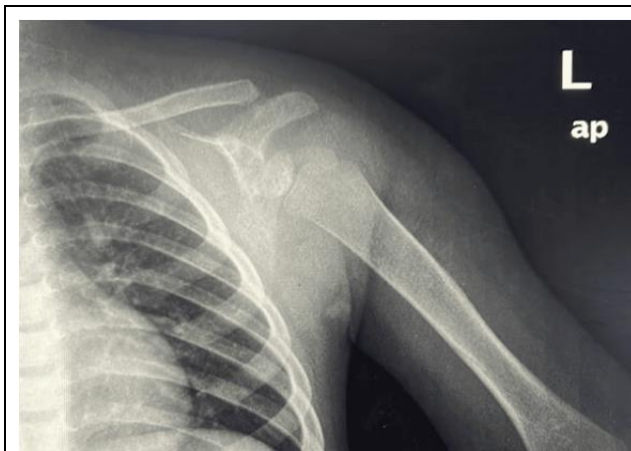


Figure 1: Left shoulder Anteroposterior radiograph revealed normal bony signal of the left shoulder joint, clavicle, scapula, and left proximal humerus.

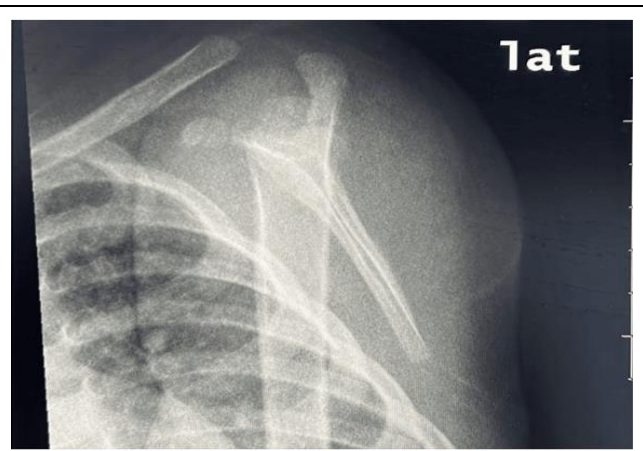


Figure 2: Left shoulder lateral scapula radiograph view revealed normal bony signals of the shoulder, scapula, clavicle, and proximal humerus.

An MRI of the left shoulder showed three fluid loculations in the supraspinatus, infraspinatus, and subscapularis muscles with cellulitis and myositis. The measurements of the fluid loculations were 1.7 x 1.3 x 0.96 cm, 3.0 x 1.8 x 1.9 cm, and 2.1 x 1.2 x 1.7 cm, respectively. The fluid loculations along the infraspinatus and the subscapularis appeared to be connected along the medial border of the scapula. The MRI also revealed mild left glenohumeral joint effusion, left axillary lymphadenopathy, and no gross bone enhancement. The MRI is shown in Figures 3 and 4 below.

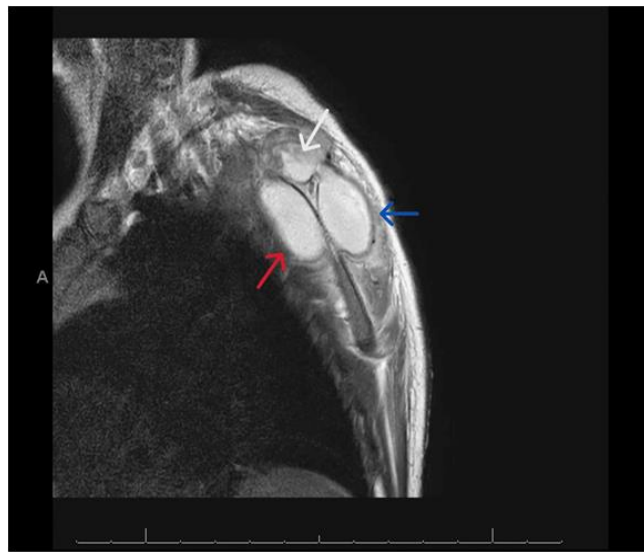


Figure 3: Left-shoulder MRI T2 Sagittal cut with contrast revealed three fluid loculations in the subscapularis (red arrow), supraspinatus (white arrow), and infraspinatus (blue arrow) muscles.

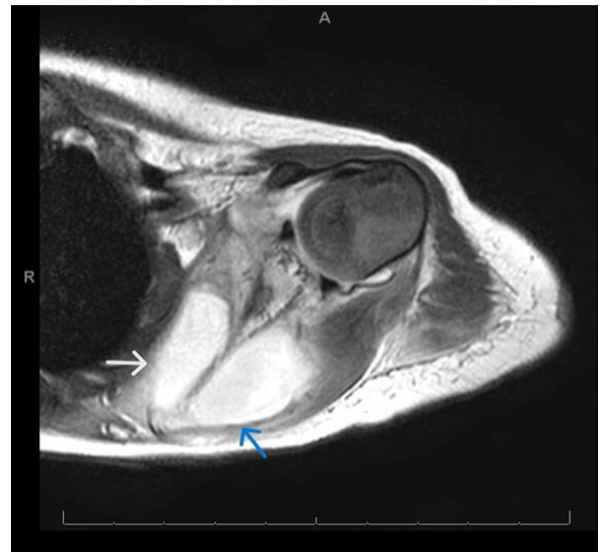


Figure 4: Left-shoulder MRI T2 Axial cut with contrast showed fluid loculations in the infraspinatus (blue arrow) and subscapularis (white arrow) muscles. The fluid loculations along the infraspinatus and the subscapularis appeared to be connected along the medial border of the scapula.

Surgical Procedure

The patient was placed in the right lateral decubitus position during the surgery. Prepping and draping were done under sterile conditions using a beanbag and an axillary roll. A limited 5 cm vertical skin incision was made just above the medial end of the spine of the scapula [6]. Superficial dissection was done with good hemostasis, reaching the medial scapular spine bone. The fascia was incised over infraspinatus and supraspinatus. A deep incision was also performed, reaching the collection of pus in the infraspinatus, supraspinatus, and subscapularis muscles. The abscesses were evacuated, and cultures were taken and sent to the laboratory for analysis. Normal saline was used for copious irrigation to clean the infected area thoroughly. A drain was applied to allow for the continued drainage of any recurrent or remaining pus. The wound was then closed in layers, and a sterile dressing was used.

Post-operative Care

The patient was started on Cefdinir, tailored to the culture results, which identified *Salmonella* spp. as the causative agent. There was notable clinical improvement following the operation. The patient became afebrile and showed increased activity and movement in the affected shoulder. Besides, inflammatory markers indicated significant progress, with a drop in (ESR) from 99 to 56 mm\hour. The patient was discharged and allowed to go home in good condition. Instructions were given to continue oral antibiotics for six weeks. Follow-up visits were scheduled to monitor recovery and ensure the infection did not recur.

Follow-Up

Two weeks after discharge, the patient returned to the clinic for a wound checkup and the removal of sutures. The wound was clean and dry and showed no signs of infection. The patient was afebrile and showed good movement of the shoulder. Six weeks after discharge, the patient showed a full shoulder range of motion, both active and passive. She remained afebrile, and her general health was good. At eighteen months, the patient was active with typical vital signs. The surgical wound had completely healed and showed no signs of infection, as shown in Figure 5 below. The shoulder showed a full range of motion without any limitations.



Discussion

Pathophysiology of Pyomyositis

Pyomyositis is a bacterial infection of the skeletal muscles mainly caused by *Staphylococcus aureus* [2]. However, patients with underlying conditions, such as sickle cell anemia, can have the infection caused by *Salmonella* species [7]. Generally, the infection results from the hematogenous spread of the bacteria and muscle abscess. The condition is more common in tropical climates but can also occur in temperate climates [8].

Sickle Cell Anemia and Susceptibility to Infections

Pyomyositis can be a rare complication of sickle cell anemia [9]. Sickle cell anemia is characterized by hemoglobin S, which can polymerize under low oxygen conditions, causing the red blood cells to assume a sickle shape. People with sickle cell disease have at least one beta-globin subunit replaced by hemoglobin S. In people with sickle cell anemia, both beta-globin subunits are replaced by hemoglobin S [9]. The sickle-shaped red blood cells are less deformable, rigid, and prone to hemolysis. Since they cannot flow smoothly through the blood vessels, they cause vaso-occlusion, leading to tissue infarction and ischemia [10]. Tissue infarction causes pain, increased infection susceptibility, and organ damage [10]. In sickle cell anemia patients, the spleen, which filters bacteria, becomes fibrotic and non-functional due to repeated infarctions. The resultant asplenia increases the susceptibility of these patients to infections, especially from encapsulated organisms, such as *Salmonella* [8].

Clinical Presentation and Diagnostic Challenges

The clinical presentation of pyomyositis can be non-specific and often resembles other conditions, such as osteomyelitis, thrombophlebitis, hematoma, malignancy, muscle strain, and arthritis [11]. It is associated with severe complications, such as sepsis and compartment syndrome, which may lead to death in the case of diagnosis and treatment delays [11]. The initial symptoms of the condition include fever, swelling, muscle pain, and tenderness of the muscle involved. It can also lead to leukocytosis. It mainly affects the muscles of the thigh, the calf muscles, the latissimus dorsi, the pectoral muscles, the gluteus muscles, and the iliopsoas [11]. In the case presented in this report, the patient presented with shoulder pain, jaundice, and fever, which progressed to swelling and redness. The involvement of different groups of muscles and the progression of symptoms showed that the infection was severe.

Imaging and Diagnosis

Imaging studies are essential for pyomyositis to be accurately diagnosed. X-rays reveal swelling of the soft tissues, while MRI provides a more comprehensive diagnosis that shows collections of fluids, muscle edema, and the extent of the infection [12]. In the case presented in this report, MRI was essential in identifying fluid loculations within the supraspinatus, infraspinatus, and subscapularis muscles, which helped confirm the pyomyositis diagnosis.

Management and Treatment

Pyomyositis management involves surgical intervention to drain abscesses and appropriate antibiotic therapy. In this case, surgical irrigation and debridement were performed, followed by intravenous antibiotics tailored to the culture results. The choice of antibiotics to be used should cover the most common pathogens [13]. In a case involving *Salmonella* spp., antibiotics, such as Cefnidir, are effective. Post-operative care is essential to prevent the recurrence of pyomyositis and ensure complete

recovery. The patient showed significant improvement following surgery, with decreased inflammatory markers and no signs of infection during follow-up visits [13]. Long-term follow-up is essential to monitor for late complications and ensure complete functional recovery.

Differential diagnosis

Differential diagnosis is essential for patients who present with symptoms that are similar to those of pyomyositis. These should include deep vein thrombosis, osteomyelitis, and septic arthritis. These conditions can present with symptoms resembling pyomyositis, including swelling, pain, and systemic signs of inflammation or infection [14]. MRI scans are essential in differentiating these conditions because they have a high resolution and can provide detailed images of joints, bone marrow, and soft tissues.

Implications for Clinical Practice

This case shows that early diagnosis and timely management of pyomyositis is essential, especially in patients with underlying conditions, such as sickle cell anemia. Clinicians should maintain a high index of suspicion for musculoskeletal infections in such patients, especially when they present with acute pain in the extremities [9]. Early interventions can improve outcomes by preventing complications.

Conclusion

This case report highlights a rare but significant complication of sickle cell anemia: pyomyositis caused by *Salmonella* species. The patient is one year old and presented to the clinic with high fever, jaundice, and shoulder pain, which progressed rapidly to severe symptoms that required surgical intervention. The case shows that maintaining a high index of suspicion for pyomyositis in patients with underlying conditions, such as sickle cell anemia, is essential, especially when they present with acute musculoskeletal pain. Early diagnosis through advanced imaging techniques, such as MRI, and timely surgical and antibiotic interventions helped achieve a good outcome for the patient. The successful management of the case using surgical drainage and targeted antibiotic therapy resulted in significant clinical improvements and, eventually, full recovery, as seen in follow-up assessments that showed the infection did not recur and that shoulder function was fully restored. Clinicians should watch out for symptoms of pyomyositis in patients presenting similar symptoms. This case shows a need for thorough post-operative care through monitoring to ensure full recovery and prevent recurrence.

REFERENCES

1. Radcliffe C, Gisriel S, Niu YS, et al. Pyomyositis and infectious myositis: A comprehensive, single-center retrospective study. *Open Forum Infectious Diseases*. 2021; 8:10.
2. Faustino FDM, Batista F. Infectious pyomyositis with intramuscular abscess in a healthy adult. *Cureus*. 2021; 13:10.
3. Kenar G, Avcı AK, Balcı A, et al. Pyomyositis and muscle abscess due to salmonella enteritidis in a patient with behçet's syndrome: A case report. *Clinical and Experimental Rheumatology*. 2020; 38: 98-100.
4. East J, Piper D, Chan S. Spontaneous intramuscular abscesses involving the rotator cuff muscles in two cases presenting during the COVID-19 pandemic. *Cereus*. 2020; 12: 10.
5. Inusa B, Hsu L, Kohli N, et al. Sickle cell disease-genetics, pathophysiology, clinical presentation and treatment. *International Journal of Neonatal Screening*. 2019; 5: 20.

6. Posterior approach to the scapular body. (2024). Accessed: 2024.
7. Chiu NC, Hsieh MC, Chi H, et al. Clinical characteristics of pyomyositis in children: 20-year experience in a medical center in Taiwan. 2009; 42: 494-499.
8. Brousse V, Buffet P, Rees D. The spleen and sickle cell disease: The sick(led) spleen. *British Journal of Haematology.* 2014; 166: 165-176.
9. Wong VK, Lissack ME, Turmezei TD, et al. Salmonella pyomyositis complicating sickle cell anemia: A case report. *Journal of Medical Case Reports.* 2010; 4: 10-1186.
10. Elendu C, Amaechi DC, Alakwe-Ojimba CE, et al. Understanding sickle cell disease: Causes, symptoms, and treatment options. *Medicine.* 2023; 102: 35237.
11. Ngor C, Hall L, Dean J, et al. Factors associated with pyomyositis: A systematic review and meta-analysis. *Tropical Medicine & International Health.* 2021; 26: 1210-1219.
12. Weaver JS, Omar I, Mar W, et al. Magnetic resonance imaging of musculoskeletal infections. *Polish Journal of Radiology.* 2022; 87: 141-162.
13. Dhivakaran G. An unresolving case of pyomyositis: A case report. *Malaysian Orthopaedic Journal.* 2023; 17: 70-75.
14. Spinnato P, Patel DB, Di Carlo M, et al. Imaging of Musculoskeletal Soft-Tissue Infections in Clinical Practice: A Comprehensive Updated Review. 2022; 10: 2329.