Pyomyositis and muscle abscess due to *Salmonella enteritidis* in a patient with Behçet's syndrome: a case report

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ABSTRACT

Salmonella infections usually present with gastrointestinal manifestations including enterocolitis especially in immunocompromised patients. Haematogenous dissemination and abscesses are very rare complications of Salmonella species. This case report documents a patient with Behçet's syndrome (BS) who has pyomyositis due to Salmonella species. A 43-year-old male patient with BS presented to the outpatient rheumatology clinic with bilateral acute-onset lower extremity pain. However, over a short time the pain gradually increased and was accompanied by fever. The magnetic resonance scans demonstrated pyomyositis and muscle abscess in the adductor and obturator muscles. The cultures showed Salmonella enteritidis *infection. The patient was successfully treated with antibiotic therapy.*

This case is important since it is one of the first in the literature to report an adult patient with BS and Salmonella pyomyositis.

Introduction

Pyomyositis is a very rare infection that arises after transient bacteremia, most often affecting the muscle groups of the pelvic girdle and lower extremities (1). Pyomyositis due to *Salmonella* species has been reported in patients with diabetes mellitus (DM), cancer, sickle cell anaemia, and human deficiency virus (HIV) infections (2). Herein, we report on a patient with Behçet's syndrome (BS) who has pyomyositis due to *Salmonella* species.

Case report

A 43-year-old male presented at the outpatient rheumatology clinic with acute-onset pain in both thighs. When the patient was evaluated, he appeared painful, with a blood pressure of 150/100 mm/Hg, a pulse rate of 100/minute and a low-grade fever of 37.8 C degree. He stated that he had difficulty in getting up from a sitting position, and the pain increased while walking. Diameter measurements of both lower extremities were similar. There was a decrease in muscle strength in the proximal lower extremities. New-onset erythema nodosum lesions were observed on the anterior face of the patients' tibia.

In his previous medical history, he had been diagnosed with BS in 2007 with posterior uveitis, recurrent oral aphthous ulcers, arthritis, erythema nodosum, and positive pathergy test. Immunosuppressive treatment was administered for the first five years of the disease with azathioprine (AZA) and corticosteroids (CS), then the patient reported that he had been diagnosed with pulmonary tuberculosis (tbc). He discontinued the immunosuppressive treatment and was treated with anti-tuberculosis agents for a year. After remission of the tbc, he did not present at the hospital for four years. Then he had a new attack of posterior uveitis for which AZA, CSs and cyclosporine treatments were started. It was learned that CS had been added at the dose of 1 mg/kg/day two months ago due to a relapse of posterior uveitis. The dose was gradually decreased, but he was still receiving 20 mg/day. He had no history of anatomic and functional asplenia and hyposplenism such as splenectomy, congenital asplenia, infarction, infiltration, sickle cell anaemia, thalassemias, chronic liver diseases, HIV or malignancies.

The preliminary diagnoses were: psoas abscess, muscle or soft tissue abscess due to tbc, infectious hip arthritis and

Table I. The laboratory assessments of the patient.

Laboratory Parameter	Results
Leu/Neu/Lym	13.6/11.9/1.3 ($10^3/\mu$L)
Hb/Plt	12.2 (gr/dL) / 221 ($10^3/\mu$L)
AST/ ALT	30/31 (U/L)
CK	91 (U/L)
Creatinine	0.55 (mg/dL)
ESR	104 (mm/h)
CRP	243 (mg/L)
Procalcitonin	0.89 (ng/mL)
Urine analysis	Prt: neg, er: 1, dens: 1032, leu:1
Serum albumin	2.74 (mg/dL)
Serum Na/K	132 / 4.5 (mmol/L)
Brucella Rose Bengal and serum agglutination tests	Negative
Blood culture	Growth of *Salmonella enteritidis*

Leu: leucocyte; Neu: neutrophil; Lym: lymphocyte; Hb: haemoglobin; Plt: platelet; AST: aspartate aminotransferase; ALT: alanine aminotransferase; CK: creatin kinase; ESR: erythrocyte sedimentation rate; CRP: serum C-reactive protein; Na: sodium; K: potassium; mg: milligram; prt: protein; dens: density; leu: leucocyte; er: erythrocyte.

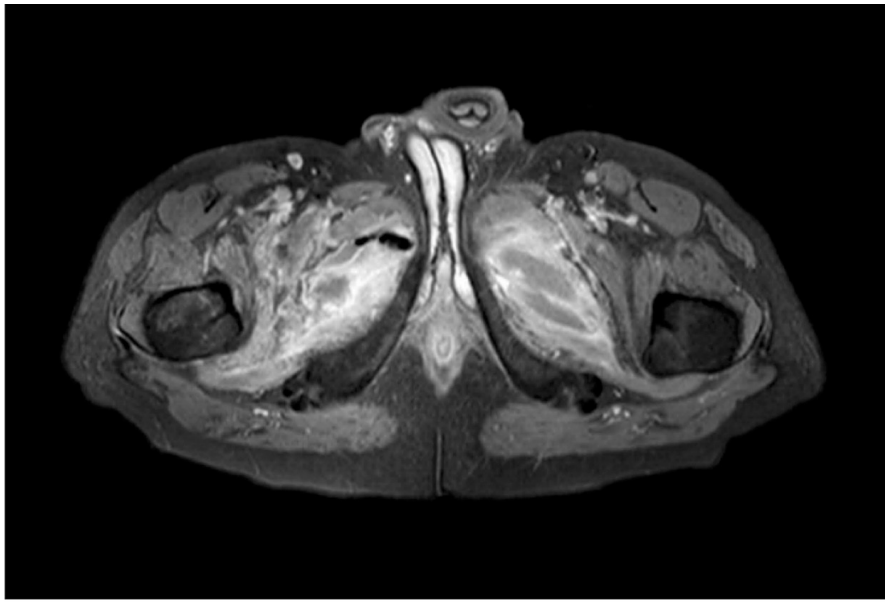

Fig. 1. The magnetic resonance imaging assessments of the patient.
The contrast enhanced, SPIR and T1 weighted, transverse plane MR images showing fluid collection and edema in both adductor and obturator muscle groups, pyomyositis in thigh muscles, air view due to abscess in muscles.

inflammatory hip arthritis, inflammatory myositis or acute venous thrombosis due to BS. Fever was observed over 38.5°C during close monitoring. AZA and cyclosporine were discontinued; only CS was continued at a lower dose. Blood culture tests were made. The laboratory data revealed high serum C-reactive protein (CRP), normal creatinin kinase (CK) and high procalcitonin (Table I). In the Doppler ultrasonography of the lower extremities there was no finding for venous thrombosis.

Magnetic resonance imaging (MRI) was performed on the hips, lumbar region, and proximal parts of the lower extremities, revealing fluid collection and oedema in the adductor and obturator muscle groups. Imaging of the pyomyositis in the thigh muscles revealed a 2 cm-sized abscess and myonecrosis in these muscle groups (Fig. 1-2). There was also femoral avascular necrosis bilaterally.

Muscle tissue cultures and biopsy were performed from the right thigh muscle abscess with a local incisional surgery. No microorganisms were isolated in the tissue cultures. Tbc polymerase chain reaction (PCR) test was negative and no signs were observed for inflammatory myositis in the pathological sampling. However, S. enteritidis was isolated in the blood cultures.

The patient was treated with ceftriaxone 2000 mg/day for 8 weeks with ciprofloxacin 1000 mg/day for 6 weeks. Drainage of the abscess was not found to be appropriate because of the difficult location of the abscess which would mean very-deep invasion into the muscle fibres. A clinic and laboratory response were observed within the first week and remission was observed in MRI within the first month.

Discussion

Pyomyositis might have serious complications such as diffuse muscle infections, abscess formation, myonecrosis, compartment syndrome, invasion to adjacent tissues and joints, and sepsis. *S. aureus* is the most common factor (more than 75%), while *S. enteritidis* in a very small proportion of cases (nearly 2%) (1).

Infections caused by Salmonella are generally derived from the gastrointestinal system and generally self-limited. Bacteremia may be seen in 5-45% of the patients in the course of the disease (3). In a literature review including patients with Salmonella pyomyositis, S. enteritidis was found to be responsible for 50% of cases (2). Patients often have diarrhoea as a primary infection. However, in this case, the patient presented with pain in the thighs and fever, without any evidence of gastroenteritis.

In previous cases it was observed that Salmonella pyomyositis was reported in elderly patients (4), children (5), patients with DM (6), acquired immune deficiency syndrome (AIDS) (7), cancer (8), and sickle cell anaemia (9). Pyomyositis due to Salmonella has rarely been reported in immunocompetent individuals (10). In previous reports there was one case of systemic lupus erythematosus (SLE) under CS treatment with Salmonella pyomyositis (11). This case is first in the literature to report a BS patient with Salmonella

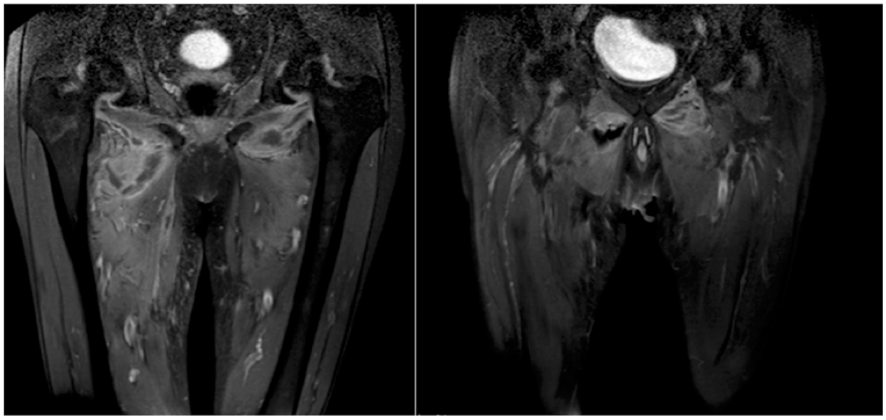

Fig. 2. The magnetic resonance imaging assessments of the patient.
The contrast enhanced, SPIR and T1 weighted, coronal plane MR images showing fluid collection and edema in both adductor and obturator muscle groups, pyomyositis in thigh muscles, air view due to abscess in muscles.

pyomyositis.

MRI is recommended as the first choice in diagnosis. In this case, besides the muscle oedema created by the infection, abscess formation and air density areas due to gas formation within muscle tissues were observed in the MRI. Gas accumulation has also been previously reported in case reports of pyomyositis due to Salmonella strains (6, 8, 11).

The preliminary diagnosis of this case included inflammatory myositis. The severe posterior uveitis, new-onset erythema nodosum lesions might also be considered as clues that this patient is in the active period of BS (12, 13). The rare findings of BS includes localised or diffuse inflammatory myositis (14-16). However, having high fever, elevated procalcitonin, the detection of abscess and gas formation in the MRI, the absence of inflammatory myositis in the pathology samples and normal CK excluded that patient had Behçet's myositis.

Pyomyositis due to Salmonella have been reported to have higher mortality rates (30%) compared to other microorganisms (17). Because the bacteremia is generally transient, it might not be possible to monitor the microorganisms in blood cultures, and also sometimes from the tissue cultures.

In summary, this case is important since it is the first to report an adult BS patient with Salmonella pyomyositis. These reports of cases have considerable importance for guiding clinicians as rare microorganisms might cause rare clinic manifestations in patients who have diseases and treatments causing immunosuppression.

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