Multiple *Salmonella enteritidis* leg abscesses in a patient with systemic lupus erythematosus

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Summary: We describe a 19 year old woman with systemic lupus erythematosus on corticosteroid therapy, who developed bilateral, multiple, gas-forming *Salmonella enteritidis* leg abscesses and osteomyelitis mimicking deep vein thrombosis. The infection was treated successfully by a combination of surgical drainage and intravenous ceftriaxone, followed by prolonged oral pefloxacin.

This rare case of gas-producing *S. enteritidis* emphasizes the difficulty in diagnosing such complications in active systemic lupus erythematosus.

Introduction

Systemic lupus erythematosus (SLE) is one of the diseases thought to predispose to severe salmonella infections.^{1–3} However, such infections rarely present as soft tissue abscesses: Cohen *et al.*⁴ found only one case with SLE out of 42 soft tissue infections in a recent review on extra-intestinal salmonella infections during the antibiotic era. The diagnosis is more difficult when the salmonella infection occurs during an exacerbation of SLE, as illustrated by the following case.

Case report

A 19 year old Arab woman with a one-year history of SLE, on corticosteroids, presented with a painful swelling in the right leg just below the knee, fever, and exacerbation of her underlying disease manifested by arthritis, massive proteinuria and high titres of anti-DNA. Symptoms had persisted for 2 weeks.

Physical examination revealed a woman with cushingoid appearance and a temperature of 38°C. Heart, lungs and abdominal examination were normal. There was a 2+ pitting oedema over her ankles and sacrum. Her hands, elbows and knees were swollen and tender, and there was a tender swelling in her right leg, extending proximally from below the knee to the ankle. The thigh and calf on both sides were oedematous and painful.

Urine collection showed a proteinuria of 15 g/24 hours, serum albumin 20 g/l, creatinine 70.9 µmol/l.

Haemoglobin was 6.2 mmol/l, white blood cell count 9000/mm³, ESR 78 in the first hour (Westergren method), antinuclear factor highly positive, and anti-DNA binding 40%.

Initial diagnosis of deep vein thrombosis was made, and treatment begun with intravenous heparin. This therapy was discontinued 4 days later because of increased swelling of the right leg. Radiographs of the legs demonstrated bilateral soft tissue swelling with gas bubbles, multiple fluid levels on the right leg, and no bone involvement in either leg. Marked muscle atrophy was also evident (Figure 1). Ultrasound showed patent veins and fluid collection in both calves. Needle aspiration with the aid of ultrasound yielded 100 ml of brown fluid, found by light microscopy to contain a few blood clots and leucocytes, suggesting an infected haematoma.

Computed tomography (CT) of the lower extremities revealed multiple fluid collections consistent with abscesses in both legs and thighs (Figure 2). A gas-producing bacterial infection was diagnosed, and surgical drainage performed. Cultures of blood and aspirated fluid yielded *S. enteritidis*. A bone scan showed increased uptake in the right tibia and left femur, correlating well with the periosteal changes on the CT scan which were most probably secondary to the soft tissue infection. Stool culture was negative.

Treatment was begun with oral co-trimoxazole, and an attempt was made to taper the steroid dosage with the addition of azathioprine. However, pancytopenia soon developed and fever persisted, and the antimicrobial treatment was changed to ceftriaxone 2 g/day, which was replaced after one

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Figure 1 Radiograph of right leg with horizontal beam showing normal bones, soft tissue swelling, separation of muscles, and air-fluid levels in both the thigh and calf.

Figure 2 CT scan through the distal third of both thighs showing muscle atrophy and large bilateral fluid collections.

week by oral pefloxacin 800 mg/day when the granulocyte count returned to normal.

Following control of the infection by surgical drainage and the described antibiotic treatment, the patient's lupus flare-up subsided: the anti-DNA reduced to 6% and the proteinuria disappeared. Computerized tomography performed one month later showed abatement of the fluid collections, and persistence of marked muscle atrophy.

The patient was discharged to an orthopaedic rehabilitation centre, where her ability to walk and function unassisted improved markedly. Since the possibility of osteomyelitis was not entirely ruled out, she continued the oral antibiotics for 3 months, according to the protocol for salmonella osteomyelitis in SLE patients described by Sattar and Molly⁵ and Ortiz-Neu *et al.*⁶

Discussion

Conditions predisposing to salmonella soft tissue infections and osteomyelitis are sickle cell disease,⁷ underlying malignancies,⁸ diabetes mellitus,⁹ renal transplants,¹⁰ previous trauma and preexisting bone disease,¹¹ steroid therapy,¹² and connective tissue disorders, including SLE.^{5,6,13,14}

The large number of patients with underlying diseases may reflect the importance of an immune mechanism in clearing salmonella organisms. Serum from patients with sickle cell disease has been shown to have deficient complement-mediated opsonizing activity for salmonella,¹⁶ while patients with diabetes mellitus or SLE with and without corticosteroid therapy, as in our case, have a defect in their cellular immune system which may be important in salmonella infections.¹⁵ Factors in our SLE patient that might contribute to this predisposition include the use of an immunosuppressive drug (prednisone), and the impaired granulocytic and monocytic phagocytosis that has been described in SLE patients.³

The combination of salmonella soft tissue infection and osteomyelitis in a SLE patient is extremely rare, and only one such case was found by Cohen *et al.*⁴ in a recent review of the literature on extra-intestinal salmonella infections. The two complications in these patients pose a difficult diagnostic problem, since they can be masked by the fever and joint swelling associated with an exacerbation of the underlying condition. In addition, as in our patient, the clinical presentation can be highly suggestive of acute thrombophlebitis, which sometimes occurs in SLE. Only the worsening of the calf swellings following anticoagulation therapy, and the demonstration of fluid collections on imaging, led to the correct diagnosis.

The *S. enteritidis* infection in our patient presented as bacteraemia, leg abscesses and osteomyelitis. It was demonstrated by X-ray of the patient's legs, and necessitated aggressive surgical drainage and prolonged antimicrobial therapy. To the best of our knowledge this is only the second reported case of subcutaneous salmonella-induced gas production.¹⁶

Until recently, antimicrobial therapy for systemic salmonella infection was limited to ampicillin, chloramphenicol and co-trimoxazole.¹⁴ Despite their potential role in the treatment of serious salmonella infections, there are only a few reports of successful treatment with third generation cephalosporins or quinolones.¹⁷⁻¹⁹

The need for long-term oral treatment in our patient led us to choose a short intravenous course of ceftriaxone followed by prolonged oral pefloxacin. Improvement expressed by normalization of temperature, sterile cultures and regression of the fluid collections was achieved over one month. The

development of salmonella enteritidis in the form of multiple gas-forming leg abscesses and osteomyelitis signifies the kind of serious life-

threatening complication that can develop in the course of a basically controlled disease.

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