

## A case of relapsing *Salmonella* osteomyelitis in a thalassaemia trait patient

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**Abstract** We report a case of chronic relapsing osteomyelitis caused by *Salmonella* Stanley in a  $\beta$ -thalassaemia trait patient who is otherwise normal. The importance of obtaining definitive bacteriological diagnosis and timely intervention to treat bone infection effectively is emphasised here.

**Keywords** *Salmonella* · Osteomyelitis · Haemoglobinopathies · Thalassaemia

### Introduction

There have been various case reports of *Salmonella* osteomyelitis in sickle cell anaemia [1, 2]. We present the first case of *Salmonella* Stanley causing a relapsing bone infection at the same site 12 years apart in a patient with  $\beta$ -thalassaemia trait. There is an increased incidence of foodborne salmonellosis in England [3]. Systemic infections may therefore become of increasing importance for clinicians. Currently, no clinical studies or antibiotic and surgical guidelines exist to guide treatment of *Salmonella* osteomyelitis.

### Case report

A 36 year old male was referred to the accident and emergency department with a suspicion of septic arthritis

of the right ankle. He complained of swelling and pain of three days duration with no history of trauma, an inability to bear weight, and the patient had an unremitting fever for two days. He had a history of infection of the right ankle 18 years back, which was treated with antibiotics only, and he had also been diagnosed with  $\beta$ -thalassaemia trait. On examination he was hemodynamically stable, the right ankle was swollen and erythematous, and ankle movements were restricted and painful. A complete blood profile was done, and it suggested a raised WCC of  $9.3 \times 10^9/L$  and a CRP of 228 mg/L.

AP and lateral radiographs of the right ankle (Figs. 1 and 2) showed mixed areas of radiodensity and radiolucency in the medial malleolus, with no periosteal reaction. Aspirate of the ankle joint did not yield any organisms, and blood cultures were negative.

He was started on intravenous antibiotics for three days, but the patient continued to suffer further spikes in temperature, increasing swelling and worsening of the pain.

His inflammatory markers were on the rise. The patient was taken to theatre for exploration and decompression of the right ankle. Twenty millilitres of pus were drained through an anteromedial incision. Following the creation of a cortical window, purulent material was drained from the medullary cavity and specimens of osseous material were sent for culture and sensitivity testing. A curette was used to evacuate any loculation of pus in the medullary canal. Thorough debridement and irrigation of the area was performed using 3 L of normal saline. Betadine-soaked gauze was then loosely packed into the intramedullary canal to allow postoperative drainage.

The patient developed pneumonic consolidation and was hence transferred to the intensive care unit for 24 h; he subsequently stabilised while on antibiotic therapy for the chest infection. Further debridement was carried out in

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**Fig. 1** AP radiograph of the ankle showing the characteristic lesion in the distal tibia

48 h, and a final washout and secondary closure was done after another 48 h. Microbiology culture and sensitivity results showed *Salmonella* Stanley that was sensitive to gentamicin and ciprofloxacin. He was started on intravenous gentamicin and ciprofloxacin for two weeks, followed by oral ciprofloxacin for six weeks. For two weeks the wound was clean and dry. The inflammatory markers WCC and CRP slowly fell to near normal. He was discharged and followed up in the community periodically due to the chronic nature of this disease.

## Discussion

It was Hodges and Holt in 1951 who reported an increasing frequency of *Salmonella* osteomyelitis in sickle cell disease [1]. *Salmonella* is a Gram-negative nonsporing bacillus belonging to the family of Enterobacteriaceae. Most cases are from contaminated food or water, with the small bowel being the habitat for the organism. Most often it results in self-limiting gastroenteritis requiring no antibiotics. Various other extraintestinal *Salmonella* infections, like meningitis and osteomyelitis, have been reported [4, 5].

*Salmonella* osteomyelitis is very rare (0.45% of all osteomyelitis), and osteitis is reported in only 0.8% of cases of typhoid fever [6]. The common way in which this infection spreads is haematogenously; the most frequent sites involved are the diaphyses of long bones and



**Fig. 2** Lateral radiograph of the ankle showing the characteristic lesion in the distal tibia

vertebrae [7, 8]. This can be attributed to increased bone marrow activity and increased blood degradation products, which favours the growth of *Salmonella* [1, 9]. The association of *Salmonella* osteomyelitis with sickle cell anaemia and other haemoglobinopathies is well recognised [8]. A preceding history of infection is not usual [10]. Stool cultures may or may not be positive [10]. Serological tests for *Salmonella* are of limited value under these circumstances [10].

Various reports suggest that non-Typhi serotypes are frequently the causative agent [8, 11–13].

*Salmonella* Stanley subtypes have been isolated from a certain brand of peanuts in three countries, namely Australia, Canada and the UK [14]. These subtypes are fully sensitive to antibiotics and have a distinct pulsed field gel electrophoresis profile. Salmonellosis is a global problem due to the international movement of foods and its high incidence in exporting countries. In September 2001, Australia isolated *Salmonella* Stanley from imported peanuts in an outbreak investigation, which resulted in wider investigations in Canada, England, Wales and Scotland [14].

There is no consensus regarding the treatment of acute and chronic osteomyelitis, as no randomised or case-control studies have been performed. The antibiotic treatment for osteomyelitis should ideally be initiated after obtaining sufficient samples from bone for microbiological assay.

Ampicillin, chloramphenicol, third-generation cephalosporins and quinolones have been used alone or in combination [15]. Hyperbaric oxygen therapy has also been used as an adjunct [15]. Ciprofloxacin has been recommended for the treatment of chronic bone infections, as it can achieve adequate levels in the bone. There are few reports of relapsing osteomyelitis in the literature [16, 17]. Lang et al. [17] described a unique case of two episodes of *Salmonella paratyphi C* osteomyelitis at different sites in the lower limbs 17 years apart. There is no consensus in the literature on ways to prevent chronic or relapsing *Salmonella* osteomyelitis. Initial investigations are full blood count, inflammatory markers (ESR, CRP), radiographs and bone scan, followed by MRI. These modalities help to identify bone infections [15].

The patient was given IV gentamycin and ciprofloxacin for two weeks followed by oral ciprofloxacin for a further six weeks. The duration of the antibiotics was based on clinical features and inflammatory markers. Due to the chronic nature of the infection the patient is still being followed up at our institution.

The patient provided informed consent to the publication of this clinical case.

**Conflict of interest statement** The authors declare that they have no conflict of interest related to the publication of this manuscript.

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