

Case Report

Unilateral Pyogenic Sacroiliitis Caused by *Salmonella typhi*: a case report

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Introduction

Typhoid fever or enteric fever is a multisystem illness with a potentially fatal outcome caused primarily by *S. enterica* serotype Typhi. The infection has various clinical manifestations that can vary from a simple gastroenteritis to severe typhoid fever. Around 21 million new typhoid fever cases are reported worldwide, annually [1]. The estimated annual mortality from typhoid is approximately 190,000 and had increased by 39% between 1990 and 2010 [2].

The number of notified cases of typhoid fever in Sri Lanka has reduced. From 2011 to 2016, the number of cases gradually declined from 1128 to 528 per annum. The distribution of typhoid fever is restricted to certain geographical areas of the country. These include the Jaffna, Vavuniya, Colombo, Kegalle and Nuwara Eliya districts. Our patient was also from the highly prevalent Nuwara Eliya district [3].

Pyogenic sacroiliitis caused by bacterial infection is a relatively rare condition. It is commonly caused by *Staphylococcus aureus*, *Streptococcus* species and *Pseudomonas aeruginosa* [4]. Sacroiliitis caused by *S. enterica* serotype Typhi is extremely rare. Only a few case reports are available in the current literature. According to our knowledge, this is the first reported case of pyogenic sacroiliitis caused by *S. enterica* serotype Typhi in Sri Lanka.

Case report

A 16-year-old schoolgirl from an estate in Nuwara Eliya had presented to the National Hospital, Kandy with a three-day history of severe left sided hip pain and difficulty in walking. On further inquiry, she had revealed a history of constipation followed by diarrhoea one week prior to the admission. In addition, she had fever, on and off, for the

same duration. There was no associated abdominal pain, red eye or dysuria. She had no past history of hair loss, oral ulcers or photosensitive rashes or small joint pain.

On examination, she was found to be febrile (104 °F) and pale and had a pulse rate of 90 beats per minute and her blood pressure was 90/60 mmHg. There was severe tenderness over the left sacro-iliac joint and the FABER test demonstrated pain in the left buttock region. On abdominal examination there was mild hepatomegaly with splenomegaly. The rest of the systemic examination was unremarkable.

Her full blood count (FBC) showed pancytopenia with neutropaenia. So, she was started on intravenous piperacillin tazobactam suspecting neutroapenic sepsis. In addition, she was found to have high inflammatory markers with deranged liver enzymes (Tables 1,2 and 3). Two blood cultures became positive for *S. enterica* serotype Typhi after 14 hours of incubation. She was started on intravenous ceftriaxone 1g twice daily and oral azithromycin 500mg once daily according to the local sensitivity pattern.

Table 1: Full Blood Count

	Day1	Day3	Day4	Day6	Day8	Day10
Haemoglobin (g/dl)	8.1	7.5	7.7	7.9	7.9	8.9
White cell count (10*3µl)	2.7 (N-21%)	3.89	4.92	4.95	8.66	8.26
Platelets 10*3µl	101	102	119	133	210	365

(N- neutrophil percentage)

Table 2: Liver function/enzymes

	Day1	Day4	Day9
Alanine transaminase U/L	128.5	137.6	86.2
Aspartate transaminase U/L	109.3	107.5	43.8
Total bilirubin µmol/l	31.57	15.94	12.21
Direct bilirubin µmol/l	22.9	11.81	8.63
Alkaline phosphatase U/L	121.3	199.9	173.4
Gamma glutamyl transferase U/L	158.3	187.2	149.9

Table 3: Inflammatory markers

	Day 1	Day5	Day10	Day21
CRP mg/L	82	31	3.8	
ESR mm/ 1 st hour	68			12

While on antibiotics, she was further evaluated for the cause of her left sided hip pain and pancytopenia. An x-ray of bilateral sacroiliac joints showed sacroiliitis in the upper half of her left sacroiliac joint (Figure 1).

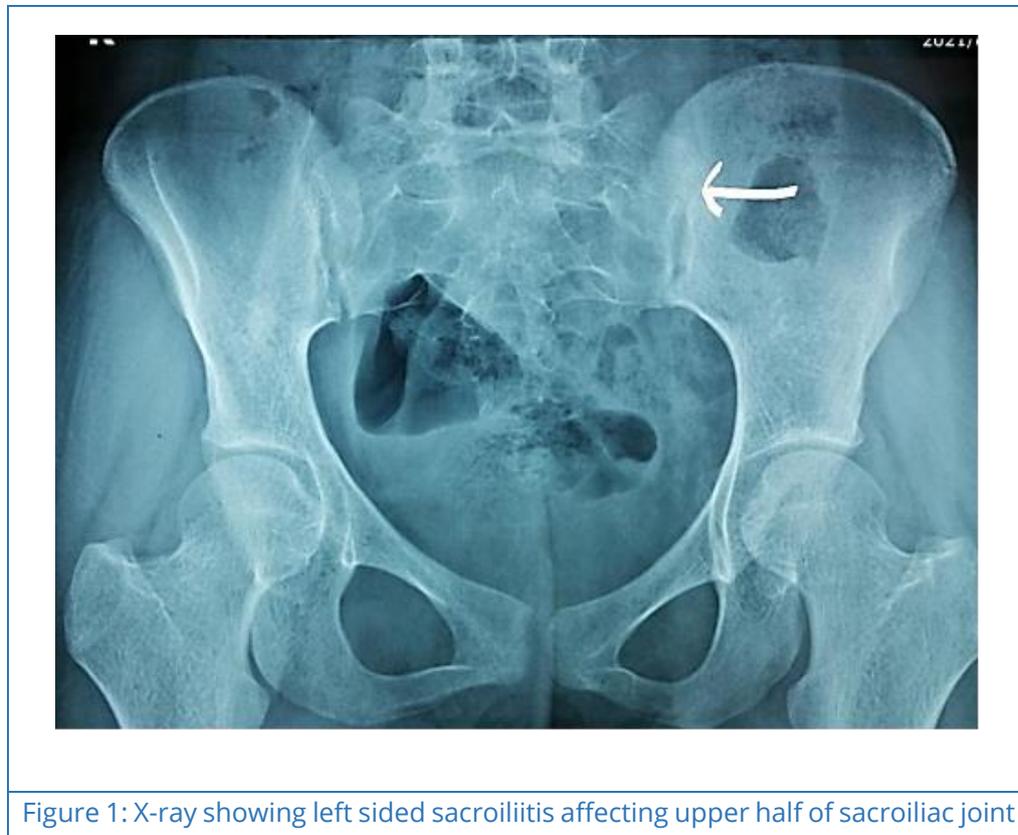


Figure 1: X-ray showing left sided sacroiliitis affecting upper half of sacroiliac joint

She was further evaluated with a magnetic resonance imaging (MRI) scan of bilateral sacroiliac joints which showed left sacroiliitis with some evidence of possible early abscess formation within the upper part of the left sacroiliac joint and inflammation extending to the left iliopsoas muscle (Figure 2).

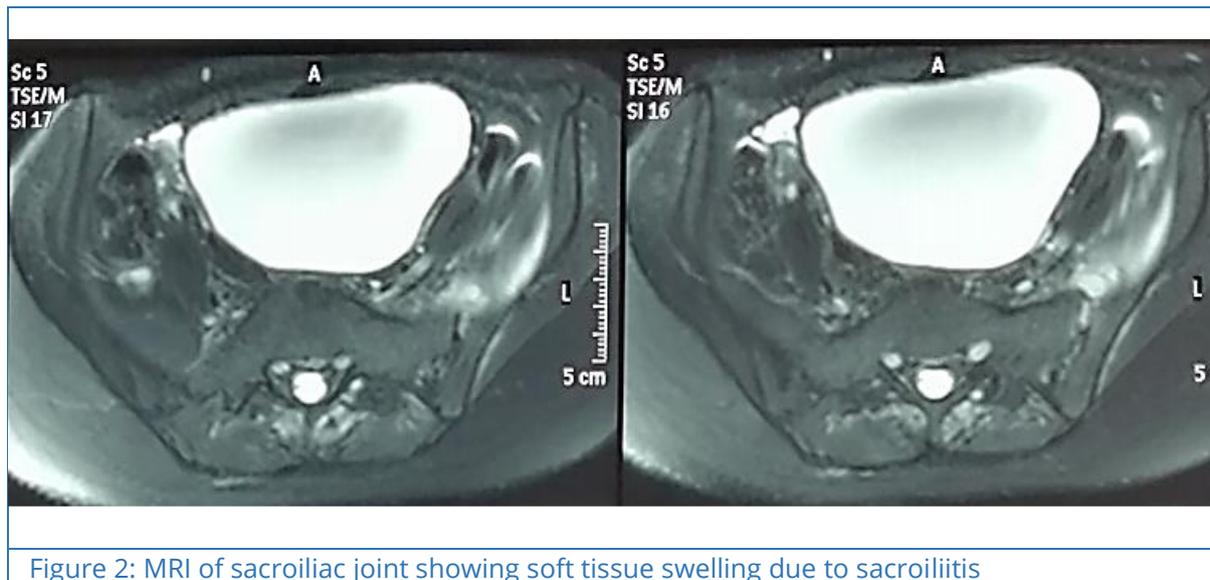


Figure 2: MRI of sacroiliac joint showing soft tissue swelling due to sacroiliitis

Her blood picture showed pancytopenia with evidence of liver pathology. The bone marrow biopsy revealed a reactive marrow with no evidence of malignancy or haemophagocytosis. Her anti-nuclear antibody was positive at a 1/100 titre and both C3 and C4 complement levels were high.

She was given a total of 14 days of intravenous ceftriaxone and oral azithromycin with a further one week of oral azithromycin. Her inflammatory markers normalised with antibiotic treatment and, therefore, drainage of abscess was not attempted. Her pancytopenia also improved over time with antibiotics and the simultaneous administration of haematinics, including folate and vitamin B12. Once inflammatory markers were normalised, she was started on a short course of non-steroidal anti-inflammatory drugs and her pain subsided. As she was pain free, with normal inflammatory markers, further imaging was not done.

Discussion

Typhoid fever is a systemic infection caused by *S. enterica* serotype Typhi, a human specific pathogen. Typhoid is contracted by consuming food or water contaminated with the pathogen. In developed countries, it occurs mainly in travellers returning from endemic countries. Endemic areas of typhoid include developing countries with poor sanitary facilities. In endemic areas, there is a high risk of acquiring disease by consuming food prepared outside the home, drinking water contaminated with the pathogen and having close contact with a person with recent typhoid fever [1,5,6]. In Sri Lanka, too, typhoid is predominantly confined to areas with poor sanitary facilities and scanty water resources [3]. In our case, the patient was from the estate sector in the Nuwara Eliya District where there is poor infrastructure and people are forced to obtain drinking water from open sources, usually contaminated by human excreta due to lack of lavatories. Further enquiry revealed poor personal hygiene in our patient who also did not use boiled and cooled water for drinking.

After ingestion of the microorganism, the patient can be asymptomatic for 7 to 14 days. When bacteraemia develops, they become symptomatic with fever and malaise. This usually occurs at the latter part of the first week. Usual history is constipation followed by diarrhoea with high progressive fever spikes. Other symptoms include headache, myalgia, abdominal discomfort, anorexia, nausea, and dry cough. Physical examination may reveal hepatosplenomegaly. Relative bradycardia is a commonly described finding which we observed in our case. Other physical findings include coated tongue and blanching erythematous maculopapular lesions occurring in the abdomen and chest known as rose spots [3,6,7]. Our patient had a dark complexion, and this was not detected. Though the imaging findings were not supportive, elevated transaminases and high bilirubin, alkaline phosphatase and gamma glutamyl transferase levels suggested co-existing hepatitis and cholecystitis. Both are well described complications of typhoid fever. Fortunately, there was no gastrointestinal perforation or haemorrhage or any complications in the cardiovascular, respiratory or nervous systems.

There are several reports describing pancytopenia in typhoid fever due to various reasons. Those include infection related bone marrow suppression, haemophagocytic syndrome and disseminated intravascular coagulation [8]. Due to the reactive marrow found on bone marrow biopsy and resolution of the cytopaenia with antibiotic treatment we made a diagnosis of infection related marrow suppression in this case.

The unusual complication our patient had was unilateral sacroiliitis and early abscess formation. A literature search showed a few case reports with the same complication. Patients with sickle cell haemoglobinopathies, systemic lupus erythematosus and immunosuppression are susceptible to bone and joint involvement in typhoid fever. This occurs in less than 1% of all *Salmonella* infections [9]. Our patient's anti-nuclear antibody was positive, and she is being followed up regularly at clinic level for possible development of an autoimmune connective tissue disorder.

All the available case reports, including our case, showed unilateral sacroiliitis involving the left sacroiliac joint. Treatment protocol for typhoid fever complicated with sacroiliitis is not established as yet. The antibiotic treatment should be individualised according to the sensitivity pattern and considering local resistance. Most patients were treated with intravenous quinolones (ciprofloxacin) or third generation cephalosporins for a minimal duration of 14 days. Then an oral antibiotic can be continued for a maximum of 4 weeks of total antibiotics [9,10,11]. In our case, the patient was initially started on intravenous ceftriaxone 2g daily and once the culture report was available oral azithromycin was added. The combination was continued for a total of 14 days and azithromycin continued for a further 7 days. Once inflammatory markers were negative, she was started on a course of diclofenac sodium together with limb physiotherapy.

An important aspect is prevention from getting the disease. As an endemic country, Sri Lanka has taken measures to prevent typhoid as well as to reduce mortality and morbidity. A diarrhoeal disease control programme commenced in 1983. This aimed

mainly to develop infrastructure and increase public awareness on improving personal hygiene [3].

Conclusions

Pyogenic sacroiliitis itself is a rare condition. Apart from the usual organisms, *Salmonella typhi* should be considered as a causative organism for pyogenic unilateral sacroiliitis in endemic areas including Sri Lanka. Early diagnosis with cultures and imaging, together with early commencement and adequate duration of antibiotic treatment are important in preventing permanent damage.

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