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Case Report

Uncommon *Salmonella* osteomyelitis in a previously healthy adult: a rare case report

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ABSTRACT

Salmonella osteomyelitis (OM) is more common in individuals with hemoglobinopathies like sickle cell anemia or thalassemia. The cardinal symptoms of OM such as fever, pain, and soft tissue swelling do not point towards a specific etiological agent. Since a preceding history of gastrointestinal infections is usually absent it causes a diagnostic challenge for *Salmonella* OM which is often overlooked causing a delay in diagnosis. In cases of fluoroquinolone resistance, third generation cephalosporins become the drug of choice. Due to absence of standardized guidelines, management is usually on a case-to-case basis. Previous studies have been focused mostly on children. Here, we present a rare case of *Salmonella typhi* OM in a 25-year male without any common predisposing factors.

Keywords: Adults, Osteomyelitis, *Salmonella typhi*, Stewardship

INTRODUCTION

Osteomyelitis (OM) is inflammation of the bone tissue usually as a result of infection. Despite advances in healthcare, this often represents a challenging condition with skin flora such as *Staphylococcus* being the most common etiological agent. *Salmonella* species (*spp.*) predominantly cause enteric fever, acute gastroenteritis, bacteremia, soft tissue infections, or asymptomatic carrier state.[1] *Salmonella* OM is a rare entity comprising of 0.45% of all OM and 0.8% of all *Salmonella* infections. Most common strains involved are *Salmonella typhi*, *Salmonella typhimurium*, and *Salmonella enteritidis* with only *S. typhi* capable of human to human transmission.[2]

Salmonella OM is more common in individuals with hemoglobinopathies like sickle cell anemia or thalassemia, splenic dysfunction, complement deficiencies, and genetic disorders. Sickling causes microscopic infarctions in bone facilitating bacterial entry from the gut to the bloodstream followed by drilling of the infracted bone by blood- borne bacteria.[3] Diabetes, autoimmune disorders, granulomatous diseases, immunodeficiencies, and HIV infection are other predisposing factors.[4] At present, most of the studies of *Salmonella* OM are focused on children with hemoglobinopathies. In this light, we present an uncommon case of *Salmonella* OM in a young adult with no common predisposing factors.

CASE REPORT

A 25-year-old male presented to the emergency department of a tertiary care hospital with the chief complaints of pain in left thigh since one and a half months which was accompanied by swelling and redness. Swelling was insidious in onset and was present from the past 3 months but pain was aggravated over the last 7-8 days. During this period, he was unable to extend his left knee completely. He also complained of intermittent fever since last 3-4 days. There was no history of trauma, bleeding disorders or hemoglobinopathies, rash, hemoptysis, increased sweating, heart disease, liver or kidney ailments, alcoholism, deep vein thrombosis, malignancy, malnutrition, or consumption of edema inducing

medications. He had taken multiple consultations from a private clinic near his home but his symptoms were not relieved. Subsequently, he came to our set up where he was admitted for further management.

On physical examination, he was well oriented to time, place and person. His vitals were within normal range. Respiratory and cardiovascular system examinations were also unremarkable. There was absence of pallor, icterus, clubbing, lymphadenopathy, and generalized oedema. On local examination, patient had a diffuse swelling over posterolateral aspect of left mid and distal thigh. Swelling was single, firm in consistency with tenderness, redness, induration, and raised local temperature. He was able to perform knee flexion from 30-80 degrees but extension was painful and restricted. Hip movements were free. Distal pulsation with ankle and toe movements were present. No crepitus or visible deformity was seen. No significant past medical or surgical history was present.

On the basis of clinical findings at the time of admission and radiological investigations (X-ray and MRI), a diagnosis of chronic osteomyelitis of left distal femur with extraosseous soft tissue abscess was made. Involvement of only one bone was present. Lytic sclerotic lesions were seen involving mid shaft of left femur with overlying periosteal thickening.

Samples for routine laboratory work up were sent. Culture reports for blood and urine sample were sterile. He also tested negative for Widal test, TyphiDot, acid fast staining, and tuberculosis polymerase chain reaction (TB PCR). Viral markers for hepatitis B and C with HIV were non-reactive. Complete blood count, liver and kidney function tests were within normal limits. Inflammatory markers such as C-reactive protein (CRP, 24.46 mg/dl) and erythrocyte sedimentation rate (ESR, 110 mm/hour) were raised. However, mean blood glucose and HbA1c were 283.35 mg/dl and 11.5% respectively. He was deemed immunocompetent but with new onset diabetes.

Incision and drainage with debridement and saucerisation of left distal femur was carried out. His pus sample was sent to the microbiology laboratory under sterile conditions. Sample was plated on blood and MacConkey agar (HiMedia®) as per standard microbiological techniques and incubated aerobically at 37°C. On the next day, grey moist colonies were observed on blood agar and pale non lactose fermenting colonies were seen on MacConkey agar. Pure culture growth was subjected to identification and sensitivity testing by VITEK® 2 compact instrument (Biomerieux) using GN and AST-N406 cards respectively as per manufacturer's instructions. Growth was reported as *Salmonella typhi* the following day with sensitivity as mentioned in Table 1. Results were interpreted as per Clinical and Laboratory Standards Institute (CLSI) guidelines.[5]

As per the culture and sensitivity report, patient was started on intravenous ceftriaxone EDTA sulbactam injection 1.5 gram twice a day for a week while inpatient. His condition improved gradually and he was discharged under satisfactory conditions. On discharge, he was advised tablet faropenem 300 mg twice daily for 2 weeks as an outpatient.

He was followed up by clinical, imaging, and laboratory work up. At the last known follow up, local examination was uneventful with no discharge, gap, or rise of temperature and patient had achieved clinical resolution of all prior symptoms. Follow up cultures were conducted twice at 2nd and 4th week post discharge and both were sterile. Pain was reduced and range of mobility was increased. Compression dressing was done and patient was advised for nil weight bearing on left leg and walker assisted gait training. After suture removal, he was deemed fit to join normal duties with precautions.

Table 1: Antibiotic sensitivity results for pus sample.

Organism isolated	*Salmonella typhi*
Antibiotic	Susceptibility
Amikacin	Resistant
Aztreonam	Sensitive
Cefepime	Sensitive
Cefoperazone/sulbactam	Sensitive
Ceftazidime	Sensitive
Ceftriaxone EDTA sulbactam	Sensitive
Ciprofloxacin	Resistant
Gentamicin	Resistant
Imipenem	Sensitive
Levofloxacin	Resistant
Meropenem	Sensitive
Minocycline	Sensitive
Piperacillin/tazobactam	Sensitive
Tigecycline	Sensitive
Trimethoprim/sulfamethoxazole	Sensitive

Continued.

Organism isolated	*Salmonella typhi*
Chloramphenicol	Sensitive
Azithromycin	Sensitive

DISCUSSION

Salmonella spp. are gram negative bacilli and a member of Enterobacteriaceae family. *S. typhi* is a rare cause of infectious OM with lack of globally recognized protocols for its treatment. Bone biopsy with histopathological examination and tissue culture are the gold standard for the diagnosis of OM.[6,7] However, most patients including ours are reluctant to go for biopsy.

Salmonella OM is a rare sighting and typically involves diaphysis of long bones, predominantly the humerus and femur. Radius, ulna, tibia, and lumbar vertebrae are other commonly involved bones.[4,8-11] Vertebral OM is usually caused by non-typhoidal *Salmonella* or *S. paratyphi.*[1] Third generation cephalosporins, fluoroquinolones, and chloramphenicol are the most commonly used antibiotics.[12] Since a preceding history of gastrointestinal infections is usually absent, *Salmonella* OM is often overlooked causing a delay in diagnosis or misdiagnosis.[3,4,11] In our case the diagnosis was delayed for over 3 months. This could be due to long incubation period or non-intestinal route of pathogen entry. S Stephanie et al quote that one quarter of patients infected with *Salmonella enterica serovar typhi* do not report gastric illnesses.[12] Widal test, TyphiDot, urine and blood culture were negative in our case.

Consequently, pus culture becomes the only source of accurate diagnosis for *S. typhi* OM. A review by Huang et al observed false negative serology and culture in literature. They also noted male preponderance and dominance of *S. typhi* OM which is similar to our report. Its presence in immunocompetent adults with no common risk factors reiterates the fact that *Salmonella spp.* should be considered in the differential diagnosis of OM even in people with no predisposing factors.[3] A diagnosis of new onset diabetes was also made for our case for which patient was given insulin injections and regularly followed up.

Inflammatory markers such as ESR and CRP were raised in our report. Huang et al mentioned that radiographical findings and abnormal inflammatory markers are common in all infectious OM and not particular to *Salmonella spp.*[3] Fluoroquinolones have a good oral bioavailability and powerful macrophage penetration but unfortunately, they were resistant in our report (Table 1). Consequently, third generation cephalosporins such as ceftriaxone are the preferred drug of choice as stated by Huang et al in their review.[3] However, cases of drug resistant third generation cephalosporin strains are also rising.[3,13,14]

Chronic *Salmonella* OM is conventionally treated by a combination of antibiotics and surgical debridement. Most of the cases cited in literature achieved clinical resolution of symptoms and no failures or recurrences were recorded.[3] Arora et al reported complete clinical and radiological resolution within 3 months with antibiotics alone.[2] Vynichakis et al observed that duration of treatment can vary from 1-6 months.[4] Our case required single surgical debridement procedure and two periods of antibiotic therapy. Resolution was achieved in about four months. Lack of globally accepted standardized guidelines for *S. typhi* OM and the subjective nature of treatment are possible limitations of the case report.

CONCLUSION

Salmonella OM is a rare clinical entity in patients without any known risk factors. However, in view of rising cases it is imperative to consider it in differential diagnosis of chronic OM especially in adults. With the emergence of drug resistant *S. typhi* strains, antibiotic sensitivity guided drug therapy and strict vigilance by the antimicrobial stewardship committee are need of the hour.

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