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Case Report

ENTEROCOCCAL MULTIFOCAL INFECTIVE OSTEOMYELITIS (MIO) IN A PATIENT WITHOUT SICKLE CELL DISEASE: A CASE REPORT

Banerjee S*, Rathore KS, Mali M and Elhence A

Department of Orthopaedics, All India Institute of Medical Sciences, Jodhpur

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ABSTRACT

Back ground: Multifocal infective osteomyelitis with simultaneous presentation in multiple bones is very rare. Specially in patients without sickle cell anaemia. We present a case of multifocal infective osteomyelitis in an otherwise healthy female.

Presentation: The patient presented with bilateral thigh pain of 3 months duration with no constitutional symptoms. The thighs were tender on palpation and on blood investigations a raised leucocyte count was observed. Radiology showed sequestrae in both femurs. MRI imaging also showed bone destruction and oedema in the bilateral femoral diaphysis.

Management: She was diagnosed as multifocal osteomyelitis and underwent saucerization and prophylactic fixation. The pus and sequestra were sent for culture and sensitivity and revealed Enterococci infection. She then underwent antibiotic therapy and prophylactic fixation for both femurs. She also developed similar symptoms in her humerus, which resolved with conservative management.

Take away: This is a first reported case of multifocal infective osteomyelitis of simultaneous onset and reaffirms the need for early diagnosis and surgical intervention both for debridement and prophylactic fixation. It also establishes the role of accurate culture and rational antibiotic therapy in management of such cases.

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INTRODUCTION

Osteomyelitis has been defined as inflammation of bone caused by an infective organism. Treatment of Osteomyelitis of a single bone in itself has been a challenge. Situation becomes worse when multiple long bones are simultaneously infected. Another similar entity called chronic recurrent multifocal osteomyelitis is simultaneous involvement of long bones due to noninfective aetiology characterised by reactive and plasma cell sclerotic process.

There have been rare case reports in literature of osteomyelitis caused by *Enterococci sps* in patients suffering from sickle cell disease^{1,2} with special predilection for vertebral bones. Other orthopedic infections caused by same organism is usually periprosthetic arthritis in elderly individuals. A deliberate and extensive search of literature was done but report of multifocal infective Osteomyelitis by *Enterococcus sps* was not found. So with avid interest we describe a case of Multifocal Infective Osteomyelitis (MIO) caused by Enterococci in a healthy young patient without sickle cell disease.

Case Presentation: A 24 year 7 months post partum lactating lady presented to our outdoor with complaints of pain in both thighs for 3 months. Pain had an insidious onset, first involved the right thigh was continuous, dull aching, non-radiating, localised to mid thighs, aggravated after prolonged walks, partially relieved with rest and over the counter analgesics and not associated with any fever. She had no history of any other constitutional symptoms such as fever, weight loss or loss of appetite. She had history of an uncomplicated, full term vaginal delivery at our institute 7 months back followed by an uneventful post partum period and was discharged next day from hospital. Her general examination was unremarkable with no pallor, icterus, cyanosis, clubbing, pedal edema or lymphadenopathy with unremarkable gait pattern. On local examination she had deep tenderness in bilateral mid thighs. The overlying skin was healthy with no scar, sinuses or discharge.

Investigations: Her Haemoglobin was 9.3 gm/dl, total leucocytes count were $13.01 \times 10^3/\mu\text{l}$ with 85% Neutrophils. Her radiographs revealed bony sequestra in both femoral diaphysis (Fig.1). Magnetic resonance imaging was performed which showed altered signal intensity all along the middle and

*Corresponding author: Banerjee S

Department of Orthopaedics, All India Institute of Medical Sciences, Jodhpur

lower third of both femur bones which was hypo intense in T1 and hyper intense in T2 weighted images with bony destruction and collection seen along the lesion in femur and soft tissue plains(Fig. 2).Her skeletal survey revealed ill formed sequestra in right humerus as well (Fig. 3).A differential diagnoses of polyosteotic skeletal tuberculosis, CRMO, multifocal osteomyelitis following *Salmonella sps* and Multi focal Infective Osteomyelitis was made.

Diagnosis and Management: Peripheral blood smear showed absence of sickle cells, so a decision for invasive diagnostic method in form of sequestrectomy, saucerization and debridement was taken to obtain specimen for ruling out other causes. She underwent the same. The decision regarding fixation was taken after calculating the Mirel score³. To prevent fractures (bilateral Mirel’s score for femur was 9), both femurs were stabilised with external fixators.In the right humerusMirel score was found to 6 and hence was managed conservatively. During debridement pus samples were collected and sent for Gram staining, AFB staining, aerobic cultures, and card based nucleic acid amplification tests for *Mycobacterium sps.*. Empirical antibiotic therapy was started with injectable Ceftriaxone and Gentamicin. Her cultures revealed *Enterococci sps.* in samples from both femur, so tubercular infection and CRMO (noninfective) causes were ruled out. The microorganism was found sensitive to Cefepime, Ceftriaxone, Piperacillin, Tobramycin, Ciprofloxacin and Meropenem but resistant to Amoxicillin- Clavulanicacid.

So Gentamycin was stopped and injection Ciprofloxacin was added.3 dayspost-



Figure 1 Lytic – sclerotic lesion seen in mid third diaphyseal region of both femur in AP and Lateral radiographs involving nearly the whole width of the bones



Figure 2 MRI both femur showing destruction and altered bone marrow signal intensity in mid and lower third of bilateral femur and increased signal intensity seen in thigh muscle on both sides and altered signal intensity seen in subcutaneous region of mid and lower third of left thigh



Figure 3 AP and lateral radiograph of right humerus bone showing Sclerotic lesion in mid third diaphyseal region involving nearly whole width.

Debridement, patient was afebrile and her TLC was $13.73 \times 10^3/\mu\text{l}$, ESR was 57 and CRP was 137.92. Relook debridement was performed on the 5th day and antibiotic (Vancomycin) mixed bone cement beads were kept in canals of both femur. IV antibiotics (Ceftriaxone and ciprofloxacin) were continued along with general nursing care and physiotherapy in form of pelvic lifting, incentive spirometry and knee mobilisation exercises. Patient remained afebrile and on 10th day her TLC was $12.06 \times 10^3/\mu\text{l}$, ESR was 43 and CRP was 100.38. At the completion of 3 weeks post surgery her ESR was 30 and hsCRP was 39.12 so from right femur antibiotic cement beads were removed, thorough debridement was done and external fixator was replaced with prophylactic intra-medullary interlocking nail for better rehab and knee physiotherapy. At the time of nail insertion repeat cultures were performed which were sterile with no growth at 48 hours of incubation. 1 week later left side external fixator was also exchanged with intra-medullary interlocking nail. During nail insertion iatrogenic fracture of shaft of femur occurred which was fixed with encirclage wiring over the nail. IV antibiotics were continued for 3 more weeks.

At completion of 6 weeks of parenteral antibiotic therapy patient was fine, the wounds had healed and she was doing her physio well. Patient was discharged on oral antibiotics (Tablet Cefuroxime & Tablet Ciprofloxacin) which were continued till 3 months. Patient was followed every month for next 18 months with serial radiographs and ESR monitoring and was allowed graduated weight bearing. Patient’s infection settled but iatrogenic fracture went in delayed union but had united at the last follow-up visit. The patient is now asymptomatic, weight bearing comfortably and back to all her routine activities of life. Her humerus lesion consolidated and healed spontaneously during this treatment.



Figure 4 AP and lateral radiographs of both femurs after bilateral tubular external fixator was applied following sequestrectomy, saucerisation and debridement



Figure 5 Postop image of Bilateral femur following exchange of external fixator by intramedullary interlocking nail. On left side, encirclement was done for intraoperative iatrogenic fracture



Figure 6 Figure shows healing of lesion and fracture site at the final follow up one and a half years after initial surgery



Figure 7 Radiograph of humerus at the last follow up visit showing healing of the osteomyelitic lesion with conservative management.

DISCUSSION

Enterococcus sp rarely described in case reports pertaining to osteomyelitis^{4,5,6}. The natural habitat of *Enterococcus sp* gut and it is frequently isolated from feces⁴. *Enterococcus sp* has been known to cause endocarditis, bacteremia, UTI, neonatal infections, CNS infections, pelvic and intra-abdominal infection, periprosthetic arthritis and even nosocomial infections and superinfections but it has been never implicated in multifocal osteomyelitis^{4,5,7}. This report describes a case of multifocal osteomyelitis caused by *Enterococcus sp* and its microbiological and diagnostic features. The patient described herein was a young woman without underlying medical problems, although may have been immunosuppressed due to pregnancy. Osteomyelitis is almost invariably the result of blood-borne seeding and can be associated with underlying endocarditis^{2,8}. Negative urine cultures made a urinary source unlikely in this case. It is not known if the patient had or was in contact of any pets or cattle. The infection described in this case was community acquired with the source of the isolate unknown. Empirical treatment for Osteomyelitis is directed towards the most common causes: *Staphylococcus aureus* and *Streptococci*^{2,4}. Such regimens are frequently in effective against enterococci (resistance to Amoxicillin and Clavulanic Acid was seen in these cases), as occurred in this case, highlighting the importance of wound culture to obtain a microbiological diagnosis, particularly in elderly patients⁴.

To the best of our knowledge this is the first report of multifocal infective osteomyelitis caused by *Enterococcus sp* and confirms the pathogenic potential of this bacterium to cause such an aggressively invasive human disease. A high index of suspicion for osteomyelitis has to be maintained while dealing with patients with chronic pain in long bones. The diagnosis hinges on early recognition of an infective process via blood investigations and a thorough radiological work up which can really bolster the chances of an optimal treatment. Early surgical intervention relies on the time-honored principles of thorough debridement, sequestrectomy and antibiotic therapy. Instillation of local antibiotic eluting devices helps in early control of the infection and reduces the need for prolonged systemic antibiotic therapy. The decision regarding prophylactic fixation/ additional stability to the weakened bone needs to be assessed and Mirel's scoring provides an excellent tool for objective treatment of such lesions. The authors also cannot over emphasize the need for obtaining adequate and representative samples during debridement and the role of microbiology in identification of the organism. The instillation of a rational antibiotic regimen is paramount in effecting eradication of the infection. We believe that the good outcome achieved in this case was a result of meticulous observation of the above-mentioned principles of management and the authors opine that a rational and objective diagnostic and management strategy can produce reproducible and effective outcome in such cases.

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