Case Report

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

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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 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 x 10^3/µl with 85% Neutrophils. Her radiographs revealed bony sequestrae in both femurs (Fig.1). Magnetic resonance imaging was performed which showed altered signal intensity all along the middle and...
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 score. To prevent fractures (bilateral Mirel’s score for femur was 9), both femurs were stabilised with external fixators. In the right humerus Mirel 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- Clavulanacid.

So Gentamycin was stopped and injection Ciprofloxacin was added. 3 days post-

Debridement, patient was afebrile and her TLC was 13.73 x 10^3/μl, 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 x 10^3/μl, 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.

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**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.

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

Enterococcus sps rarely described in case reports pertaining to osteomyelitis3,5,6. The natural habitat of Enterococcus sps gut and it is frequently isolated from feces5. Enterococcus sps 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.3,5,7 This report describes a case of multifocal osteomyelitis caused by Enterococcus sps 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 endocarditis2,8. 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 ineffective 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 microbiological diagnosis, particularly in elderly patients4.

To the best of our knowledge this is the first report of multifocal infective osteomyelitis caused by Enterococcus sps and confirms the pathogenic potential of this bacterium to cause such 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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