Journal of Health, Medicine and Nursing
ISSN 2422-8419 An International Peer-reviewed Journal
Vol.37, 2017

Bilateral Tibial Chronic Osteomyelitis in a Non-Sickler: A Case Report and Review of Literature

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ABSTRACT

BACKGROUND: Osteomyelitis in children has various clinical manifestations causing diagnostic and therapeutic difficulties. Inappropriate treatment of acute osteomyelitis may lead to a more chronic, serious and complicated form. Chronic osteomyelitis continues to be a major cause of morbidity and disability in children especially those living in developing countries.
OBJECTIVE: To present a case of bilateral tibial chronic osteomyelitis in a non-sickler and review the literature on the subject.
METHODS: An 8 year old boy presented with a 23-day history of high grade intermittent fever and a 22-day history bilateral leg swelling. Detailed history was obtained and physical examination carried out. He was further evaluated with laboratory and radiological investigations and received adequate treatment before he was discharged home.
RESULTS: He had periodic bouts of fever which was unresponsive to anti-pyretics. His legs were swollen and tender with inability to stand. Investigations revealed anaemia, leucocytosis, neutropenia, lymphocytosis and raised Erythocyte Sedimentation Rate of 128mm/hour. Soft tissue swelling around the tibia was seen on the radiograph. He had incision and drainage done on both legs with wounds packed and dressed. The drained purulent fluid was sent for microscopy, culture and sensitivity and it revealed growth of *Staph. aureus* sensitive to azithromycvin, genticin and ofloxacin. He was principally treated with antibiotics, analgesics and hematinics which yielded a good result.
CONCLUSION: Multiple bone chronic osteomyelitis occurs in non sickle cell disease patients who are immune competent. It can also be treatable simple incision and drainage of the soft tissue abscess and appropriate antibiotics, without classical bone operative procedures if diagnosed early, with a good outcome.
Keywords: Osteomyelitis, Chronic Osteomyelitis, Multiple Bone infections, Non-sickle cell patient.

INTRODUCTION

The term osteomyelitis was first used by the French surgeon Chassaignac in 1852.[1] It is defined as an inflammation of bone and bone marrow caused by pyogenic bacteria, mycobacteria or fungi.[2] It may also rarely be non infective[3]. Osteomyelitis may lead to formation of "sequestrum" (pockets of dead cortical bone), if left untreated with abscess formation, and "involucrum" (new bone incorporating the sequestrum).[4-7] Multiple openings in the involucrum develop which is called "cloaca" through which pus and sequestrum comes out of the bone.

Osteomyelitis in children continues to cause diagnostic and therapeutic difficulties. The clinical manifestations vary and classical signs and symptoms of the infection are not always present.[8] Bilateral tibial chronic osteomyelitis has rarely been reported in the literature and is usually accompanied by an underlying chronic disease, such as sickle cell disease[9].

Recent prospective studies found that uncomplicated osteomyelitis can be treated by three to four weeks of antibiotics therapy.[10] Recent studies have also demonstrated a 90% response rate to medical therapy alone when treatment was initiated promptly and continued for an appropriate time.[10]

CASE REPORT

History

An 8 year old primary school boy was referred to the surgical unit from the paediatric unit of our centre on his 8th day of admission. He was said to have presented with a 15-day history of high grade intermittent fever, 14-day history of progressively worsening bilateral leg swelling and 9-day history of inability to walk. There was a positive history of trauma to the left leg. There was also a history of purulent swelling over the posterior aspect of his right elbow which was drained by his mother with added self medication of ampiclox. He is not a known sickle cell disease patient (sickler) and there was no family history of such. At presentation, he was diagnosed

with cellulitis to rule out sepsis, malaria and sickle cell disease. Investigations were requested for and he received a full course of anti-malaria and a week course of antibiotics. The illness, however, showed no sign of improvement and he was referred to the surgical team for expert management.

Physical Examination

Clinical evaluation revealed an ill looking boy, mildly pale, febrile (38.2^{o}C), anicteric, not cyanosed, not dehydrated, with tachycardia of 102 bpm. There was no lymphadenopathy and no pedal oedema. Musculoskeletal system examination revealed bilateral leg swellings with shiny flaky skin, differential warmth, positive fluctuancy and marked tenderness. There was no discharging sinus.

Investigations

Results of his laboratory investigations showed genotype as AA, Haemoglobin 10 g/dl, White Blood Cell Count 12,180/mm^3, Neutrophils 20%, lymphocytes 77.5%, Erythocyte Sedimentation Rate (ESR) 128mm/hour. Radiographs of the legs done at presentation showed a soft tissue swelling. There was periosteal reaction, but no cortical destruction, sequestrum or involucrum was seen. A diagnosis of bilateral tibial chronic osteomyelitis was made based predominantly on the clinical and laboratory findings as well as the duration of the clinical features.

Treatment

Incision and drainage was done on the right and left legs, with drainage of 400mls and 250mls of purulent and seropurulent fluid from the right and left legs respectively. The cavities were irrigated and packed with guaze. Both legs were dressed with guaze and crepe bandage. The patient was placed on Intravenous Ceftriaxone and analgesics while awaiting results of microscopy, culture and sensitivity of the drained fluid. Weight bearing was restricted by wheelchair ambulation, to avoid a pathological fracture. The mucopurulent fluid was cultured and yielded growth of *Staphylococus aureus* sensitive to genticin, azithromycin and ofloxacin. The antibiotic therapy was changed to Ofloxacin tablets and he showed marked improvement with the swelling subsiding and cessation of the fever.

He was discharged 2 weeks later from the surgical unit on antibiotics and hematinics and was followed up as an outpatient in the orthopaedics cliinc. The response to treatment was good, and he was finally discharged from the clinic after 12 months.

DISCUSSION

Pathology

Osteomyelitis is mostly caused by a micro organism. In neonates, the bacteria most frequently associated with acute haematogenous osteomyelitis are those which cause neonatal sepsis, notably Lancefield group B streptococci (*Streptococcus agalactiae*) and *Escherichia coli* as well as *Staphylococcus aureus*.[11] In older children, *S.aureus* infection predominates and in some countries, such as the US, community-acquired methicillin-resistant strains (CA-MRSA) are increasingly recognised.[12] In a retrospective study carried out over a 30-month period from 2010 to 2012 on twenty-one cases of chronic tibial osteomyelitis in which treatment involved the use of OSTEOSET -T as a space filler and local antibiotic delivery system for the management of chronic osteomyelitis, microbiological analysis of the samples revealed 38% of the infections caused by strains of Staphylococcus species (half caused by S. aureus only) with a significant number having either a mixed growth or no identified growth.[13] *Staphyloccus aureus* was the offending organism in our patient being reported. This finding is in tandem with findings of other authors described above.

Bilateral chronic osteomyelitis is rarely seen, also when chronic osteomyelitis occurs simultaneously in two or more bones, it is usually associated with an underlying chronic illness, such as sickle cell disease. Bachmeyer and colleagues reported bilateral tibial osteomyelitis caused by Pantoea agglomerans (a rare gram-negative bacillus) in a patient with sickle cell disease.[9] This is not the case in our patient, who had no know underlying disease nor showed any clinical feature of any co-morbidity.

Clinical features

Common presenting signs and symptoms of chronic osteomyelitis which include pain, fever, tarchycardia, tenderness, fluctuancy, warmth, edema, swelling, abscess, intermittent acute exacerbations were all seen in the index patient presented. However other features such as chronic discharging sinus, surrounding hyper pigmentation and pathologic fractures were not found.

Radiological features

Generally, radiological examination is performed to evaluate the extent of bone involvement (for example, the extent of active intramedullary infection or abscess superimposed on areas of necrosis, sequestrum and fibrosis) and to identify soft tissue involvement (areas of cellulitis, abscess, and sinus tracts). Findings on radiographs may include: periosteal reaction, metaphyseal patchy rarefaction, soft tissue swelling, sequestrum and involucrum. Radiology is usually normal within the first 10 days of onset of osteomyelitis and by 10^{th}-14^{th} day, an extra-cortical outline denoting periosteal new bone formation is usually visible.[14] This may explain the reason why only periosteal reaction was seen in our patient, whereas other classical features of involucrum, cloaca, sequestration and pathological fractures were not seen in the radiograph of the legs of the patient which were taken less than 15 days of the onset of fever.

Other more sensitive methods of early diagnosis exist. Radionuclide bone scan with Technetium^{99} is useful for identifying other sites of skeletal involvement,\. The lesions demonstrate increased uptake on technetium bone scans, even if they are clinically silent.[15] Magnetic resonance imaging can also be done. It is very sensitive and is more useful for soft-tissue assessment and revealing early bony oedema.[16] Computed tomography is superior to plain radiography in vertebra or complex bones. Ultrasound is useful in children and it detects features like fluid collections, abscess and edema within 1 - 2 days of onset of disease. It readily depicts soft tissue involvement, but it provides only limited information about bone changes.[9] These were however not done for our patient, because they are not easily accessible in our centre.

Hematology (Haemoglobin, White Blood Cell count, ESRor C-reactive protein, blood culture), stool culture, aspiration and culture, ASO – Antistreptolycin O –antibody titre shows generalized affectation of the general body system. Those available at our centre were done for the patient presented and the results were supportive of diagnosis of chronic osteomyelitis. Also, the genotype result of AA, ruled out sickle cell disease.

Diagnosis

In consideration of diagnostic criteria, chronic osteomyelitis exists in the presence of any of the following conditions;[19]

1. Infection lasting 6 weeks or more (with radiological evidence)
2. Sequestrum formation or sclerosis
3. Relapse or persistence after initial treatment of acute osteomyelitis
4. Osteomyelitis associated with foreign bodies
5. Osteomyelitis associated with peripheral vascular disease
6. Osteomyelitis from organisms that produce chronic, indolent disease such as *Mycobacterium tuberculosis*

Emphasis should however, be based on the clinical history, physical examination and radiological findings.

Treatment

The first step in managing chronic osteomyelitis is to make the diagnosis. The mainstay of treatment of chronic osteomyelitis is surgical by complete removal of all infected and devascularized tissue which provides the only opportunity to eradicate the infection, because antibiotics cannot penetrate devascularized tissue.[20-24] However, recent studies have demonstrated a 90% response rate to medical therapy alone when treatment was initiated promptly and continued for an appropriate time.[25]

Non-operative management is indicated when there is no discrete sequestrum or inadequate involucrum. This is done using antibiotics based on sensitivity result. Resistant organisms such as Methicillin-Resistant Staphylococcus aureus (MRSA), Methicillin-Resistant Staphylococcus epidermidis (MRSE) and Vancomycin-resistant enterococci (VRE) have stimulated research to discover new drugs like Streptogramins (Quinupristin and Dalfopristin) effective against MRSA and VRE; Coumarin group (Novobiocin) inhibit DNA- gyrase and is effective against VRE.[26]As part of non-operative management, the ulcer and sinus are then dressed and cast placed to support pathologic fractures or prevent potential fractures. We were successful in treating our patient with initial use of parenteral Ceftriaxone and subsequently oral ofloxacin. The affected limbs were also drained of abscesses and resulting surgical wounds dressed till healing. To prevent pathological fractures, patient was kept non weight bearing in wheel chair ambulation.

The only operative intervention in the patient presented was the incision and drainage of the leg abscesses. In chronic osteomyelitis, operative treatment varies from simple drainage and debridement to ablation of a part or

the entire limb. [27] The principles of surgical treatment are; adequate sequestrectomy and curettage; saucerisation; management of ensuing dead space; management of bone loss and possible instability post sequestrectomy; reconstruction of soft tissue defects; adjunctive antibiotic therapy based on the outcome of the marrow curretings/sequestral culture and sensitivity result. The index patient, having had incision and drainage, was successfully treated with ofloxacin and genticin which were the antibiotics sensitive to the offending organism, *S. aureus* cultured on the abscess aspirate.

Complications

Complications of osteomyelitis include septic arthritis, destruction of the adjacent soft tissues, malignant transformation of the sinus or ulcer, secondary amyloidoses, and pathologic fractures. Our patient did not have any of these complications as the infection was diagnosed early and adequately treated. He was also placed on wheelchair which aided in preventing pathological fractures.

CONCLUSION

Bilateral tibial chronic osteomyelitis isa very rare entity in children who are non-sickle cell disease patients nor immune compromised. This makes this case report an interesting one. Early diagnosis, prompt and adequate treatment can ensure complete cure of the disease even without classical bone operative procedures. It also helps in preventing complications as was demonstrated in the index patient.

DECLARATIONS:

The authors categorically state that this manuscript has not been previously published and the manuscript is not under consideration elsewhere.
All the information contained in this manuscript are original except otherwise clearly stated and duly acknowledged.

Ethics approvals: Ethical approval was secured from the Ethical Review Committee of State House Medical Centre Abuja, Nigeria to publish this case report.

Consent for Publication: Not applicable

Funding: None

Conflicting interests: None

Authors' contributions: All the authors made substantial contributions in the study design, Literature Search, Literature review, and write up of the study..

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