

Case report:

Challenges in Diagnosis of Chronic Osteomyelitis : A Case Report from Sri Lanka

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Abstract:

The prevalence of osteomyelitis has been continuously decreasing in children with improvement of health care services and introduction of Hemophilus and Pneumococcal vaccines. Despite this, diagnosis and management of osteomyelitis are often a challenge to pediatricians as well as orthopedic surgeons. We report a 13-year old boy who had been treated as for rheumatic fever over 2 years with Benzathene penicillin, but ultimately turned out to have chronic osteomyelitis of right tibia. Evidence of chronic osteomyelitis was radiologically confirmed by X-ray and Computerized tomogram (CT) of right tibia and pus cultures grew *staphylococcus aureus*. Clinical features and biochemical markers completely resolved upon debridement of pus and intravenous antibiotics. He is currently on follow up at the orthopedic and pediatric clinics in the local hospital.

Keywords: Challenges in diagnosis, chronic osteomyelitis, debridement, *staphylococcus aureus*.

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Introduction

Chronic osteomyelitis frequently affects long bones of arms and legs in children, but other bones are not exempt¹. It often represents a catastrophic continuum of delay in diagnosis². The main causative organisms are bacteria but other organisms include fungi and tuberculosis especially in immunocompromised children³. The organisms spread to bone either from blood or adjacent tissues⁴. Diagnosis is naturally based on clinical features and supported by microbiological and radiological confirmation⁵. Variable and unusual clinical presentations often hinder accurate diagnosis of osteomyelitis⁶. Although chronic osteomyelitis is a rare disease in children⁷, it might cause severe sequelae such as growth failure, septic arthritis, destruction of joints, and permanent disability⁸. While delayed diagnosis can cause anxiety and apprehension in parents, the disease can have a lifelong impact due to complications⁹. We report a child, in whom chronic osteomyelitis was initially mistakenly diagnosed as rheumatic fever and chorea, and 4-weekly prophylaxis with Benzathene penicillin

was given for one year, before the diagnosis of chronic osteomyelitis was confirmed in retrospect.

Case report

A-13-year old child presented with right knee joint pain and right sided limp for one year and right knee joint swelling for one month duration. One year ago, he had been investigated for right knee joint pain, and fever with blood and radiological investigations at the local hospital. The investigations revealed elevated C-reacting protein (CRP), increased erythrocyte sedimentation rate (ESR), antistreptolysin O titre (ASOT) >800 U/mL and increased white blood cells. X-ray and ultrasound of right knee joint had been reportedly normal. Subsequently, he was treated as for acute rheumatic fever and had been on 4-weekly intramuscular Benzathene penicillin up to the current presentation. Notably, there had been no improvement in knee joint pain while on antibiotic prophylaxis and limp persisted. Then he was re-evaluated for abnormal lower limb movements which were misinterpreted as rheumatic chorea and treatment was continued. Over the course, he was on antibiotic prophylaxis, several courses

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of oral antibiotics and non-steroidal anti-inflammatory drugs (NSAIDs) were prescribed as for pharyngitis and knee joint pain by the general practitioner; however, the clinical improvement of right knee joint pain was only temporary and pain recurred upon discontinuation of antibiotics. There had been no history of falls or trauma to right lower limb.

Physical examination revealed an ill-looking, mildly febrile (37.8C), and limping child with objectively demonstrable swelling over anterior aspect of the right knee joint. The movements of the right knee joint were restricted by pain. Rest of the musculoskeletal and other system examination were normal. Investigations revealed leucocytosis (15.6×10^3 , N-74%), elevated CRP (190mg/dL), and ESR(78mm/hour). X-ray right knee joint showed periosteal elevation which was suggestive of osteomyelitis (Figure 1).

This was further explored by CT right knee joint as MRI(magnetic resonance imaging) was not available and CT revealed evidence of periosteal reactions, cortical destruction, and sequestrum formation. Pus and necrotic tissues were drained and sent for cultures by orthopedic surgeon; Pus cultures grew staphylococcus aureus and were sensitive to intravenous Cloxacillin. Blood cultures yielded no growth. Following diagnosis of chronic osteomyelitis, he was initially treated with intravenous Ampicillin and Cefotaxime, and later changed to intravenous Cloxacillin for total of two weeks, followed by further 4-week course of oral antibiotics. Repeated X-ray, non-contrast CT and blood investigations 4 weeks after commencement of antibiotics revealed normal findings. In addition, he was free of pain and limping had completely resolved. He was able to resume his normal day-to-day activities. At 6 months review, he was found to have normal function of right knee joint and was discharged from care. Figure 1 shows periosteal erosions of proximal anterior tibia and Figure 2 shows sclerotic changes which are suggestive of osteomyelitis.

Discussion

This report highlights the importance of revisiting initial diagnosis when clinical features persist despite initial management. Clinical features of rheumatic fever often overlap with suppurative arthritis and adjacent osteomyelitis¹⁰. During the first two weeks of the course of osteomyelitis, X-ray findings are often normal and repeating X-ray is crucial for children with persistent joint and bone pains managed in centers with limited



Figure 1: Periosteal erosions in proximal anterior tibia (X-ray image).



Figure 2: Sclerotic bone changes in proximal anterior tibia (CT image).

diagnostic facilities. Although ultrasound is helpful in early detection of septic arthritis, the value of ultrasound in diagnosing osteomyelitis is limited¹¹. A low index of suspicion of chronic osteomyelitis is one of the main reasons for its delayed diagnosis¹².

Osteomyelitis is seen more commonly in children with risk factors such as indwelling intravascular catheters, systemic diseases such as sickle cell anemia and chronic granulomatous disease¹³. Children who have had penetrating or puncture wounds are also at risk of direct inoculation of bacteria causing osteomyelitis¹⁴. The disease has been reported predominantly in male children¹⁵. In the absence of specific risk factors, the likely route of entry of infection in the reported child is

hematogenous.

In addition to clear radiological evidence of osteomyelitis, pus cultures yielded the etiological agent in this child. However, blood culture was negative. Studies have reported that cultures could be negative in up to 50% of cases with osteomyelitis¹⁶. This group of children had a longer duration of bone pain but excellent response to a course of empirical anti-staphylococcal antibiotics with better long term outcomes.

Acute osteomyelitis almost unvaryingly happens in children because of the rich blood supply to growing bones. Further, long bones are commonly involved in children compared to adults¹⁷. Microorganisms infect bone through one or more of three basic methods which are via the bloodstream (*haematogenous*), nearby areas of infection (as in cellulitis) and penetrating trauma. *Staphylococcus aureus* the most common organism causing osteomyelitis¹⁸. Other organisms include *Group B streptococci*, *Escherichia coli*, *Streptococcus pyogenes*, and *Haemophilus influenza*¹⁸. Gram-negative bacteria, including enteric bacteria, are significant pathogens in splenectomized patients and intravenous drug users¹⁹. Pus culture grew *Staphylococcus aureus* in our child which was sensitive to several antibiotics including cloxacillin.

Once the infection enters into metaphysis of the bone via the blood stream, leucocytes march to engulf the organism and lyse and form pus in the bone²⁰. Pus, in turn, tracts into blood vessels and impedes blood flow leading to formation of sequestra²⁰. Subsequently, body tries to form a new bone at the place of sequestra. When this process is chronic, organisms start exhibiting resistance to antibiotics²¹. The continuum of this pathological process leads to chronic disability²². The gold standard for diagnosis of chronic

osteomyelitis is considered to be supportive histopathological findings in bone biopsy and positive bone cultures²³. Magnetic resonance imaging (MRI) is superior to other radiological methods in soft tissue characterization in chronic osteomyelitis and is the imaging modality of choice²⁴. A high index of suspicion is crucial for early diagnosis and early treatment prevents complications of bone destruction. MRI and specific histopathological evaluation facilities were not available in current treatment facility; however, empirical treatment led to complete resolution of clinical and radiological features and physical disability.

Osteomyelitis frequently needs lengthy antibiotic therapy for several weeks or months with or without surgical debridement. Antibiotics are selected empirically by the patient's history and epidemiological pattern of common infective organisms. A number of centers suggest treatment for 6 weeks²⁵. Our child received 6 weeks treatment with surgical intervention.

Whilst chronic osteomyelitis is rare in the pediatric age group, early diagnosis could be challenging due to overlap with other rheumatological diseases and variability in clinical presentation. High index of suspicion is required for early diagnosis. Aggressive treatment with parenteral antibiotics and surgical debridement are likely to prevent long term physical disability.

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Ethical Approval Issue: Written consent has been obtained from index patient's parents.

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