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Case Report

A rare case of cystic tuberculosis osteomyelitis of the distal tibia in infancy: A case report

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Abstract

Tuberculosis can affect one-third of the musculoskeletal system, but isolated bone tuberculosis in infants is extremely rare, often presenting with non-specific symptoms that can lead to misdiagnosis. Cystic forms of tuberculosis can resemble various other pathological conditions Here, we present a case of tuberculosis osteomyelitis affecting the distal tibia in a nine-month-old infant who visited our outpatient department due to swelling and pain in the left ankle. The lesion was treated with curettage and debridement on two occasions. The patient was fitted with an under-knee splint for three months and received anti-tuberculosis therapy for twelve months. During a one-year follow-up, there were no signs of relapse, and no epiphyseal injury or deformity was observed. This case emphasizes the need to consider tuberculous osteomyelitis in the differential diagnosis of lytic lesions in the distal tibia metaphysis in infants.

Introduction

Isolated bone tuberculosis in children is exceedingly rare, typically affecting the metaphyses of the long bones in the lower extremities.[1] Diagnosing this condition is difficult due to non-specific clinical and radiological findings.[2] Isolated lesions may also mimic bacterial and fungal osteomyelitis, as well as simple and aneurysmal bone cysts, cartilage tumors, and osteoid osteoma. The literature contains no reports of patients under 11 months with isolated tuberculosis.[3] Here, a case of tuberculosis in the distal tibial metaphysis, a rare site for infants under one year of age was reported.

Case Report

A 9-month-old female presented with pain and a superficial wound on her left ankle, which had occurred two weeks ago due a physical injury after falling. The patient did not show any signs of fever, anorexia, weight loss, or cough.

During the physical examination, the left ankle exhibited swelling and tenderness, with painful movements. There was no evidence of skin involvement on the lower limb, and the neurovascular status of the lower limbs was NORMAL. X-rays indicated osteolysis affecting the metaphysis and epiphysis of the left distal tibia (Figure 1). Laboratory findings included a white blood cell count of 13.6×10^3, an erythrocyte sedimentation rate of 28 mm/h, and a C-reactive protein level of 13.5 mg/L, with normal lung radiography results.

Based on a preliminary diagnosis of acute osteomyelitis of the left distal tibia, debridement and curettage were performed. A bone biopsy revealed granulomatous inflammation without atypical cells. Microbiological analysis also identified an acid-fast bacillus through Ziehl-Neelsen staining, and *Mycobacterium tuberculosis* was cultured from the bone sample.

The infant had received a Bacillus Calmette-Guérin (BCG) vaccination at birth. Following the confirmation of distal tibia tuberculosis, anti-tuberculosis therapy, including isoniazid, rifampin, ethambutol, and pyrazinamide, was initiated. No familial history of tuberculosis was found in the patient or her family. The patient has been undergoing follow-up examinations every three months at our orthopedic clinics, with no relapses noted after one year. Follow-up assessments indicated full, pain-free ankle motion, and radiographs showed a smaller lesion compared to pre-treatment images (Figure 2).

Discussion

Infection with *M. tuberculosis* (MTB) can affect various

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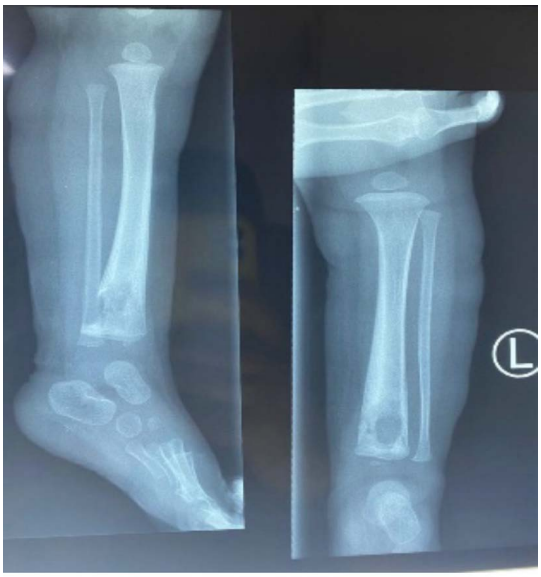

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Figure 1. Osteolytic lesion prior debridement and anti-tuberculosis treatment. **(a)** Lateral view of tibia and fibula x-ray. (b) AP view of left tibia and fibula X-ray

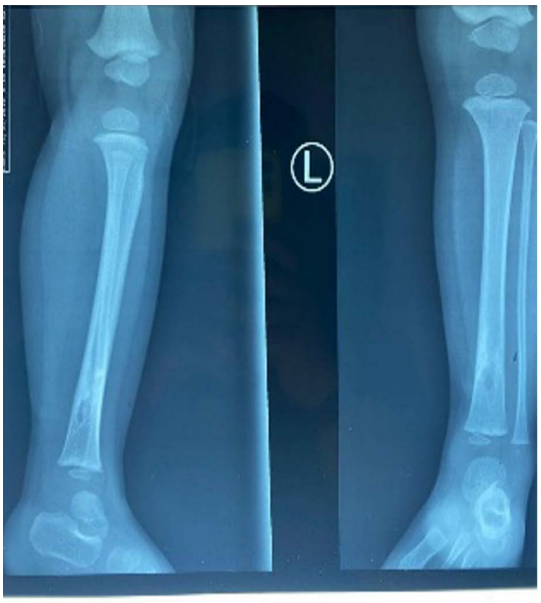

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Figure 2. Smaller lesion at 1 year follow-up. (a) Lateral view of left tibia and fibula x-ray. (b) AP view of left tibia fibula x-ray

organs and tissues. While 75% of cases are pulmonary, the musculoskeletal system is involved in only 1-3% of cases, with half of those affecting the vertebrae and 4-11% involving the foot and ankle.[4] Regarding bone and joint tuberculosis, a case series by Martini et al. reported that 11% of cases involved pediatric patients. In children, skeletal involvement is often multifocal and typically follows lung tuberculosis, often associated with an immune deficiency.[5] Isolated bone involvement in children is rare, highlighting the importance of lung radiography and family screening for patients with bone and joint issues. In this case, there was no evidence of tuberculosis in the lungs or elsewhere in the musculoskeletal system, and family members were also found to be tuberculosis-free.

Diagnosing bone tuberculosis in children poses challenges due to non-specific clinical and radiological signs that can lead to misdiagnosis. Symptoms such as pain, swelling, and limping are common, with atypical presentations including low-grade fever, weight loss, fatigue, local muscle wasting, sinus tracts, and pathological fractures.[6] Diagnostic evaluations typically include tuberculin skin tests, erythrocyte sedimentation rates, complete blood counts, and direct lung radiography. Initial imaging may reveal soft tissue swelling, while advanced stages present with multiple lytic, oval cysts extending from the metaphysis to the epiphysis, often accompanied by osteoporosis.[7] Bone lesions in tuberculosis osteomyelitis can be categorized into four types: cystic, infiltrative, regional erosion, and spina ventosa, with our patient's lesion classified as cystic.[8] Bone involvement carries a risk of injury and pathological fractures, necessitating a differential diagnosis that includes bacterial and fungal osteomyelitis as well as bone tumors causing lytic lesions; thus, biopsy is warranted in suspicious cases. In our patient, a frozen section was performed to rule out potential tumoral lesions.[3]

Histological examination revealing granulomatous inflammation supports the diagnosis of tuberculosis. Ziehl-Neelsen staining can also identify acid-fast bacilli, although detection rates in tuberculosis osteomyelitis are low, resulting in a high false-negative rate. Growing *M. tuberculosis* in culture remains the definitive diagnostic method, though culture results can take time, delaying treatment. Polymerase chain reaction (PCR) tests also suggest a faster diagnosis but may also produce false-negative results. In this case, the diagnosis was confirmed through the detection of acid-fast bacilli with Ziehl-Neelsen staining and *M. tuberculosis* culture, along with granulomatous inflammation observed in the pathological tissue.

Treatment for bone tuberculosis typically involves curettage of the lesion and the administration of anti-tuberculosis medications. Large defects resulting from the curettage of cystic lesions are at increased risk of pathological fractures, necessitating careful and gentle surgical techniques to avoid permanent damage. While there have been reports of filling defects with bone grafts, this approach is generally discouraged.[9] In cases where continuity of the bone structure is preserved, the application of a cast may help reduce fracture risk. Standard treatment regimens include isoniazid, rifampin, ethambutol, and pyrazinamide, with the first-line combination of isoniazid and rifampin. Approximately 85% of patients with bone tuberculosis can be effectively treated within 9-10 months; however, rapid progression may require surgical intervention. Recurrences can occur

even after medical treatment, necessitating potential surgical repeat.[10] In this patient, no recurrence was observed, and no graft was placed following curettage, as the infant was not yet ambulating. Anti-tuberculosis treatment was continued for one year.

Conclusion

This case report underscores the necessity of considering bone tuberculosis in the differential diagnosis of lytic lesions in the metaphyses of long bones in infants. Attention should be given to the patient's history of travel or exposure to regions with high tuberculosis prevalence, such as Sabah, which accounted for 20% of Malaysia's tuberculosis notifications between 2012 and 2018 despite representing only 10% of the population.

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Authors' Contribution

Conceptualization: Anis Anwar Zahari, Abdul Qayyum, Norazian Kamisan, Norhaslinda Bahaudin.
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Competing Interests

No conflicts of interest declared by the authors.

Ethical Approval

This study was approved by the Ethics Committee of Hospital Tuanku Ja'afar, Seremban Malaysia. Consent was explained and obtained from the subject no HTJ43151. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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