Cystic tuberculosis osteomyelitis of the distal tibia in infancy

Seyit Ali Gümüştas, Talat Çağırılmaz, Mehmet Müfit Orak, Gökhan Pehlivanoglu, Sedat Öktem

Clinic of Orthopaedics and Traumatology, Kartal Dr. Lütfi Kırdar Education and Research Hospital, İstanbul, Turkey

Abstract

One-third of tuberculosis cases affect the musculoskeletal system. Solitary bone tuberculosis is a rare condition in infancy, has non-specific findings, and can be misdiagnosed easily. Cystic form tuberculosis may mimic many other pathologic conditions. In our case report, we present tuberculosis osteomyelitis of the distal tibia in a baby aged ten months who visited our outpatient department with swelling and pain in their left ankle. Curettage and debridement was performed twice for the lesion. An under-knee splint was applied for 3 months and anti-tuberculosis treatment was given for 12 months. There was no relapse during a five-year follow-up period. There was no epiphysis injury and deformity. In this case report, we discuss that bone tuberculosis, as a rare condition, must be considered in lytic lesions of the distal tibia metaphysis in infancy. (Turk Pediatri Ars 2017; 52: 53-6)

Keywords: Infancy, tibia, tuberculosis osteomyelitis

Introduction

Solitary bone tuberculosis is seen substantially rarely in children (1-4). It frequently affects the metaphyses of the long bones in the lower extremities. The diagnosis is difficult because the clinical and radiologic findings are not specific (5). Solitary lesions may mimic osteomyelitis related with bacteriae and fungi, simple and aneurismal bone cysts, cartilage tumors, and osteoid osteoma (2). In the literature, there is no patient younger than 11 months with solitary tbc (6). Here, we present a case of tuberculosis of the distal tibial metaphysis which is a rare localization in infants aged younger than 1 year.

Case

A 10-month-old male patient presented to our Emergency Outpatient Clinic with pain and swelling in the left ankle, which he had had for the last 15 days. Physical examination revealed edema and tenderness in the left ankle. Ankle movements were limited and painful. Direct radiography revealed osteolysis involving the metaphysis and epiphysis in the distal tibia (Figure 1).

The laboratory findings were as follows: white blood cells (WBC): 13.6×10^3 , erythrocyte sedimentation rate: 28 mm/h, C-reactive protein: 13.5 mg/L. Lung radiography was found normal. The patient was underwent surgery with a prediagnosis of acute osteomyelitis. A frozen biopsy was performed; atypical cells were not observed and granulomatous inflammation was found. Debridement and irrigation was performed. Acid-resistant bacterium was observed with Ziehl-Neelsen staining in microbiologic examination of the samples obtained. *Mycobacterium tuberculosis* was grown in culture. Caseification necrosis was observed on histopathologic evaluation. A tuberculin skin test was not performed.

Additional investigations in terms of immune deficiency were not performed because the patient did not have recurrent pneumonia, otitis, sinusitis, diarrhea, organ and intramuscular abscess, familial history of immune deficiency or laboratory findings such as lymphopenia, which could suggest immune deficiency.

A Bacillus Calmette-Guérin (BCG) vaccination had been administered when the patient was 2 months old. Qua-

Address for Correspondence: Talat Çağırılmaz E-mail: talatcagirmaz@gmail.com

Received: 05.12.2014

Accepted: 19.10.2015

©Copyright 2017 by Turkish Pediatric Association - Available online at www.turkpediatriarsivi.com

DOI: 10.5152/TurkPediatriArs.2017.2479

Figure 1. a, b. Postero-anterior and lateral radiographs of the ankle at the time of the first presentation

Figure 2. a, b. Clinical appearance at the end of the fifth year

druple tuberculosis treatment (isoniasid, rifampin, ethambutol, and pyrazinamide) was initiated for the first two months. The treatment was completed with isoniasid and rifampin for 10 months. Investigations performed revealed no lung tuberculosis in the patient or in the patient's family. Infection recurred in the sixth week after surgery and curettage and debridement was performed once again. A culture of the samples obtained in the second operation was negative. Pathologic examina-

tion revealed granulomatous inflammation findings. An under-knee splint was applied to the patient for a total period of three months as from the first operation. Relapse was not observed in the follow-up. The laboratory values returned to normal in the fourth month.

The patient is being examined at follow-up visits by the Department of Pediatric Chest Diseases and in Orthopedic Clinics once a year. Relapse was not found in the follow-up in the fifth year. The laboratory findings were found as normal. The ankle range of motion was full and painless (Figure 2). Direct radiography revealed no deformity or involvement of the epiphysis (Figure 3). The X-rays taken in the seventh year revealed no abnormal findings, shortness or deformity (Figure 4). The family was informed that the patient was going to be reported in a case presentation and verbal informed consent was obtained.

Discussion

Tuberculosis infection caused by *Mycobacterium tuberculosis* may affect all organs and tissues (7). Lung localization is observed in 75% of cases and infection occurs in the musculo-skeletal system in only 1-3% of cases (8). The vertebrae are involved in 50% of cases of musculo-skeletal system tuberculosis and the foot-ankle is involved in only 4-11% (8, 9).

In the bone and joint tuberculosis series of Martini et al. (9), which included 652 patients, 11% of the subjects were in the pediatric age group. Skeletal system involvement in children is generally observed in multiple localizations and develops secondary to lung tuberculosis in association with immune deficiency. Solitary bone involvement is found rarely in children. Therefore, lung radiography and family screening are recommended in patients with bone and joint involvement (1-5). Immune deficiency was not found in our patient.

Figure 3. a, b. Postero-anterior and lateral radiographs of the ankle at the end of the fifth year

Figure 4. a, b. Postero-anterior and lateral radiographs of the ankle at the end of the seventh year

In addition, no other tuberculosis focus was found in the lungs or musculoskeletal system. Tuberculosis infection was not found in the family members.

It is difficult to confirm the diagnosis of bone tuberculosis in children. The diagnosis can be easily missed in the early stage because the clinical and radiologic find-

ings are not specific (4). The most common symptoms include pain, swelling, and limping (1-7). The other rare findings include mild fever, weight loss, weakness, muscle atrophy around the joint, sinus tract, and pathologic fracture (3, 7). The assistive diagnostic tests include a tuberculin skin test, erythrocyte sedimentation rate, complete blood count, and direct lung radiography (1). Only soft tissue swelling may be found on direct radiography of the involved region in the early stage. In the late stage, multiple lytic, oval cysts with lobulated borders extending from the metaphysis to the epiphysis are typical and osteoporosis accompanies frequently (1-3, 6, 7). Bone lesions are observed as 4 types in tuberculosis osteomyelitis: cystic, infiltrative, regional erosion, and spina ventosa (3). In our patient, bone involvement was observed as cystic type. Bone involvement carries a risk for bone injury and pathologic fracture. In the differential diagnosis, osteomyelitis caused by bacteriae and fungi and bone tumors causing lytic lesion should be investigated. Therefore, biopsy is recommended in suspicious cases (2). Frozen section was performed in our patient and atypical cells were investigated. In this way, we tried to exclude possible lytic tumoral lesions.

Granulomatous inflammation found in a histologic examination supports the diagnosis (7). Acid-resistant bacterium may be observed with Ziehl-Neelsen staining. Acid-resistant bacterium is found with a low rate in tuberculosis osteomyelitis, and the rate of false-negative results is high. Growth of *M. tuberculosis* in culture is diagnostic (7). However, treatment may be delayed because *M. tuberculosis* culture takes a long time. For a faster diagnosis, polymerase chain reaction (PCR), which is a nucleic acid reproduction method, may be used, but one should be careful about false-negative results (10). In our case, the diagnosis was made by observation of acid-resistant bacterium with Ziehl-Neelsen staining and growth of *M. tuberculosis* in culture. In addition, the diagnosis was supported with the finding of granulomatous inflammation on the pathologic tissue sample.

Treatment of bone tuberculosis involves curettage of the lesion and administration of anti-tuberculosis drugs. Bone defects that form after curettage of cystic lesions pose a risk for pathologic fracture. In addition, curettage should be performed carefully and gently so that no permanent physical damage occurs (7). Although cases in which defects were filled with bone graft have been reported, this application is not recommended (1-3, 7, 9). The risk of fracture should be reduced by applying a cast in cases in which continuity in bone anatomy is present. Isoniazid, rifampin, ethambutol, streptomycin and pyrazinamide are used in the treatment of tuberculosis.

Isoniazid and rifampin constitute the first-line combination (1-3, 6, 10). Eighty-five percent of patients with bone tuberculosis can be treated with medical treatment for 9-12 months, but curettage and debridement should be performed in rapidly progressing cases. Recurrence may be observed despite drug treatment and surgical intervention may be needed again (7). Recurrence occurred 6 weeks after the first surgical intervention in our patient and a second operation was performed. A graft was not placed in the cavity formed after curettage and a cast was applied for three months. In addition, anti-tuberculosis drug treatment was continued for one year.

This case report serves to remind that bone tuberculosis should be considered in lytic lesions observed in the metaphyses of the long bones in infancy.

Informed Consent: Verbal informed consent was obtained from patient's parents, who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - S.G., T.Ç.; Design - S.G., M.O.; Supervision - M.O., S.Ö.; Funding - S.G., T.Ç., G.P.; Materials - T.Ç., G.P.; Data Collection and/or Processing - S.G., G.P.; Analysis and/or Interpretation - S.G., M.O.; Literature Review - S.G., T.Ç., S.Ö.; Writing - S.G., T.Ç.; Critical Review - M.O., S.Ö.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

References

- Shih HN, Hsu RW, Lin TY. Tuberculosis of the long bone in children. Clin Orthop Relat Res 1997; 335: 246-52.
- Rasool MN, Govender S, Naidoo KS. Cystic tuberculosis of bone in children. J Bone Joint Surg Br 1994; 76: 113-7.
- Rasool MN. Osseous manifestations of tuberculosis in children. J Pediatr Orthop 2001; 21: 749-55. [\[CrossRef\]](#)
- Martini M, Adjrad A, Boudjemaa A. Tuberculous osteomyelitis. A review of 125 cases. Int Orthop 1986; 10: 201-7. [\[CrossRef\]](#)
- Agarwal A, Qureshi NA, Khan SA, Kumar P, Samaiya S. Tuberculosis of the foot and ankle in children. J Orthop Surg (Hong Kong) 2011; 19: 213-7. [\[CrossRef\]](#)
- Malik S, Joshi S, Tank JS. Cystic bone tuberculosis in children-a case series. Indian J Tuberc 2009; 56: 220-4.
- Kao HK, Yang WE, Shih HN, Chang CH. Physeal change after tuberculous osteomyelitis of the long bone in children. Chang Gung Med J 2010; 33: 453-60.
- Alvarez S, McCabe WR. Extrapulmonary tuberculosis revisited: a review of experience at Boston City and other hospitals. Medicine 1984; 63: 25-55. [\[CrossRef\]](#)
- Martini M. Tuberculosis of the bones and joints. Berlin: Springer-Verlag; 1988. [\[CrossRef\]](#)
- Sağlık Bakanlığı, Tüberküloz Tanı ve Tedavi Rehberi, Çocukluk Çağı Tüberkülozu. 2011; 71-84.