

# Widespread Skeletal Tuberculosis in a two years old Girl

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## Introduction

Presentations of tuberculosis are protean and are not limited to the pulmonary disease alone. The spectrum of pediatric tuberculosis includes asymptomatic primary infection, progressive pulmonary TB and extrapulmonary disease including lymphadenitis, meningitis and osteomyelitis. Skeletal TB occurs in about 1-3% and in some studies accounts for 1-6% of the extrapulmonary cases.<sup>1-3</sup> Any part of the skeletal system can be involved but in order of frequency the sites most commonly involved are the spine, femur, tibia and fibula. The involvement of spine accounts for about 50 - 65% of the cases and the knee and hip joint involvement each account for 15% of cases.<sup>2</sup> Involvement of small bones of hand and feet leading to tuberculous dactylitis is a rare presentation of TB. Even rarer is the association of tuberculous dactylitis with tuberculous osteomyelitis of multiple bones in a single patient. We are reporting a rare case of a young child with disseminated skeletal tuberculosis along with tuberculous dactylitis. It is to emphasize how a confusing clinical and radiological picture may lead to the diagnostic and therapeutic delay and result in sequelae. This can be avoided if clinicians working in communities, where tuberculosis is common have an awareness that tuberculosis can present in least expected sites and in most unusual combinations. A high index of suspicion therefore, is the key to timely diagnosis and treatment.

## Case Report

A two years old female child vaccinated for TB, resident of Sargodha presented with the complaints of fever and multiple bony swellings with discharging pus for the last nine months. Swellings involved multiple fingers, both elbows and feet. There was a history of weight loss and night sweats. There was no history of contact with a known tuberculous patient although grandfather was diagnosed and treated for pulmonary tuberculosis 10 years ago, much before the little girl was born. She belonged to a low socio-economic strata and was living in a crowded house. She had been to many general practitioners, district hospitals and private clinics and was repeatedly diagnosed as osteomyelitis and received multiple courses of oral as well as injectible antibiotics ranging from amoxicillin to 3rd generation cephalosporins. The swellings showed no improvement and continued to discharge pus. Over a period of time new swellings continued to appear with eventual formation of sinuses.

On examination she was afebrile, with height and weight below 10th percentile. Local examination revealed multiple small swellings with discharging sinuses on left index and little fingers, right metatarsal bone, both elbows

and metacarpal bones. There were signs of inflammation with restricted movements. Rest of the examination was unremarkable. She was admitted with the provisional diagnosis of chronic osteomyelitis with the possibility of an underlying immunodeficiency.

On investigations complete blood picture showed a high total leucocyte count of 18000/mm<sup>3</sup> with normal breakup. There was hypochromic microcytic anemia. Erythrocyte sedimentation rate was elevated to 79mm in first hour. Mantoux was positive with 10 x 10 mm induration. X-rays chest and spine, CSF study, immunoglobulins and nitroblue tetrazolium test (NBT) were all within normal range. Skeletal survey showed multiple lytic lesions with varying degree of sclerosis and expansion in many bones. Epiphysis of left ulna, right humerus, metaphalangeal region of right radius, third metatarsal bone and bones of hands and feet had significant expansion (spina ventosi) with extensive destruction of metacarpals.

A diagnostic incision and drainage was done and material sent for ZN stain, culture and histopathology. AFB stain turned out to be negative. Histopathology showed granulomatous inflammation consistent with TB.

The patient was diagnosed as having widespread tuberculous osteomyelitis involving multiple bones. Chemotherapy for tuberculosis was started. Four drugs regimen with isoniazid, rifampicin, pyrazinamide and streptomycin was planned for first two months. She was discharged and then followed up. Her swellings completely disappeared and her sinuses healed within two months. After two months she was advised to continue with isoniazid and rifampicin for another 10 months.

## Discussion

Tuberculosis has been a problem for humans since antiquity. A reemergence of TB is occurring in the world including developing and underdeveloped countries.<sup>4</sup> An estimated 1.3 million cases of TB and 450,000 associated deaths occur annually in children and extrapulmonary TB accounts for up to one third of all newly diagnosed cases of tuberculosis.<sup>5</sup> Disseminated bone tuberculosis and dactylitis is very rare but may occur in those parts of the world where TB is endemic.<sup>6-12</sup> In Nelson R. Mandela School of Medicine 42 children were diagnosed to have TB osteomyelitis from 1984-1999 and only 5 children had multifocal bone involvement.<sup>8</sup> Department of radiology from King Edward Memorial Hospital, Mumbai, presented radiological findings of 4 children diagnosed to have tuberculous osteomyelitis involving several bones over a period of nine months and only one child presented with involvement of a metacarpal bone.<sup>13</sup> In another study in Ethiopia out of 41 children having bone and joint involvement, metatarsals

Medicine 42 children were diagnosed to have TB osteomyelitis from 1984-1999 and only 5 children had multifocal bone involvement.<sup>8</sup> Department of radiology from King Edward Memorial Hospital, Mumbai, presented radiological findings of 4 children diagnosed to have tuberculous osteomyelitis involving several bones over a period of nine months and only one child presented with involvement of a metacarpal bone.<sup>13</sup> In another study in Ethiopia out of 41 children having bone and joint involvement, metatarsals were involved in two children and only one child had phalangeal involvement.

The clinical picture caused by mycobacterium tuberculosis may simulate many different diseases. This can result in delay in diagnosis and institution of chemotherapy. Multiple unwanted investigations are performed and serious consequences are faced by the patients.<sup>14</sup> The spread of infection to skeletal system is either through haematogenous dissemination or through lymphatic drainage. Radiographic evidence of the primary pulmonary focus is present only in about 16-50 % of the cases.<sup>15</sup>

Tuberculous dactylitis or spina ventosi occur in children under five years of age and is most common in infants.<sup>16</sup> The metacarpal, metatarsal or phalanges may be affected. Hands as compared to feet are more frequently involved. Disease may involve several digits, which become swollen, fusiform or spindle shaped usually with little pain. A cold abscess may form and drain spontaneously.

Diagnosis of skeletal tuberculosis should be considered in any child who has a positive Mantoux test, evidence of present or past TB, history of contact with a tuberculous patient and any child having persistent, otherwise unexplained bone or joint lesion.<sup>16</sup> Radiological changes are not pathognomic. Findings range from soft tissue swelling, periostitis with bone expansion and cortical destruction, diffuse uniform infiltration and localized osteitis. In the early stages, when plain radiographs are normal, CT scan or an MRI may be helpful in localizing the lesion.

No single test of synovial fluid is diagnostic of tuberculosis except for a positive AFB smear or culture. As the disease is paucibacillary, a positive AFB smear is rare, so the diagnosis usually is confirmed by obtaining granulomatous tissue on biopsy.<sup>6,8,17-19</sup> Other investigations should include ESR, X-ray chest, X-ray spine and CSF study to rule out any concomitant extraskelatal lesion.

Treatment includes both chemotherapy and orthopedic intervention. The orthopedic procedures can be used for several purposes such as diagnosis, evacuation of caseum and necrotic bone, immobilization of joint, and reconstruction or strengthening of damaged bone.<sup>1,4,7</sup>

Antituberculous drugs are the mainstay of treatment. Two months of isoniazid, rifampacin, pyrazinamide and streptomycin once daily is followed by 7-10 months of isoniazid and rifampacin once daily or twice a week.

The prognosis for recovery without deformity is significantly good provided a timely diagnosis is made and proper chemotherapy initiated before the development of complications.<sup>18</sup>

## References

1. Dhillon MS, Sharma S, Gill SS, et al. Tuberculosis of bones and joints of the foot: an analysis of 22 cases. *Foot Ankle* 1993;14:505-13.
2. Jesus GV, Lydia T. Tuberculous osteomyelitis of the long bones in children. *Pediatr Infect Dis J* 1995;14:542-6.
3. Bohl JM, Janner D. Mycobacterium tuberculosis sternal osteomyelitis presenting as anterior chest wall mass. *Pediatr Infect Dis J* 1999;18:1028-9.
4. Wessels G, Hesselting PB, Beyers N. Skeletal tuberculosis: dactylitis and involvement of the skull. *Pediatr Radiol* 1998;28:234-6.
5. Maltezou HC, Spyridis P, Kafetzis DA. Extra-pulmonary tuberculosis in children. *Arch Dis Child* 2000;83:342-6.
6. Urovitz EP. Tuberculous dactylitis: a rare entity. *Can J Surg* 1982;25:698-90.
7. Clarke JA. Tuberculous dactylitis in childhood. The need for continued vigilance. *Clin Radiol* 1990;42:287-8.
8. Rasool MN. Osseous manifestations of tuberculosis in children. *J Pediatr Orthop* 2001;21:749-55.
9. Schaaf HS, Donald PR. Radiological case of the month: multiple bone tuberculosis and dactylitis. *Arch Pediatr Adolesc Med* 2000;154:1059-60.
10. Zoga A, Lee VW. Pediatric case of the day. Tuberculosis dactylitis and primary pulmonary Tuberculosis. *Am J Roentgenol* 1999;173:815-17.
11. Jensen CM, Jensen CH, Paerregaard A. A diagnostic problem in tuberculous dactylitis. *J Hand Surg* 1991;16:202-3.
12. Negusse W. Bone and joint tuberculosis in childhood in a children's hospital, Addis Ababa; *Ethiop Med J* 1993;31:51-61.
13. Morris BS, Varma R, Garg A, et al. Multifocal musculoskeletal tuberculosis in children: appearances on computed tomography. *Skeletal Radiol* 2002;31:1-8.
14. Martini M, Adjrad A, Boudjemaa A. Tuberculous osteomyelitis. A review of 125 cases. *Int Orthop* 1986;10:201-7.
15. Singh SB, Saraf SK, Singh LI, et al. Osteoarticular tuberculosis in children. *Indian Pediatr* 1992;29:1133-7.
16. Otachbjian M. Tuberculosis of bone, pediatric orthopedic, 2nd ed. Philadelphia: Saunders,1990, pp. 1127-8.
17. Vohra R, Kang HS, Dogra S, et al. Tuberculous osteomyelitis. *Br J Bone Surg* 1997;79:562-6.
18. Dhillon MS, Nagi ON. Tuberculosis of the foot and ankle. *Clin Orthop* 2002;398:107-13.
19. Chen SC, Huang SC, Wu CT. Nonspinal tuberculous osteomyelitis in children. *J Formos Med Assoc* 1998;97:26-31.