Case Report

Tuberculous dactylitis (spina ventosa) with concomitant ipsilateral axillary scrofuloderma in an immunocompetent child: A rare presentation of skeletal tuberculosis

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Abstract

Tuberculous dactylitis is a distinctly uncommon, yet well recognized form of tuberculosis involving the small bones of the hand or foot. It occurs in young children in endemic areas under 5 years of age. Tuberculosis of the short tubular bones like phalanges, metacarpals or metatarsals is quite uncommon beyond 6 years of age, once the epiphyseal centers are well established. The radiographic features of cystic expansion have led to the name "*Spina Ventosa*" for tuberculous dactylitis of the short bones. Scrofuloderma is a mycobacterial infection affecting children and young adults, representing direct extension of tuberculosis into the skin from underlying structures e.g. lymph nodes. An 8-year-old malnourished girl had multiple axillary ulcers with lymphadenopathy. Tuberculous dactylitis with ipsilateral axillary scrofuloderma was suspected on clinical and radiological grounds. The suspicion was confirmed by histology and bacteriology. The patient responded to antitubercular drugs with progressive healing of the lesions without surgery. Concomitant presence of these dual lesions suggesting active disseminated tuberculosis in immune-competent child over 6 years is very rare and hardly reported.

Key Words: Tuberculosis, cutaneous, hand, lymphadenitis axilla, osteoarticular tuberculosis, immunocompetence,

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INTRODUCTION

Skeletal tuberculosis accounts for only 1 to 5% of all tuberculous infections and roughly, half of them occurs in the spinal column.[1-3] Tuberculous dactylitis is a form of secondary, extrapulmonary tuberculosis involving the small bones of the hand or foot; distinctly rare in adult,[4] but it is a well recognized form of tubercular osteomyelitis in young children in endemic areas. However, even in the pediatric age, tuberculosis of the short tubular bones like phalanges, metacarpals or metatarsals is quite uncommon after the age of 5 years, once the epiphyseal centers are well established.[5] Radiographic features of cystic expansion of the short tubular bones has led to the name of "*Spina Ventosa*" for tuberculous dactylitis of the short bones.[6] Scrofuloderma (SCF) on the other hand is a tuberculous or nontuberculous mycobacterial infection affecting children and young

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adults, representing direct extension of tuberculosis into the skin from underlying structures such as the lymph nodes (especially the cervical), bone, or lung, or by contact exposure to tuberculosis.[7,8] Here a case involving an 8-year-old destitute girl having tuberculous dactylitis of hand with ipsilateral scrofuloderma in axilla is described; to the best of our knowledge, this combination in immune-competent children over 5 years is very rare and hardly reported in the literature.

CASE REPORT

An 8-year-old destitute girl was brought by members of an internationally acclaimed humanitarian organization, with complaints of pain and swelling in the left index finger of five month's duration. On examination, she had features of a draining abscess on the dorso-ulnar aspect of the proximal phalanx of the left index finger [Figure 1]. On detailed medical history, she admitted having low-grade fever and significant weight loss over the last four months. Her general examination revealed emaciation with pallor and three small scantily draining axillary ulcers with accompanying axillary lymphadenopathy and an additional satellite ulcer in the midaxillary line near the anterior axillary fold [Figure 2]. Rest of her general and systemic examination was unremarkable. The radiograph showed features of osteomyelitis in the proximal phalanx distal to the physis [Figure 3]. Chest X-ray was normal but tuberculin test was strongly positive about 12 mm. Sedimentation rate was 73 mm/first hour. Aspirate from the small abscess around the sinus of the index finger showed no acid fast bacilli (AFB) on Ziehl Neelsen (ZN) staining and no pyogenic bacteria grew on culture. Fine needle aspirate from the ipsilateral axillary lymph node however, showed granulomatous inflammation with necrosis and additional slide subsequently came positive for AFB on ZN staining. Abdominal ultrasonography was normal. A diagnosis of tuberculous osteomyelitis of the second proximal phalanx, with disseminated tuberculosis in the form of axillary (lymph node origin) scrofuloderma was made. The patient was started on a four drug regimen (isoniazid + rifampicin + pyrazinamide + ethambutol for the first two months followed by isoniazid + rifampin for a total of seven months) as per DOT guidelines. Attention was also given to improve on her nutrition including high protein diet with zinc, calcium, vitamin D supplements and hematinics. The patient was given no splint immobilization for the index finger and active range of motion exercises was encouraged early. The patient was doing better with progressive healing of the bony lesion without any surgery [Figure 4]. The axillary scrofulodermic sinuses were healing, as well and patient was consistently gaining weight and hemoglobin level over months. We have not come across any literature showing concomitant presence of tuberculous dactylitis,

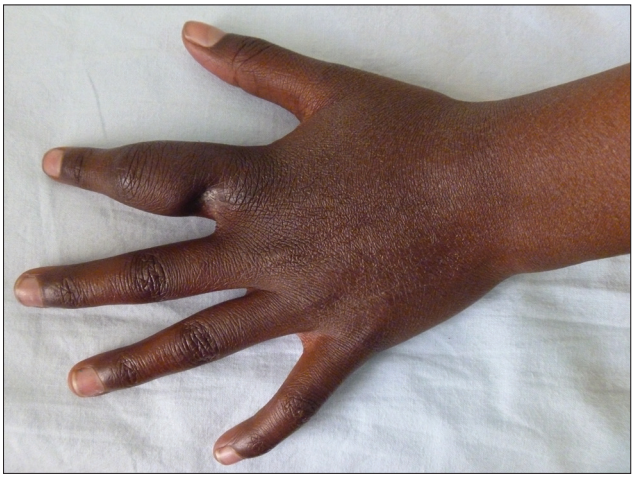

Figure 1: Fusiform swelling of tubercular dactylitis: Note the healing sinus

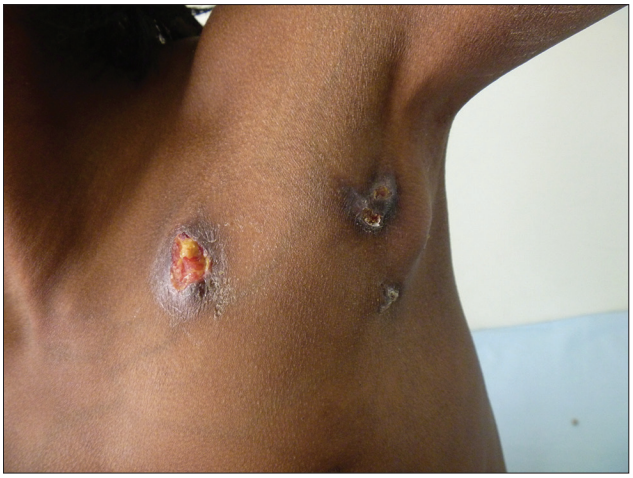

Figure 2: Axillary scrofuloderma with satellite lesion in anterior axillary fold

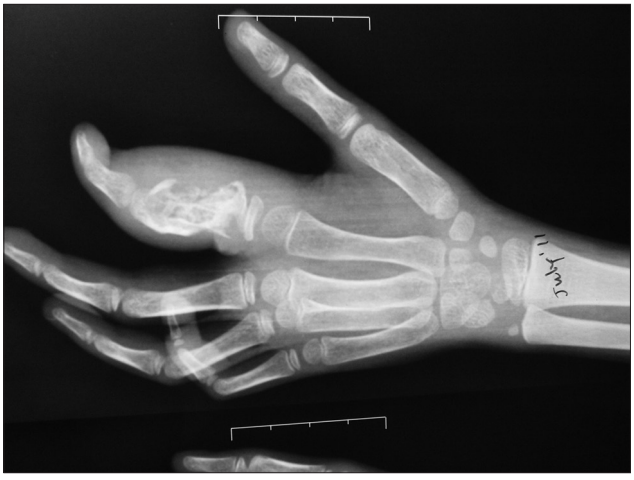

Figure 3: Radiological features: Lytic destruction distal to proximal phalangeal physis

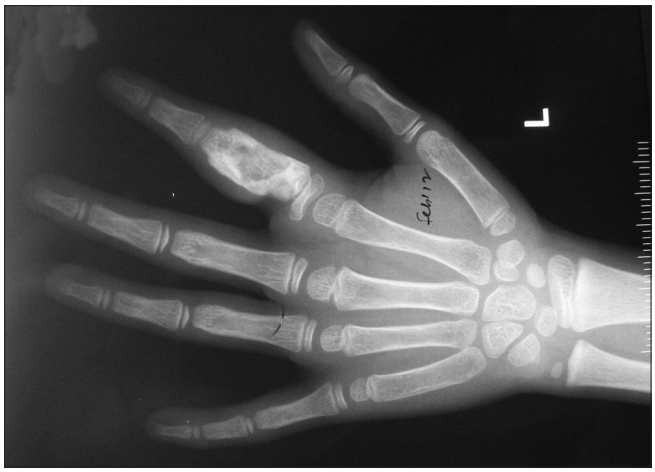

Figure 4: Progressive radiological healing after 6 month of ATT

axillary scrofuloderma suggesting active disseminated tuberculosis in an immunocompetent child above 6 years age.

DISCUSSION

Most tubercular infections of the bones are caused by the human strain of mycobacterium tuberculosis. Infection of the musculoskeletal system is a secondary disease caused by hematogenous spread from a primary lesion; it may occur shortly after the primary infection or years later as a disease reactivation. Osteoarticular involvement occurs in 1 to 3% of patients with extrapulmonary tuberculosis[2] and spine represents 50% of these lesions.[1,3,9] Tuberculous involvements of the metacarpals and phalanges is a rare presentation of extrapulmonary tuberculosis, especially in adults.[4] A whopping majority of 85% of patients with tubercular dactylitis are younger than 6 years of age.[10] The short bones of the hand are the most frequent location of skeletal tuberculosis in infancy and early childhood before the epiphyseal centers are well established. At this tender age, the hematopoietic marrow in those bones offers a fertile field for hematogenous bacterial implants; pulmonary lesions usually can be demonstrated. The infection rapidly involves the entire marrow space. Tuberculous granulation tissue expands the relatively soft cortex as it is resorbed or infarcted by the underlying process. The resultant fusiform expansion of the bone with thinned cortex and relatively radiolucent marrow space due to trabecular destruction resembles an inflated balloon. Expansion of the bone with cystic quality is termed *spina ventosa*.

Radiological features are well described, and findings are mention in published papers.[11] The affected bone appears expanded with lytic lesions in the middle (distal to the physis of proximal phalanx as seen in present case) and subperiosteal new bone formation along the involved bone. The cavity may contain soft coke like sequestra. Other findings on plain radiographs include osteopenia, soft-tissue swelling with minimal periosteal reaction, cysts in bone adjacent to joint and subchondral erosions. Typically, there is no periosteal layering or thickening, and sequestration ordinarily does not occur. In natural course, the disease may heal with shortening of the involved bone and deformity of the neighboring joint. Diaphyseal lesions tend to respond to therapy with slow healing. The bone density returns with slow filling of defects by new bone, which may become sclerotic, coarsely trabeculated, or relatively normal. The non-specific nature of these radiographic findings may potentially delay the diagnosis when spina ventosa is the lone clinical presentation.

Also called 'tuberculosis colliquativa cutis', scrofuloderma is manifested by the development of painless subcutaneous swellings that evolve into cold abscesses, multiple ulcers, and draining sinus tracts.[7,8] Initially, in SCF there are firm painless subcutaneous nodules that gradually enlarge and suppurate. The overlying skin breaks down to form an undermined ulcer with a granulating tissue at the base. This leads to ulcers and sinus tracts with undermined edges and ultimately puckered scars. Progression of the disease often leads to irregular adherent masses, densely fibrous at some places while fluctuant and discharging at others. It heals with a characteristic puckered scarring at the site of infection [Figure 2]. Diagnosis is usually performed by needle aspiration biopsy or excision biopsy of the lesion or node and the microbiological demonstration of stainable acid fast bacteria. In in our case, swellings with draining sinuses, histopathology report, strongly positive tuberculin test and a response to DOTs regime of ATT favored the diagnosis of tubercular scrofuloderma with tubercular dactylitis. In an epidemiological study conducted in south India in a Medical College Hospital, over a period of 7 years a total of 38 patients with scrofuloderma, the majority of cases presented with discharging sinuses overlying caseating tuberculous lymph nodes of the cervical nodes, followed by inguinal, axillary and submandibular groups, respectively.[12] There were only two cases of SCF with concomitant tuberculous synovitis out of which one was that of the thumb. The youngest female was 8-year-old and most cases occurred in the second decade (Mean age: 31 years).[12] As mentioned earlier, spina ventosa is seen in children, mainly between the ages of 1 and 3 years. Our patient was also just 8-year-old; again showing the rarity of our case to have axillary SCF with concomitant spina ventosa.

The gold standard for the diagnosis of osseous tuberculosis remains the positive culture of mycobacterium tuberculosis from bone tissue.[11] Differential diagnosis worth considering in such cases includes pyogenic osteomyelitis, Brodie's abscess, Atypical mycobacterial infection (due to mycobacterium scrofulaceum and M. avium-intracellulare), Actinomycosis or other deep mycosis (Sporotrichosis, Botryomycosis, Nocardiosis etc), Kaposi's sarcoma and luetic dactylitis.[13] There is recent published report of diagnostic dilemma and radical excision of such lesion as tumor in a young 2-year-old child.[14] Presence of draining sinus in the tuberculous osteomyelitis may hinder its distinction from pyogenic disease. Clinically, pyogenic osteomyelitis tends to be acutely painful, swollen, and hot with fever. Tuberculous osteomyelitis is relatively benign with mild pain and minimal pyrexia. The diffuse osteopenia associated with tuberculous infection may help distinguish it from pyogenic infection, as also the absence of sequestration. In syphilis, the bone is thickened by periosteal reaction. The definitive diagnosis relies on the detection M. tuberculosis by PCR or culture.[15] Tuberculin test may provide useful supportive evidence in difficult cases.[1] The optimal treatment duration for tubercular dactylitis remains unknown. Surgery has a limited role in the treatment in general but may play a supportive role, and curettage of the cavity has been recommended for avascular lesions.[15]

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