Isolated Spina Ventosa: An Extremely Rare Presentation of Skeletal Tuberculosis in Adolescent Age

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Abstracts: Even though the prevalence of tuberculosis is very high in the developing world, skeletal tuberculosis of extremities rarely occurs. Here we report an uncommon form of bone tuberculosis in a 16 year old female presenting as tubercular dactylitis of proximal phalanx of right middle finger confirmed by Histopathology.[Agrawal A NJIRM 2014; 5(5):111-113]

Key Words: Tuberculosis, Dactylitis, Skeletal

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Introduction: Rankin in 1886 first identified tuberculous dactylitis by histological technique[1]. Phalangeal tuberculosis is quite uncommon form of skeletal tuberculosis and rarely appears after the age of 5 years[2]. It is also known as spina ventosa (literally meaning 'short bones inflated with air') due to its radiographic feature of cystic expansion of short tubular bone[3]. Compared to the paediatric age group, this condition is quite uncommon in adolescence. The present case report describes isolated tubercular dactylitis of right proximal phalanx of middle finger in a teen age girl.

Case History: A 16 year old Muslim female, high school student presented in the OPD with complaint of swelling of right middle finger of one year duration, which had gradually progressed from the size of a peanut to its current size of a sphere of 2 cm diameter. The swelling had bluish discoloration at the periphery and undermined edges. For the last one month, a sinus had also appeared over the swelling & the patient complained of frequent sero-sanguinous discharge from the same (fig1). She also complained of on & off low grade fever, malaise and mild pain in the affected finger for the last three months. The patient denied any history of trauma, previous anti-tubercular treatment or family history of tuberculosis. On examination, the chest was bilaterally clear with absence of any respiratory symptoms, or, lymphadenopathy. Systemic, examination was unremarkable.

On routine investigations, Hb: 7.8gm% with a,microcytic hypochromic picture demonstrated on the blood smear; Total Leukocyte count: 4,600; Differential count: Polymorph 63%, Lymphocyte 34%, Monocyte 3%. ESR: 35mm fall in the first hour. Mantoux test was positive with an induration of 20 mm diameter after 72 hours. Urine examination showed protein in traces with occasional calcium oxalate crystals. Blood sugar, Liver & Kidney function tests and serum electrolytes including calcium & phosphate were within normal range. ZN Staining of the sputum was negative for AFB.

Skiagram of chest in PA view was normal for age. Skiagram of hand in AP and Lateral view showed punched out osteolytic lesion in proximal phalanx of right middle finger (fig2). A biopsy of soft tissue and bone from the site was taken and sent for histopathological analysis which revelaed well-formed epithelial cell granuloma suggestive of chronic granulomatous inflammation. In addition, inflammatory exudates with degenerated granulomas having Langhans and foreign body type of giant cells (fig3.) which are the hallmark of tuberculosis were also observed. The smear, however, was negative for AFB which can be attributed to the paucibacillary nature of this condition.

Figure 1: Right Middle Finger Shows Bluish Discoloration with Discharging Sinus.

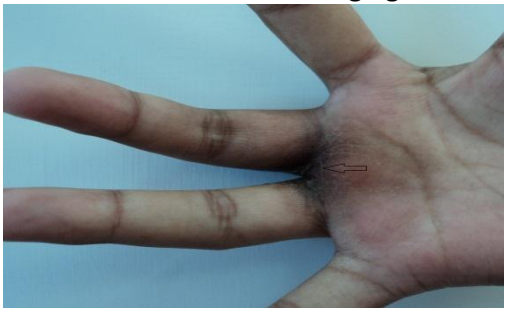


Figure 2: Skiagram of Right Hand in Anterioposterior And Lateral View Shows Osteolytic Lesion In Proximal Phalanx Of Middle Finger.

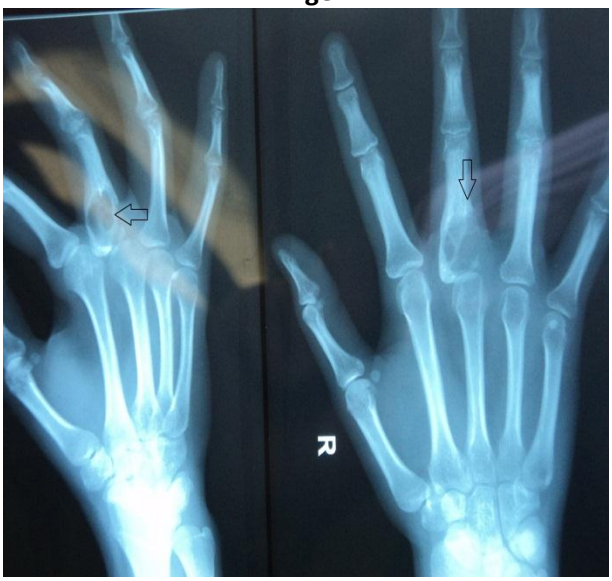
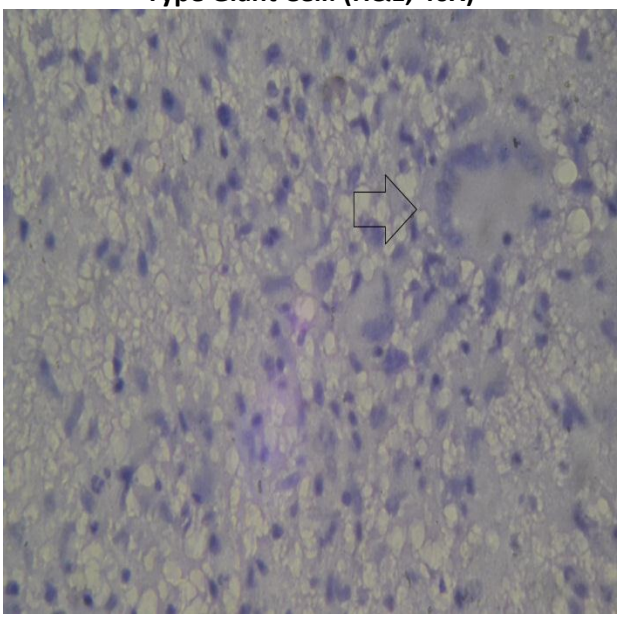


Figure 3: Epitheloid Cell Granuloma with Langhans Type Giant Cell. (H&E, 40X)



Discussion: Boyer first described spina ventosa (spina = short bone; ventosa = inflated with air) in the short long bones in 1803, while Nelaton proved the tuberculous etiology of the condition in 1837[3] and Feilchenfeld in 1896 described Tuberculous Dactylitis roentgenographically in children[1,4]. Bone and joint tuberculosis is not a very common form of tuberculosis with a prevalence of only 1%–3%[5] of all extra-pulmonary tuberculosis cases and spine & hip being the most commonly affected sites. Dactylar tuberculosis is extremely rare in adolescent age and 85% of children with this disease are younger than 6 years of age. Its incidence among children with tuberculosis was reported to be 0.65%-6.9%[6], and the bones of hands are more frequently affected than the bones of feet with the proximal phalanx of index and middle finger being the commonest sites of infection[2].

Generally bacteria migrate through primary foci of infection like lung or lymph node[7], and hematopoietic marrow in the short bones offers granulation tissue for haematogenous bacterial implants. The infection rapidly involves the entire marrow space and granulation tissue expands the relatively soft cortex. The resultant fusiform expansion of the bone with thinned cortex and relatively radiolucent marrow space due to trabecular destruction resembles an inflated balloon[5]. The radiographic features of cystic expansion of the short tubular bones have led to the name of “spina ventosa” being given to tuberculous dactylitis of the short bones of the hand[1].

The definite diagnosis of tuberculous dactylitis is made by bacteriological and histological studies, although direct visualization of mycobacterium under microscope is quite difficult in paucibacillary infection. Mantoux skin test is beneficial in the diagnosis of skeletal disease in childhood, particularly in communities where the incidence of TB is high. It was also reported that the rate of false-negative results of Mantoux test was 14%. Therefore a positive Mantoux test result can be helpful in confirming a diagnosis of tuberculosis, but a negative result cannot exclude it [8].

The differential diagnosis of tuberculous dactylitis includes osteomyelitis, metabolic diseases (gout), sarcoidosis, and tumours[2,9]. However osteomyelitis is rapidly responsible for bone destruction whereas the rate of lesion progression is far slower with mycobacterium. In sarcoidosis, well-demarcated cystic lesions are found in the phalanges of the fingers, although bony expansion and periosteal new bone formation are not found.

Draining sinus is frequently associated with tuberculous osteomyelitis and may confuse the distinction from pyogenic disease [4]. Clinically, pyogenic osteomyelitis tends to be acutely painful, swollen, and hot, with generalized fever. Contrary to this tuberculous osteomyelitis is more often only mildly painful, pyrexia is minimal, and the whole condition is relatively benign[3].

Current recommendations for the treatment of osseous tuberculosis include a 2-month initial phase of isoniazid, rifampin, pyrazinamide, and ethambutol followed by a 6- to 12-month regimen of isoniazid and rifampin[5]. Although few studies have endorsed the paucibacillary nature of the lesion and advocate a 6-month treatment course but contrary to this Subasi, et al. describe a 4-drug regimen for 2 months, followed by a 2-drug regimen for 10 months[2]. Therefore in the present case the anti-tubercular treatment was started with four drugs for two months and in follow up the patient showed symptomatic improvement.

Conclusion: Tuberculosis (TB) is still the second most frequent infectious disease after malaria on a worldwide basis[10]. Hence we cannot deny the possibility of tubercular infection at any rare site in the body where has not previously been reported or mentioned in standard text books as in present case.

It is recommended that physicians as well as surgeons always consider the tuberculosis as differential diagnosis in the infectious disease of rare site before the commencement of treatment to avoid future complication.

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