INTERNATIONAL JOURNAL OF MEDICAL AND APPLIED SCIENCES
E-ISSN:2320-3137

Case Report

RARE PRESENTATION OF TUBERCULOSIS AFFECTING BOTH BONES OF FOREARM

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ABSTRACT

Tuberculosis (TB) of long bones in leg or forearm is a rare entity. Usually in these sites, either of the two bones are involved. Lesion of long bones usually affects the metaphysis region. In this case , the patient has involvement of both bones of forearm, further more, the region of epiphysis, metaphysis and to some extent diaphysis of ulna is involved along with metaphyseal affection of radius on medial side giving a misinterpretation during diagnosis as vanishing bone disease. Presentation of kochs as disappearing bone disease in very rare.
Keywords:Tuberculosis,destruction lower third ulna,curetting,AKT,REAPPEARING OF BONE

INTRODUCTION

Tuberculosis is still endemic in many developed countries.Extrapulmonary tuberculosis involvement of the musculoskeletal system is uncommon, accounting for only 10% of cases.Bone and joint tuberculosis is next to pulmonary affection in tuberculosis. Spine, hip and knee are common joints affected in that chronological order. Double or multiple lesions in bone or joint is uncommon except in spine and small bones of hands and feet (eg.Metacarpal and metatarsals).Involvement of the both bones in forearm is extremely rare, and in lytic lesions where bone is destroyed, the diagnosis may be misinterpreted as Gorham's disease (disappearing bone disease).We were unable to trace such type of presentation of involvement of both bones of forearm in literature.We report a case of a 45 year old male , presenting with pain in left lower end forearm, presumptively diagnosed as disappearing bone disease. Biopsy revealed as kochs.

CASE REPORT

45 year old male came to Orthopaedic OPD with complain of pain ,deformity, restricted painful movements and wasting in left lower third forearm since 4 months with history of occasional fever. No significant past history. There is history of chronic smoking. No history of preceding trauma.

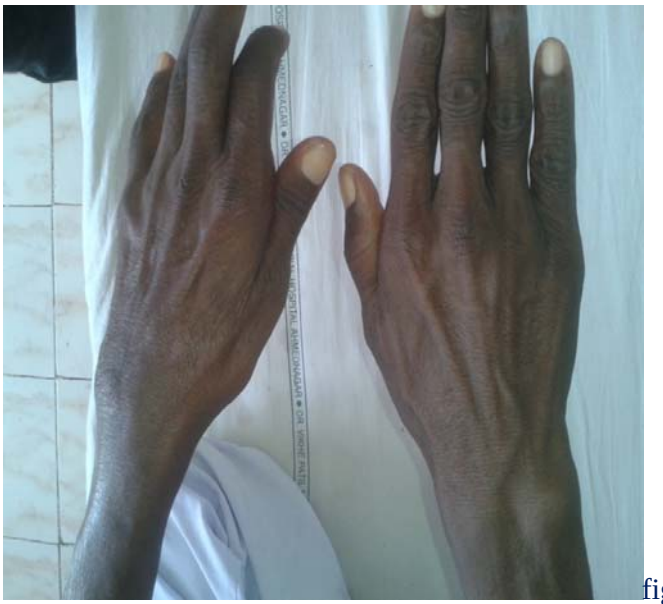


fig1 – profile view of both forearm

On examaintion, patient was poorly nourished, anaemic, with marked wasting of left forearm , lower end ulna was hardly palpable on medial side of distal forearm, movements of wrist and forearm (pronation- supination) were painful and restricted. Power of small muscles in hand was decreased. Since last month, he was unable to do day to day activities with his left hand. No palpable lymphadenopathy in axilla, no distal neurovascular deficit was elicited.

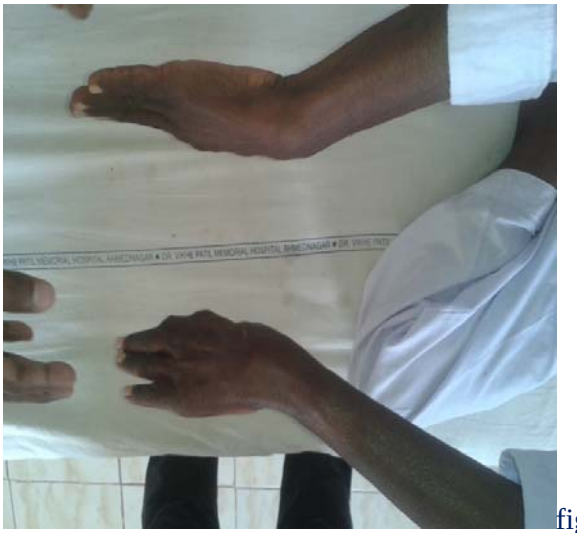


fig 2- lateral view of both forearm

Xray revealed destruction of medial part of radius in metaphyseal area and distal part of ulna involving epiphysis, metapyhsis and diaphysis. Laboratory parameters revealed anaemia, raised ESR (30mm at the end of 1st hour). Mantoux was inconclusive. Chest Xray was normal.

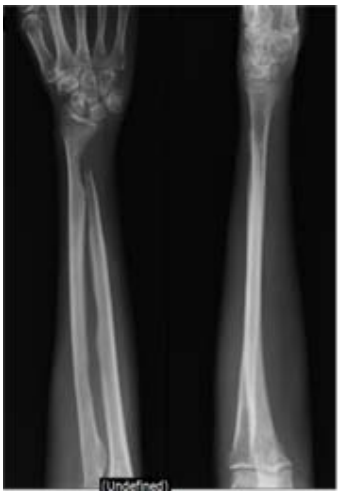
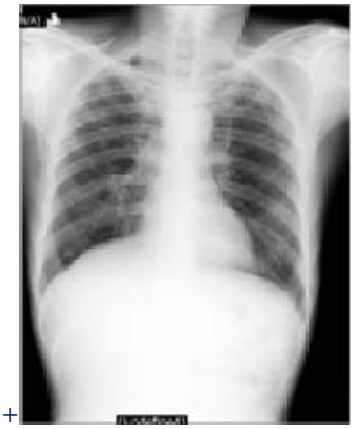


fig 3- preop xray of the patient



+ fig 4- chest xray did not show any abnormality

Provisional diagnosis of disappearing bone disease was made. After obtaining fitness and valid consent, patient underwent biopsy of the lesion. Intraoperatively, there was no bony or cartilaginous tissue , on medial side of distal forearm, there was erosion of radius in metaphyseal region with presence of granulation-like tissue . Distal end of ulna was not seen in the operative field . Curretting of the tissue was done and sent for histopathology. HPE findings revealed thickened bone of lamellar structure without marrow cavities next to fibrous tissue, with few fibroblasts and a small number of newly formed vascular channels.

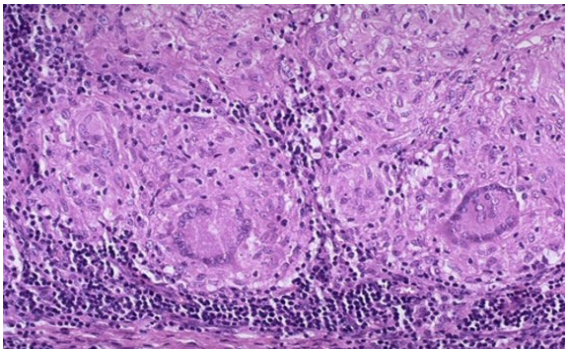


fig 5 – HPE of slide demonstrating tubercular foci

HPE revealed kochs. Patient was started on AKT. Postoperatively, after removal of stitches, he was kept in above elbow cast for 6 weeks in midprone position with wrist in 5 degrees dorsiflexion.

He was assessed radiologically ,clinically. Lab investigations (CBC) was done at follow up. Active movements and physiotherapy was started at end of 2 months . Patient was advised removable cock-up splint for next 2 months.

Serial follow up every monthly was done for first 3 months and then quarterly subsequently .

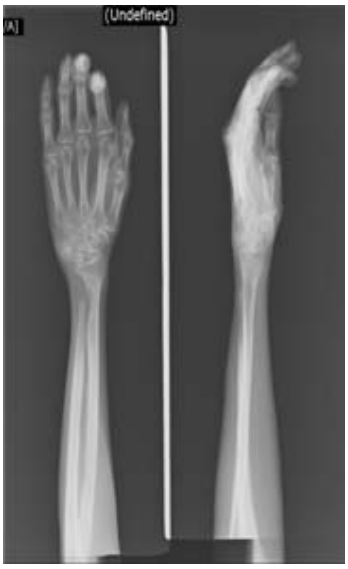


fig 6- post op xray at 3 months

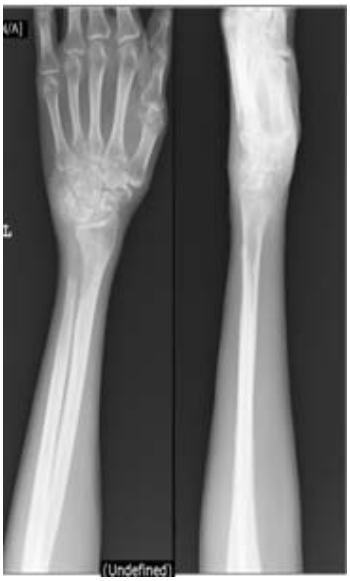


fig 7- xray after 11 months showing growth of ulna

Now, at the end of 1 year, patient has relief of symptoms and there is regrowth of distal end of ulna.there is subsidence of pain and there is improvement in power of hand and having fair movements at wrist to perform simple day to day activities .

DISCUSSION

Osteoarticular involvement occurs in 1 to 3% of patients with extrapulmonary tuberculosis[16] and spine represents 50% of these lesions[21]. Tuberculous involvement of the forearm bones is a rare presentation of extrapulmonary tuberculosis[18,19.] Only 1/3rd of patients with tuberculosis of the bone are diagnosed with concomitant active pulmonary disease[22]. Disseminated skeletal tuberculosis without primary foci is rare[23]. The skeletal infection often becomes symptomatic within 1-3 years after initial infection.

Diagnosis of kochs is made on radiographic features and culture of Mycobacterium Tuberculosis. Radiologically the affected bone appears expanded with lytic lesions in the middle (as seen in present case) and sub periosteal new bone formation along the involved bone. The cavity may contain soft coke like sequestra[24-25.] Other findings on plain radiographs include osteopenia, soft-tissue swelling with minimal periosteal reaction, narrowing of joint space, cysts in bone adjacent to joint, and subchondral erosions. The non-specific nature of these radiographic findings can often delay the diagnosis.

The gold standard for the diagnosis of osseous tuberculosis is culture of Mycobacterium tuberculosis from bone tissue[26]. Differential diagnostic considerations include pyogenic osteomyelitis, Brodie's abscess, Kaposi's sarcoma and lytic dactylitis. Clinically, pyogenic osteomyelitis tends to be acutely painful, swollen, and hot with fever. Tuberculous osteomyelitis is relatively benign with mild pain and minimal pyrexia.

Management is essentially by antitubercular drugs, rest to the involved part in functioning position and early active exercise. 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[27]. Few studies argue that 6-month of antitubercular treatment is appropriate for tubercular of forearm bones because of its paucibacillary nature.[28]

Tuberculous is difficult to diagnose during early stages. Tubercular should be suspected in cases of long-standing pain and swelling . It is necessary to keep tubercular in mind while making the differential diagnosis of several osseous pathologies.

Vanishing bone disease is a rare idiopathic disease, leading to extensive loss of bony matrix, replaced by proliferating thin-walled vascular channels and fibrous tissue. There are >191 cases reported in the English literature. Gorham and Stout made the first overview of the disease in 1955 and they first presented 24 cases known at that time. The etiology remains speculative, the prognosis unpredictable, and effective therapy still unknown. The disease can be monostotic or polyostotic although multicentric involvement is exceptional.

Vanishing bone disease is a rare idiopathic disease leading to extensive loss of bony matrix, which is replaced by proliferating thin-walled vascular channels and fibrous tissue.[8] Although the disease can be monostotic or polyostotic, multicentric involvement is unusual.[9] The process often extends to the soft tissues and adjacent bones, especially at the shoulder girdle. There are no associated endocrine or metabolic abnormalities.[10]

Genetic transmission or sex predilection is not evident. A pregnancy with Gorham disease was described in 1993.11 The infant was delivered by low forceps, with a good outcome. Two other natural pregnancies also were reported in 1990.[12]

Pathogenesis and Pathophysiology

The mechanism of bone destruction and resorption remains unclear.[7] The etiology is unknown. Incidence of the disease may be linked to a history of minor trauma, although as many as half of the patients have no history of trauma.[9]

Most cases occur in children or in adults <40 years. However, the disease has been described in patients as young as 1 month[8] to as old as 75 years.[13] The bones of the upper extremity and the maxillofacial region are the predominant osseous locations of the disease. Approximately 60% of all cases with vanishing bone disease occur in men.[1]

Leriche's hypothesis that post-traumatic arterial hyperemia was responsible for bone resorption was rejected first by Mouchet and later by Gorham et al.[3] The same authors postulated an angioma might act as a shunt, increasing local oxygen tension.[3]

In most cases, trauma was relatively trivial, and in some cases, trauma did not occur. As with many other diseases, the role of trauma in vanishing bone disease may be to signal the presence of a preexisting abnormality. Knoch[15] suggested a previous silent hamartoma becomes active after a minor trauma, leading to bone resorption.

Neurovascular changes such as Sudeck atrophy also have been described.[16] Thompson and Schurman[17] suggested the disease is a primary aberration of vascular tissue in bone, related to hyperemic granulation tissue. According to Young et al,[18] endothelial dysplasia of blood and lymphatic vessels could lead to osteolysis. Osteoclasts are the only cells capable of resorbing bone. Thus, it is presumed vanishing bone disease may represent a pathologic derangement of osteoclastic activity. Any defect of the osteoclasts could lead to idiopathic osteolysis.[19]

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