ORIGINAL RESEARCH PAPER

Paediatrics

TUBERCULOUS DACTYLITIS- AN UNCOMMON PRESENTATION OF A COMMON INFECTION

KEY WORDS: tuberculosis, dactylitis, nonhealing ulcers

Dr Sudipta Mishra Post Graduate Pediatrics Final Year, Hitech Medical College, bhubaneswar.

Prof Dr Bishwajit Mishra Hitech Medical College, Bhubaneswar- *Corresponding Author.

Prof Dr Suryakanta Swain Professor And Hod Department Of Pediatrics Hitech Medical College, Bhubaneswar.

ABSTRACT

Background- Tuberular dactylitis is an unsual form of osteoarticular tuberculosis involving short tubular bones of hands and feet which is uncommon beyond 6years of age. Case Brief-We report a case of 16 year old male who was diagnosed with tubercular dactylitis involving unlateral hand. His diagnosis was delayed due to lack of suscpicion of this rare entity. He presented to the OPD with fever for 1 month and swelling of his right hand with non healing ulcers since 15 days. On examining the laboratory reports and radiographs, findings correlated with tuberculosis, suggestive of osteoarticular tuberculosis. MRI of hand and wrist was suggestive of osteomyelitis and infective arthritis. Histopathology reports revealed chronic granulomatous inflammation but AFB was not isolated. CBNAAT sputum and gastric aspirate was positive for mycobacterium tuberculosis. PPD was positive and anti tubercular treatment was started.

INTRODUCTION

The disease tuberculosis is old and existed even in early ages .It still continues to be a problem in developing countries. It is estimated that 1-6% of children with primary infection may develop bone and joint Tuberculosis in 1 -3 yrs if left untreated.[1,2] Osteoarticular TB(tuberculosis) can be joint TB ,Bone TB ,Spine TB .Infection in a bone or joint with Mycobacterium tuberculosis is almost always secondary to a primary focus, in the lymphatic gland ,lungs or mesentry ,from where it disseminates by hematogenous route. There may be a history of trauma , after which hematoma is formed which can become a nidus for infection with tuberculosis bacilli. Epithelioid cells, giant cells, fibrosis at periphery, deposit to form follicles which enlarge to become tubercle .As the tuberculosis lesions heal,sclerosis takes place at sites like short long bones ,clavicle, intense sclerosis activity by layer of subperiosteal bone and characteristic of a tuberculous lesion, Which ultimately bursts leading to formation of tubercular sinus. Osteoarticular TB mostly occurs during childhood, which is insidious in onset, starts as monoarticular or mono osseus involvement. The child complaints of night cries, mild grade fever, loss of weight and apetite. In later stages, all movements get restricted, however muscle atrophy occurs early. Tuberculosis of metatarsal and phalanges starts in the diaphysis as opposed to metaphysis in long bones .This is because of the short long bones- the nutrient artery which enters the bone in the middle of the shaft .Within a short time ,the finger gets enlarged and painful .As the bone gets involved ,new subperiosteal bone is laid giving rise to typical enlargement of the shaft. The prognosis is good ,often heals with anti tubercular drugs.

Tubercular dactylitis a rare form of osteoarticular tuberculosis involving short tubular bones of hands and feet(phalanges ,metacarpals ,metatarsals)[3].It is more common in younger population with 85%cases seen in children less than 6years of age and accounts for 0.65 to 6.9% of all forms of tuberculosis cases in children.[4]Tubercular dactylitis can present without constitutional symptoms further adding to diagnostic difficulty[5]. Cytopathology (FNAC and/or biopsy of bone or synovium) rules out other conditions. The gold standard for the diagnosis of osseous tuberculosis remains the positive culture of mycobacterium tuberculosis from bone tissue. Tubercular dactylitis responds well to anti-TB drugs. Current recommendations for the treatment of osteoarticular TB include a two-month initial phase of isoniazid, rifampicin, pyrazinamide and ethambutol as followed by a ten-month regimen of isoniazid and rifampicin. Few reports clearly define the optimal duration for the treatment of tuberculous dactylitis. Surgery has a limited role. Diaphyseal lesions show slow healing. In general, the bone heals by formation of new bone following slowly filling defects which can become sclerotic or trabeculated.[6]

Associated tuberculosis lesions: Pulmonary tuberculosis was the most common associated lesion followed by osteo-articular tuberculosis at other sites and tuberculous lymphadenitis .Skin involvement in the form of lupus vulgaris or scrofuloderma can also be present. Some might also have associated tuberculous tenosynovitis of flexor sheath of hand and associated tuberculous abscess of the temporal lobe of brain.

Case Summary

Fig 1 : General appearance of child

Fig 2 : Gross appearance of lesion

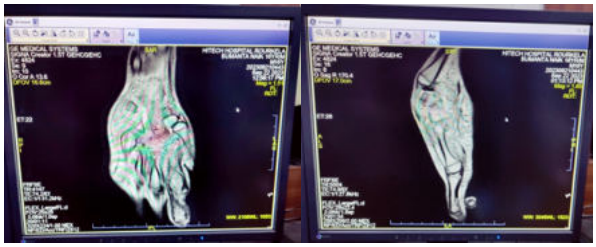

Fig 3 a : MRI findings
Fig 3 b : MRI findings

A 16yr old boy,29kg,born out of non consanguineous marriage ,presented to the OPD with fever since 30 days, back pain since 30days,with swelling of right hand and ulcers(painless) on dorsum of hand since 15days. Fever was insidious in onset, of low grade in nature, persisted throughout the day, was not relieved with medications. The patient also complained of decreased appetite and generalized weakness. On physical examination, the vitals were normal, pallor was present., no hepatospleenomegaly was seen. All systemic examination were within normal limit. The clinical study found a swelling and inflammatory aspect of the skin of the finger. The finger motion was painful and limited. On local examination, non healing punched out ulcers measuring 5cm *4cm on dorsum of right hand with discharging sinus was seen. He had restriction of movements around elbow and knee joints. Family history of exposure to open case of pulmonary TB was positive and the boy has personal history of abuse of bidi. However the child had no history of cough and night sweats. Anthropometry showed weight = 29kg. (less than 3rd centile), height151cm(3rd to 15th centile).

Total leucocyte count was 9,900/mm3 with differential count being predominantly-neutrophilic 82%, Hemoglobin was 7.5% and total platelet count was 5.15 lac. Liver function tests and renal function tests were normal. The analytical study showed elevated acutephase reactants with ESR (erythrocyte sedimentation rate) of 90 mm/hr and peripheral smear showing moderately hypochromic cells with anisopoikilocytosis. However the sickling test was negative which was confirmed with HPLC (high performance liquid chromatography) which was normal. Montaux test was strongly positive (22*20mm) induration after 72hrs of 10 tuberculin units., The Xray of right hand showed Fusiform soft tissue swelling with joint space narrowing with cystic degenerative changes and lytic lesion.The Admission chest radiography demonstrated right hilar lymph node enlargement with multiple scattered nodules. The special coloring of Ziehl–Neelsen in sputum did not highlight the Mycobacterium tuberculosis (MTB). She was nonreactive for retroviral infection. A fine needle aspirate showed sheets of degenerating cells and epitheloid cell clusters in a hemorrhagic background suggesting a granulomatous inflammation consistent with tuberculous dactylitis. CBNAAT sputum and gastric aspirate was positive . MRI showed infective arthritis like tuberculosis of radiocarpal, intercarpal, carpometacarpal, 2nd and 5th metacarpophayngeal joints. Osteomyelitis of entire 2nd and 5thmetacarpals,proximal phalanx of index finger , proximal half of 3rd and 4thmetacarpals. Gross radiocarpal, intercarpal, carpometacarpal, 2nd and 5th metacarpophayngeal joint effusion with synovitis, extensive ill defined abscess surrounding flexor and extensor tendons , the distal forearm, wrist, and deep spaces of palm. Extensive soft tissue edema of distal forearm, wrists, fingers , thenar and hypothenar muscle.In hospital treatment included injection piperacillin tazobactam for 14days .Anti tubercular treatment was started(HRZE2 and HR4).While the patient was on follow up after 4 weeks of treatment,the size of swelling has decreased 2cm*1cm and repeat ESR showed 12mm/hr ,with restoration of hand and finger movements .The patient was advised to continue total 10 months of isoniazid and rifampicin in continuation phase after seeing the good response to treatment.

DISCUSSION

Tubercular dactylitis is a rare presentation of extra pulmonary tuberculosis and it constitutes less than 1% of skeletal tuberculosis.[7] There are some underlying predisposing factor for tuberculosis. Low socioeconomic status and immunodeficiency were the most common predisposing factors. Malnourishment and vitamin D deficiency, and diabetes mellitus were also each. Immune deficiencies included human immunodeficiency virus (HIV) infection, severe combined immunodeficiency (SCID) and chronic granulomatous disease (CGD) . Vitamin D deficiency and diabetes were the other rare factors in our review. Vitamin D deficiency may impair T-cell function and decrease the production of cytokines T helper 1 (Th 1), interleukin 2(IL-2) and gamma interferon, thus the increasing risk of primary tubercular infection and its reactivation .[8,9]. Local trauma prior to development of local symptoms of tubercular dactylitis whereas some also have it after development of symptoms leading to burst of abscess and subsequent sinus formation.].Single sinus was 6 to 7 times more common than multiple discharging sinuses. Sinus and fistula formation is a feature of long standing neglected cases.[10]. Tuberculous dactylitis causes a subacute to chronic painless swelling of the digits as observed in this case. It can lead to destruction of the involved bone with shortening.

The disease is slowly progressive , and diagnosis is often delayed due to multiple factors, including the paucibacillary nature of the lesion, as smears are often negative; non-specific clinical manifestations of the disease; the ability of occurrence of the disease in the absence of pulmonary tuberculosis; the lack of a rapid microbiological diagnostic method; and poor awareness among treating physicians and surgeons who do not have much experience with this disease[.11]. Diagnosis is confirmed by imaging and cytopathology. In this case, Chest xray showed hilar lymphadenopathy ,montaux test was positive, CBNAAT sputum was positive and the diagnosis was confirmed by MRI imaging and histopathology. The benign course of dactylitis with absence of fever distinguishes it from bacterial osteomyelitis.

As per literature, bones of hand are more commonly involved than of the foot. In hand proximal phalanx of index and middle finger are most often affected[4,10,12].Involvement of toes is less common [10].In hands, phalanges were more commonly infected than metacarpals; whereas in feet, involvement of metatarsals was more common than that of phalanges. Surprisingly, there was no case in literature review of distal phalanx tuberculous dactylitis of fingers or toes. Radiological findings in tuberculous dactylitis are of non-specific nature and include a combination of soft tissue swelling, lytic lesions, cystic expansion of the bone as if it appears filled with air, cortical erosions, cortical destruction, periosteal reaction, sclerosis, sequestrum formation, reduction of adjacent joint space, subchondral erosions and pathological fracture [13]. There is no significant periosteal layering or thickening and sequestration ordinarily does not occur. Sclerosis is seen in only long standing cases and during the healing phase [3,4,14,15]. Marked periosteal reaction is seen in cases with sinus formation due to associated secondary pyogenic infection of which periosteal reaction is a hallmark. Sequestrum formation, expansion (endosteal resorption), multiple osseous involvement, and a positive chest roentgenogram is more frequent in children while as pathological fracture is more frequent in adults [16].Tuberculous dactylitis can virtually mimic any disease involving short tubular bones of hand and feet. The spectrum of disease it can mimic include infectious or non-infectious granulomatous disease, tumours, hemoglobinopathies, endocrinopathies, metabolic disorders and others [5,17,18,10,19]. Among infections, it resembles pyogenic osteomyelitis, brodie's abscess, fungal osteomyelitis, parasitic infections, atypical mycobacterial infection, syphilitic dactylitis, brucellosis and Madura mycosis [3,5,10,20,21,22]. Tuberculosis of short tubular bones may be confused with tumours, both benign and malignant. Among benign lesions it resembles enchondroma, cortical fibrous defects (monostotic fibrous dysplasia), aneurysmal bone cyst, giant cell tumor, chondroblastoma, osteoid osteoma, florid reactive periostitis, pagets disease, eosinophillic granuloma (histiocytosis X) [3,17,10,19,21].

Malignant lesions include secondary deposits, Ewings sarcoma, osteosarcoma, myeloma, leukemia, malignant giant cell tumor, Kaposi sarcoma [5,17]. It also mimics chronic granulomatous disease like sarcoidosis [17,18,2]. Hemoglobinopathies such as sickle cell disease can present as sickle cell dactylitis which mimics TD [5]. Among endocrinopathies and metabolic disease it resembles hyperparathyroidism (brown tumor) and gout respectively [5,17,18]. Rarely, it can be confused with hereditary acro-osteolysis Musculoskeletal tuberculosis was previously considered rare extra-pulmonary form of tuberculosis accounting for 10 – 18% of extra-pulmonary cases, but recent studies show it represents 27 – 35% cases and hence the most common site of extra pulmonary tuberculosis.

Diagnostic delay was due to;

1- lack of high index of suspicion and poor awareness among clinicians.
2- non specific clinical manifestations
3- presentation at unusual age (uncommon beyond 6years once the epiphyseal centres are well established)

Therefore after careful history taking,examination of findings and appropriate diagnostic work up , the basis of prompt diagnosis of tuberculous dactylitis could be made.

Ethical consent-The participant gave informed consent to participate in the study.

REFERENCES

1. Starke JR, Smith MHD. Tuberculosis. In: Feigin RD, Cherry TD (Eds): Textbook of Pediatric Infectious Diseases, 4th edn. London: WB Saunders company 1998;1196-1239.
2.. Luckheie M.Tuberculosis of cervical spine. South Afr Med J 1996;86:553-6.
3. Bandyopadhyay R, Mukherjee A, Mondal RK. Case Report:"Spina Ventosa" Tuberculous Dactylitis in a 2 Year Old Boy - A Very Rare Disease. Open Orthop J 2012; 6:11820. Report: "SpinaVentosa" TuberculousDactylitis in a 2 Year Old Boy - A Very Rare Disease. Open Orthop J2012; 6:118-2
4. Salimpour R, Salimpour P. Picture of the month. Tuberculousdactylitis. Arch PaediatrAdolesc Med 1997;151 (8):851-852
5. Engin G, Acunas B, Acunas G, Tunaci M. Imaging of Extrapulmonary Tuberculosis. RadioGraphics 2000;20(2):471-488.
6. World Health Organization (WHO), author *Guidance for national tuberculosis programmes on the management of tuberculosis in children.* World Health Organization; 2014.
7. Subasi M, Bukte Y, Kapukaya A, Gurkan F.Tuberculosis of the metacarpals and phalanges of the hand.*Ann Plast Surg.* 2004;53(5):469–72
8. Nnoaham KE, Clarke A. Low serum vitamin D levels and tuberculosis: a systematic review and meta-analysis. Int J Epidemiol 2008;37 (1):113-119.
9. Dai G, Phalen S, Mcmurray DN. Nutritional modulation of host responses to mycobacteria. Front Biosci 1998;3:110-122.
10. Sunderamoorthy D, Gupta V, Bleetman A. TB or not TB: an unusual sore finger. Emerg Med J 2001;18(6):490-491.
11- Karanas YL, Yim KK. Mycobacterium tuberculosis infection of the hand:a case report and review of the literature.*Ann Plast Surg.* 1998;40:65–67.
12. Mohan V, Gupta SK, Agrawal AK. Disseminated multicystic tuberculosis. Indian Paediatr 1980;17 (12):987-990
13. Kushwaha RAS, Kant S, Verma SK, Sanjay, Mehra S. Isolated metacarpal bone tuberculosis-a case report. Lung India 2008;25 (1):17-19.
14. Gyanshankar PM, Dhamgaye TM, Amol BF. Spina ventosa discharging tubercle bacilli – A case report. Indian J Tuberc 2009;56 (2):100-103.
15. Kothari PR, Shankar G, Gupta A, Jiwane A, Kulkarni B. Disseminated Spina Ventosa. Indian J Chest Dis Allied Sci 2004;46 (4):295-296.
16. Feldman F, Auerbach R, Johnston A. Tuberculous dactylitis in the adult. Am J Roentgenol Radium Ther Nucl Me 1971;112(3):460-479.
17. Maruschke L, Baumann T, Zajonc H, Herget G. Monostotic Fibrous Dysplasia of the Middle Phalanx of the Hand. J Med Cases 2013;4(5):318-322 .
18. Hassan FOA. Tuberculous dactylitis pseudotumor of an adult thumb: a case report. Strat Traum Limb Recon 2010;5(1):53-56.
19. Kali L, Lund F. Tuberculosis of the hand. Tidsskr Nor Laegeforen2000; 120(4):445-446.
20. Wani IH, Muzaffer N, Jan M, Salaria AQ. Tubercular dactylitis with secondary involvement of tendon sheath in an adult. A rare manifestation of adult skeletal tuberculosis. Basic Res J Med Clin Sci 2012;1(3):47-50.
21. Roy AK, Khanduri S, Girisha KM. Fusiform swellings of fingers in a3-year-old girl. J Postgrad Med 2006;52(4):314-324.
22. Chrispal A, Prabhakar T, Booruga HK.A bizarre appearance of a common disease, tuberculous dactylitis, involving multiple digits in an adult. Trop Doct 2009;39 (1):51-52.