Tuberculous Dactylitis: A Case Series and Review of Literature

Nadeem Ali[1], Abedullah Bhat[1], Akeela Fatima[2], Khalid Muzzafar[3], Irfan Ahmad Latoo[4], Rajinder Singh[5]

[1]*M.B.B.S, M.S, Senior Resident, Department of Orthopedics, SHKM GMC Mewat, Nuh, Haryana, India*
[2]*M.B.B.S, Junior Resident, Department of Microbiology, Sher-i-Kashmir Institute of Medical Sciences Soura, Srinagar, Jammu and Kashmir, India*
[3]*M.B.B.S, M.S, Fellow limb reconstruction surgery, Resident, Ganga Hospital, Coimbatore, Karnatka, India*
[4]*M.B.B.S., M.S, Senior Resident, Bone and Joint Hospital Barzulla, Government Medical College Srinagar, Srinagar, Jammu and Kashmir, India*
[5]*M.B.B.S., M.S, D.N.B, Associate Professor, Department of Orthopedics, Government Medical College Jammu, Jammu, Jammu and Kashmir, India*

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Correspondence to: Dr Nadeem Ali

Address: Resident of Orthopedics, Government Medical College Jammu, Kashmir, India

Email: drnadeeem@gmail.com

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ABSTRACT

The literature and statistics on tuberculous dactilitis (TD) are scarce and most literature consists of isolated case reports. The aim of this case series is to examine trends and diagnostic difficulties faced by the clinicians in diagnosing this rare disorder. Google search engine and MEDLINE were searched for key words 'tuberculous dactylitis' and 'spina ventosa'. From all published papers and unpublished reports, 58 cases were extracted and 61 cases, including the three presented by us in this review, were analyzed for a set of 16 parameters. There is re-emergence and increase in the incidence of this form of extra-pulmonary tuberculosis (EPTB) especially in the industrialized countries, which poses a diagnostic challenge to physicians in these countries, as they are not well versed with this entity.

Keywords: Spina Ventosa; Extra Pulmonary Tuberculosis; Tuberculous Osteomyelitis; Osteo-Articular Tuberculosis

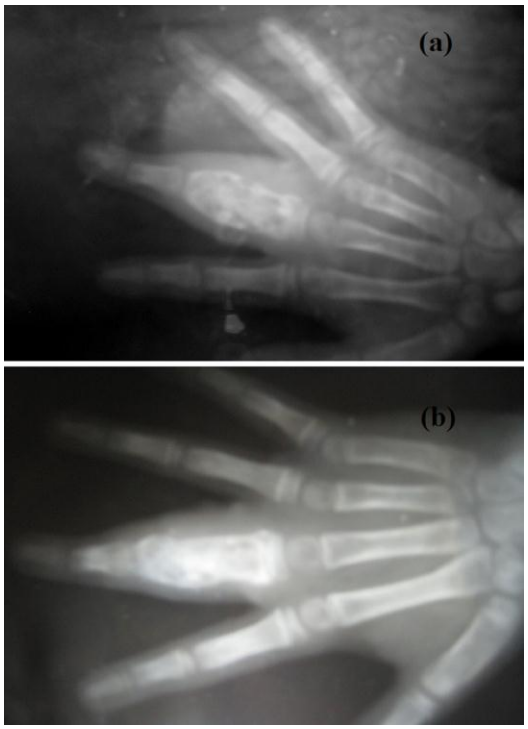
INTRODUCTION

Tuberculous dactylitis (TD) is a rare form of osteo-articular tuberculosis involving short tubular bones of hand and feet (phalanges, metacarpals and metatarsals) [1]. It is more common in younger population with 85% cases seen in children younger than 6 years and accounts for 0.65% to 6.9% of all forms of tuberculosis cases in children [2]. There are only scattered reports of adult cases with no statistics [3]. TD always poses a diagnostic challenge for orthopedic surgeons especially in the developed countries where tuberculosis is not as rampant as in the developing countries. Over a period of three years at our tertiary care centre, we have encountered three such cases. Besides, TD is a radiological mimicker of countless tumors, infections, hematological, and non infectious granulomatous diseases.TD can present without constitutional symptoms or evidence of tuberculosis in any other part of the body, further adding to diagnostic difficulty [4-8]. The incidence of this form of tuberculosis is expected to increase in the developed countries because of HIV infection and emigration from tubercular endemic areas [8]. Here we present three cases of TD that we encountered at our centre from May 2010 to April 2013 and provide a review of the published literature of this rare form of osteo-articular tuberculosis.

CASE REPORT

Case 1: An eight years old boy presented with progressively increasing swelling and dull pain of the left middle finger for the past seven months. There was no history of fever, night sweats, night cries, loss of weight, anorexia, cough, trauma or similar swelling in any other part of his body. His mother had pulmonary tuberculosis and she had received treatment two years ago. There was a fusiform swelling of bony consistency involving the proximal half of the left middle finger with mild increase in local temperature and tenderness on deep palpation. Movements of the finger were restricted. Examination of the respiratory system was normal. Erythrocyte sedimentation rate (ESR) was 36 millimeter (mm) and Mantoux reaction was positive. Radiograph of left hand showed soft tissue swelling of the middle finger, bone expansion, lytic lesions, sequestrum, cortic-

Figure 1: a) Proximal phalanx of middle finger expanded with cortical erosions and destruction. b) Radiograph at 5 months showing sclerosis and healing.

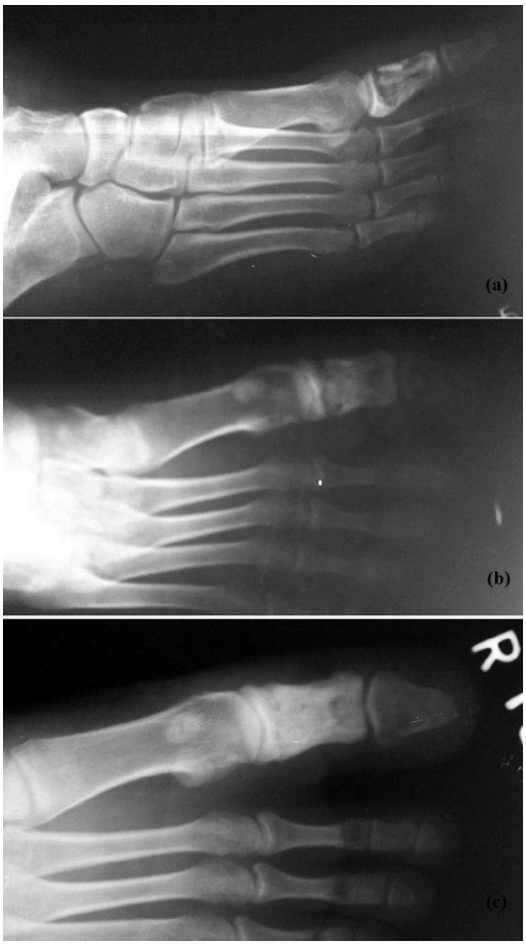


-al erosions and destruction of proximal phalanx of middle finger (Figure 1a).Chest radiograph was normal. Diagnosis of TD was made based on family history of tuberculosis, raised ESR and radiological picture of spina ventosa. Treatment with four anti-tubercular drugs (isoniazid, rifampicin, pyrazinamide, ethambutol) was instituted for 2 months after which only two drugs (isoniazid, rifampicin) were continued. There was substantial decrease in swelling and restoration of finger movements but patient was lost to follow up at 5 months (Figure 1b).

Case 2: A fifteen year old boy presented with swelling of the right great toe for last one year and a sinus with serous discharge on the dorsal surface of the same toe for the past 5 months. The patient had consulted a dermatologist and many general practitioners who prescribed antibiotics but the condition did not improve. There was no history of trauma, fever, night sweats, loss of weight, loss of appetite, contact with a case of tuberculosis and no family history of tuberculosis. Examination revealed soft tissue swelling with a single draining sinus on the dorsum of toe. Systemic examination was unremarkable. ESR was raised (45 mm in first hour). Mantoux test was positive. Ziehl Nielsen staining and culture for mycobacterium of sinus discharge were negative. There was a bony lytic lesion, cortical erosions, sequestrum in the proximal phalanx of right big toe on radiography (Figure 2a, Figure 2b). Chest radiograph had no abnormality. A preliminary diagnosis of TD was made and four anti-tubercular drugs (isoniazid, rifampicin, ethambutol and pyrizinamide) were administered for 3 months followed by two drugs (isoniazid, rifampicin) for another 12 months. Swelling subsided and discharge diminished within 2 months and the sinus healed completely within 5 months of treatment. Final radiograph at the end of treatment (15 months) showed sclerosis and healing of the lesion (Figure 2c).

Case 3: Two years old boy admitted in the pedia-

Figure 2: Lytic lesion containing feathery sequestrum with cortical erosions and destruction of proximal phalanx of great toe a) Oblique view b) AP view. c) Healing and sclerosis at 18 months.

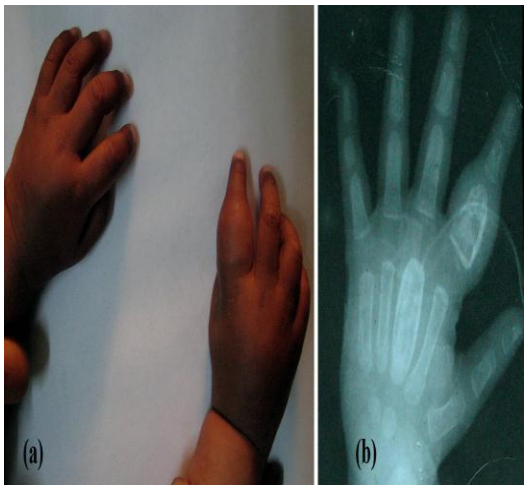


-trics department for the chief complaint of right sided weakness of the body for the past one month. He was diagnosed with tubercular left temporal lobe abscess and iron deficiency anemia with simple partial seizures. He was referred to us with 6 months history of progressively increasing painless swelling of the right index finger. There was history of evening rise of temperature above normal, weight loss and loss of appetite. There was no history of trauma, no history of tuberculosis contact or family history of tuberculosis. General physical examination of the patient revealed body temperature above normal, pallor of palpebral conjunctiva, and generalized wasting. On systemic examination, there was right sided hemiparesis. Examination of the respiratory system was normal. Locally there was a fusiform, non-tender, bony swelling of the proximal half of right index finger (Figure 3a). Epitrochlear lymph node was palpable (1.5 × 1.5 cm) and freely mobile on the right side. Hemoglobin was 8grams/deciliter and ESR of 92 mm. Radiograph of right hand had spina ventosa like picture of proximal phalanx of index finger and third metacarpal (Figure 3b). Bilateral apical infiltrate was seen on chest radiograph. Fine needle aspiration cytology of the epitrochlear lymph node showed multinucleate giant cells, epitheloid cells and plenty of plasma cells suggestive of granulomatous inflammation but staining for mycobacterium was negative. Based on these findings a diagnosis of disseminated multifocal tuberculosis with TD was made with advice of anti-tubercular drug therapy as prescribed by pediatrician and follow up after every 2 weeks. The patient never returned for follow up.

LITERATURE SEARCH

Google search engine and MEDLINE were searched using key words 'tubercular dactylitis' and 'spina ventosa'. All published and non-published results were evaluated for case(s) of TD. Case reports that mentioned age, sex of the patient, and the site of TD were included for review. Fifty-eight case reports of TD from 41 published papers and 3 unpublished reports fulfilled inclusion criteria. Thus, 61 cases including our above mentioned three cases were included in this review. In addition to inclusion criteria, data on 13 other parameters were extracted; presenting symptom(s), duration of symptoms before consulting a specialist, presence of constitutional symptoms of tuberculosis, any draining sinus from TD lesion,

Figure 3: a) Swelling right index finger and dorsum of hand. b) AP view showing Spina Ventosa of proximal phalanx of index finger and third metacarpal



predisposing factors, any history of local trauma, history of contact or positive family history, recent emigration from endemic area, radiological findings, associated tubercular lesion(s), ESR (raised or normal), mantoux/PPD reaction (positive or negative), and the initial diagnosis.

LITERATURE REVIEW

Included case reports were published between 1971 and 2013. Age of the patients ranged from 5 months to 77 years (average = 18.29 years), 41% (n = 25) were 10 years or younger, and 24.6% (n = 15) were between 11 and 20 years. A majority were males (56%, n=34).

Clinical features: Of the 61 cases, chief presenting complaint was mentioned in 55 cases; 96% (n = 53) presented with swelling, of which 57% (n = 30) had a painful swelling. One case (2%) presented with only pain as his complaint and another one as discharging sinus. The duration of symptoms was known in only 38 cases. Patients presented for the first time between 2 weeks to 2 years from the onset of symptoms (mean duration = 5.99 months), 39% (n = 15) reported within 2 months of the onset of symptoms, and 2 (5%) out of 38 patients had first encounter with orthopedic specialist 2 years after the onset of symptoms. Of the 36 patients in whom data were reported on constitutional symptoms, one or more constitutional symptoms of tuberculosis were present only in 42% (n =15). Examination was reported in 50 patients of

which 15 showed a discharging sinus (es) related to bone with TD, of which 13 cases had a single sinus whereas 2 had more than one discharging sinuses. ESR was elevated above normal range in 24 (80%) out of 30 patients. Mantoux or PPD intra-dermal skin sensitivity test was positive in 22 out of 29 patients.

Risk factors: Of the 36 patients with data, 11 (31%) had some underlying predisposing factor for tuberculosis. Low socioeconomic status and immunodeficiency were the most common predisposing factors, each present in 3 patients (8%). Malnourishment was seen in 2 (5.56%) and vitamin D deficiency, and diabetes mellitus in 1 (3%) each. Immune deficiencies included human immunodeficiency virus (HIV) infection, severe combined immunodeficiency (SCID) and chronic granulomatous disease (CGD) each in one patient.

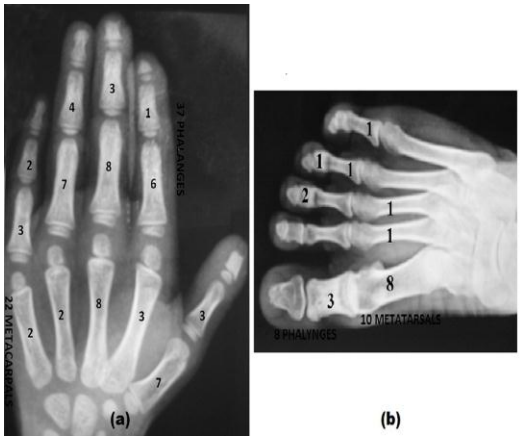
5 cases (13%) had history of local trauma. 3 cases (8%) had trauma prior to development of local symptoms of TD whereas 2 (5%) had it after development of symptoms leading to burst of abscess and subsequent sinus formation. 7(23%) of the 31 patients with relevant data had history of contact with a case of tuberculosis or had family history of tuberculosis. 5 (16%) cases had family history of tuberculosis while as 2 (6%) had contact outside the family. Of the 61 cases, 4 patients (6.56%) were actually immigrants from tuberculosis endemic regions.

Of the 61 cases of TD, 45 cases had only hand involvement, 12 had foot involvement and 4 had lesions both in hand and foot. 5 cases, 4 of hands and 1 of feet, had bilateral involvement. More than one bone affected by TD was seen in 13 cases and 9 of these 13 cases with multiple TD lesions were of 6 years or younger.

In 61 cases, 77 was the total number of bones involved by TD, 59 (77%) in hands and 18 (23.38%) in feet. In hands, 37 (63%) bones involved were phalanges and 22 (37%) metacarpals. Most commonly involved bones of hands were second metacarpal (n = 8), proximal phalanx of middle finger (n = 8), proximal phalanx of ring finger (n = 7), first metacarpal (n = 7) and proximal phalanx of index finger (n = 6). There was no case involving distal phalanx of fingers (Figure 4a).

In feet, 8 (44%) bones were phalanges and 10 (56%) were metatarsals. Most commonly involved bones were first metatarsal (n = 8) and and proximal phalanx of first toe (n = 3). Distal phalanx of toes was not involved in any case (Figure 4b).

Figure 4: a) Distribution of involvement of the bones of hand. **b)** Distribution of involvement of the bones of foot.



Radiological Findings: Radiological picture of TD varied with a combination of different radiological signs. Different radiological findings in 77 bones of hand and feet are enumerated (Table I).

Associated tuberculosis lesions: In 50 cases of TD, 33 had associated tubercular lesion at another site. Pulmonary tuberculosis was the most common associated lesion seen in 19 cases followed by osteo-articular tuberculosis at other sites in 10 and tuberculous lymphadinitis in 9cases. 4 patients had skin involvement in the form of lupus vulgaris or scrofuloderma.One patient had associated tuberculous tenosynovitis of flexor sheath of hand and another had associated tuberculous abscess of the temporal lobe of brain.

Differential diagnosis: Of the 54 cases, initial diagnosis was TD in 41 and correct diagnosis was missed in 13 cases. The most common pathology which mimicked TD was pyogenic osteomyelitis (n=5) followed by tumors, syphilitic dactylitis, fungal or parasitic infections and metastatic pyogenic abscess.

DISCUSSION

Extra pulmonary tuberculosis (EPTB) constitutes 15–25% of all the tuberculosis cases [9]. In USA, the most frequent forms of extra- pulmonary tuberculosis are lymphatic gland (41.3%), pleura (20.7%), and bone and joint (11.2%) [9]. Skeletal tuberculosis constitutes less than 2 % of all tuberculosis cases [10]. TD, also known as spina ventosa (Spina="a thorn"; Ventosa = "full of

Table I: Different radiographic findings in 77 bones with tuberculous dactylitis

RADIOLOGICAL FINDING	NUMBER OF BONES (%)
Lytic lesions	53 (68.8%)
Expansion or ballooning of the bone	42 (54.5%)
Mild periosteal reaction	32 (41.6%)
Soft tissue swelling	31 (40.3%)
Cortical erosions	10 (13.0%)
Cortical destruction or cortical breach	8 (10.4%)
Sclerosis	4 (5.2%)
Involvement of adjacent articular surface	3 (3.9%)
Severe perisoteal reaction	2 (2.6%)

wind or distended"), is a rare form of tuberculous osteomyelitis involving phalanges, metacarpals and metatarsals [1]. It constitutes 4 – 8% cases of skeletal tuberculosis [7, 11]. Literature on this entity is scarce and there are not many TD reports [1, 12, 13, 14]. Eighty-five percent cases of TD are younger than 6 years [2]. In our literature review, 83% cases were 30 years old or younger with a second peak at later age. As the life expectancy is expected to increase further, the second peak is likely to expand further in the near future due to associated co-morbidities and compromised immunity with increasing age.TD is reported to be three times more common in males than females [7]. However, in the present review, we found only a slight preponderance of males with a male to female ratio of 1.26. The presenting complaint is either swelling and pain not responding to analgesics or a painless swelling [2, 15, 16]. Soft tissue swelling associated with osseous lesions of foot is minimal because bones of feet are covered by tendons and tight facial sheaths which resist distension [17].

Tuberculosis of bone often becomes symptomatic few months to 3 years after the initial infection [6, 18]. There is often a long delay in the diagnosis of osteo-articular tuberculosis with a mean delay of 5 to 12 months [15, 16, 19]. One reason for the delay in diagnosis of osseous tuberculosis is due to the paucibacillary nature of the lesion [12].

Other factors for the diagnostic delay of TD are attributed to insidious onset, slow progression, paucity of symptoms, non-specific clinical manifestations, occurrence of disease in absence of pulmonary tuberculosis, non-specific nature of radiographic findings, lack of rapid microbiological diagnostic method, occasional inconclusive microbiological and histopathologic findings, and poor awareness among treating physicians and surgeons who do not have much experience with this disease [8, 11, 15, 16, 19, 20-24]. In our review series, there was a mean delay in presentation of 6 months.

In this review case series, 30.6% patients had some underlying predisposing factor for tuberculosis. Low socioeconomic status and immunodeficiency were the most common predisposing factors followed by malnutrition. In a case series of 66 patients with extra-spinal osteo-articular tuberculosis by Ali R et al, low socioeconomic status was the most common predisposing factor seen in 90% of their patients [19]. In our review, immune deficiencies that predisposed to TD included severe combined immunodeficiency (SCID), chronic granulomatous disease (CGD) and acquired immune deficiency such as HIV infection. All patients with congenital impairment of immunity (such as SCID or CGD) developed tuberculosis in infancy. Orthopedic surgeons should have low threshold for considering this form of tuberculosis of bone because of an increase in the number of HIV positive and immune suppressed children [25, 26, 27]. 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 [28, 29]. Less than one-tenth of our cases had history of trauma prior to the development of symptoms. In tuberculosis of foot there is prior history of trauma in one-third of the patients but significance of trauma is unclear [17].

In our review series 23% cases of TD had history of contact with tuberculosis. In the case series by Ali R et al of extra-spinal osteo-articular tuberculosis, 18.18% had positive family history which is comparable to that of our literature review [19]. 6.56% cases in our review were emigrants from tuberculosis endemic regions. The incidence of tuberculosis has increased in industrialized countries and one of the factors attributing to it is immigration from countries with high prevalence of tuberculosis [11,30,31].

Single or multiple discharging sinuses were present in 30% cases in our review. 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 [7]. Ali R, et al in their series of extra-spinal osteo-articular tuberculosis had a discharging sinus present in 20% [19]. Comparing it with our review series, sinus formation is slightly more common in TD than in other forms of extra-spinal osteo-articular tuberculosis.

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 [2, 7, 32]. Involvement of toes is less common [7]. In our review, bones of hand were four times more commonly involved than those of the foot with only 7% cases having simultaneous involvement of hand and foot. 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 TD of fingers or toes. About 21% cases had TD of more than one bone and two-third of these cases had age of 6 years or less in our review series. Multiple site involvement is seen in 25 to 30% and it is more often reported in children than in the adults [7, 11, 31, 33]. Radiological findings in TD 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 [22]. 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 [1,2,34,35]. Marked periosteal reaction is seen in cases with sinus formation due to associated secondary pyogenic infection of which periosteal reaction is a hallmark [17]. 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 [33]. In our review series, ESR was elevated above normal range in 80% and Mantoux test or PPD intra-dermal skin sensitivity test was positive in 75% cases. Raised ESR is a non-specific marker and Vohra et al had raised ESR in 88% cases in their series on extra-spinal osteo-articular tuberculosis whereas Rasool et al had raised ESR in 60% cases [15,36]. Mantoux positivity varies from 60 to 80% in multifocal osteo-articular tuberculosis [37]. In Ali R et al series of extra-spinal osteo-articular tuberculosis, Mantoux test was positive in 62% (26 out of 42) patients [19].

TD 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, tumors, hemoglobinopathies, endocrinopathies, metabolic disorders and others [4,5,6,7,8]. Among infections, it resembles pyogenic osteomyelitis, brodie abscess, fungal osteomyelitis, parasitic infections, atypical mycobacterial infection, syphilitic dactylitis, brucellosis and Madura mycosis [1,4,7,13,14,38]. Tuberculosis of short tubular bones may be confused with tumors, both benign and malignant. Among benign lesions it resembles enchondroma, cortical fibrous defects (mono ostotic fibrous dysplasia), aneurysmal bone cyst, giant cell tumor, chondroblastoma, osteoid osteoma, florid reactive periostitis, pagets disease, eosinophillic granuloma (histiocytosis X) [1,5,7,8,14,39]. Malignant lesions include secondary deposits, Ewings sarcoma, osteosarcoma, myeloma, leukemia, malignant giant cell tumor, Kaposi sarcoma [4,5,22,40]. It also mimics chronic granulomatous disease like sarcoidosis [5,6,13,3]. Hemoglobinopathies such as sickle cell disease can present as sickle cell dactylitis which mimics TD [4]. Among endocrinopathies and metabolic disease it resembles hyperparathyroidism (brown tumor) and gout respectively [4-6]. Rarely, it can be confused with hereditary acro-osteolysis [14].

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 EPTB [19]. Statistical data is not available in literature pertaining to any rise in the incidence of TD in the recent era.

CONCLUSION

Diagnosis of TD should always be kept in mind while dealing with pathology of short tubular bones of hand and feet as it is often missed because of usual absence of stigmata of tuberculosis in other parts of the body especially lungs, absence of constitutional symptoms and clinico-radiological mimicking with other infections, tumors, endocrinopathies, metabolic disease, hemoglobinopathies and chronic granulomatous disease. Any delay in diagnosis and treatment of TD will likely decrease the chances of good functional outcome.

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