

CASE REPORT

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# Bilateral tuberculous dactylitis of both hand and feet in a female toddler: a case report on a rare presentation of skeletal tuberculosis in children

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## Abstract

**Background** Even though tuberculosis is a common disease among children in developing countries, tuberculous dactylitis is an uncommon form of Skeletal tuberculosis specially with involvement of both the hands and feet.

**Case Presentation** A one-and-a-half-year-old previously healthy female Ethiopian toddler presented to our pediatric outpatient clinic with a history of two-month duration of painful multiple swellings over both her hands and feet. The swelling involved the proximal phalanx of the left index finger, dorsum of the right hand, and dorsum of both feet over the first metatarsal bone. Physical examination, radiologic findings, and histopathology suggested tuberculous dactylitis. The patient was treated with anti-tuberculosis drugs for one year and she showed clinical and radiologic improvement and recovery.

**Conclusion** Tubercular dactylitis should be considered in the differential diagnosis of children from endemic areas presenting with bone and joint pain or swelling. Our experience of a twelve-month course of antitubercular treatment, which is in line with WHO recommendations, for skeletal tuberculosis, showed excellent outcomes.

**Keywords** Tuberculous dactylitis, Extra-pulmonary tuberculosis, Skeletal tuberculosis, Tuberculosis, Spina ventosa

## Background

Tuberculosis is an infectious disease caused by *Mycobacterium tuberculosis* and is characterized by the formation of granulomas with caseous necrosis histopathologically. Its commonest presentation is pulmonary involvement. Skeletal tuberculosis (TB) accounts for 10 to 20% of extrapulmonary tuberculosis (EPTB), with Pott's disease and tuberculous arthritis being the two most common forms [1].

TB dactylitis is a term used when it involves the bones of the hands and feet. It usually presents with soft tissue swelling with or without pain and radiographically appears as expansile cyst-like cavities with cortical bone

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destruction along the diaphysis. It is usually spread haemogenously or via the lymphatic system. TB dactylitis is a very uncommon form of EPTB in children. Around 85% of patients with TB dactylitis are below 6 years of age [2]. It usually involves the short bones of the hands than feet and multifocal involvement of both hands and feet is a rare occurrence of TB dactylitis.

The diagnosis of TB dactylitis is usually made based on radiologic, histopathologic, and in rare cases bacteriologic results. Juvenile inflammatory arthritis, Bacterial osteomyelitis, leukemias, and syphilitic dactylitis are among the differentials that are usually considered initially in patients with TB dactylitis.

### Case presentation

A 1-year and 6-month-old female Ethiopian toddler presented at the outpatient department with complaints of swelling on the left proximal index finger, dorsum of the right hand, and dorsum of bilateral feet of 8 weeks duration. The swelling started in the right hand and involved bilateral feet and left proximal index finger within a week.

The mother noticed that the swelling was progressively increasing in size and over the last two weeks it has caused limping and pain over the feet which was aggravated by walking. The patient has a low-grade fever,

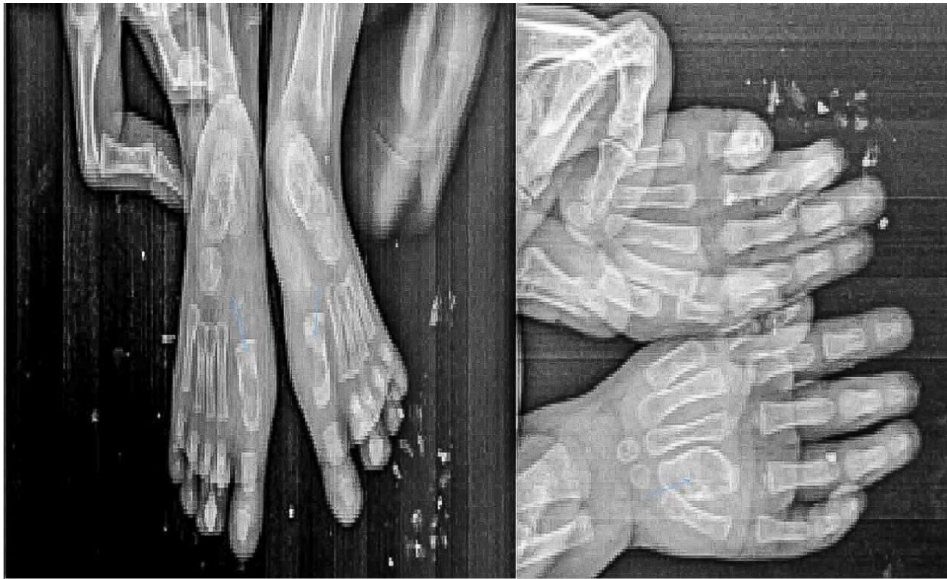
weight loss, and loss of appetite for the same duration. There was no family or personal history of similar illness and no contact with a chronic cough or TB-diagnosed patient. She was completely vaccinated for her age and had taken the BCG vaccine at birth.

On physical examination, the patient was moderately malnourished based on weight for length percentiles and her vital signs were within normal range. Musculoskeletal examination revealed tender, warm, erythematous swelling at the dorsum of the bilateral foot over the first metatarsal bone with no discharge. There was also a non-erythematous, tender, warm swelling at the dorsum of the right hand over the 4th metacarpal bone with no discharge and a similar swelling of her left proximal index finger (Fig. 1).

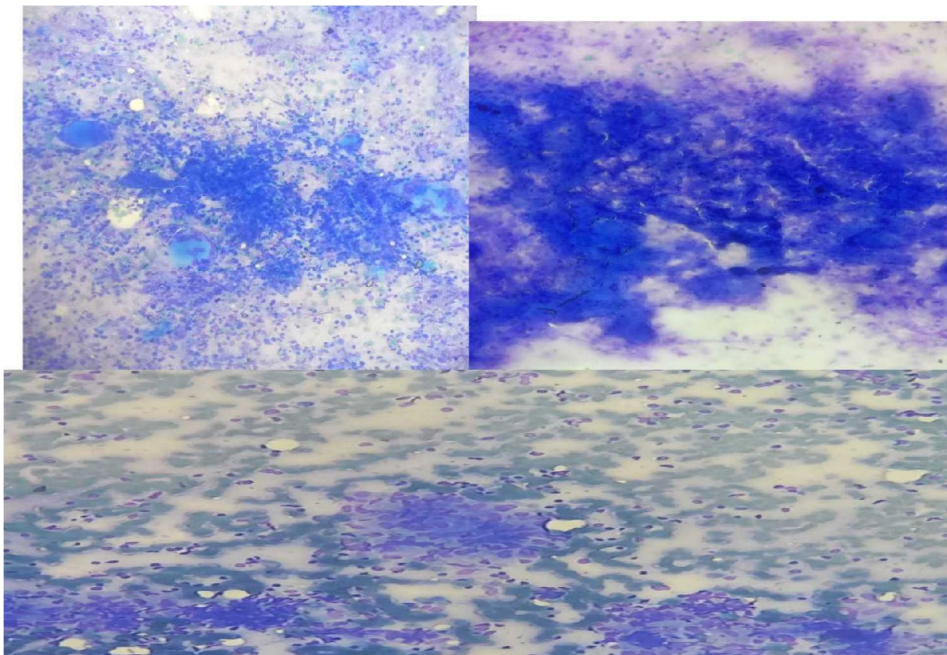
With a provisional diagnosis of osteomyelitis, the patient was admitted for further workup. Her complete blood count showed mild microcytic anemia, ESR was 62, and Gene-Xpert from gastric aspirate was negative. Chest radiography was normal. Anteroposterior radiography of bilateral feet and hands showed lytic lesions with cortical erosion and normal joint spaces (Fig. 2). Further imaging of the extremities with MRI was planned but could not be done because the MRI machine was out of service. Finally, FNAC from bilateral feet and right hand



**Fig. 1** Erythematous swelling at the dorsum of the bilateral foot over the 1st metatarsal bone, swelling at the dorsum of the right hand over the 4th metacarpal bone, swelling of the left proximal index finger



**Fig. 2** A bilateral mildly expansile lytic lesion with cortical erosion of metatarsal bone with adjacent soft tissue swelling. A diaphyseal expansile lytic lesion with cortical erosion over the right 4th metacarpal bone



**Fig. 3** Smears show epithelioid granuloma with multinucleated giant cells admixed with heterogeneous lymphoid cell population against a necrotic background

was taken and it showed granulomas with necrotic backgrounds (Fig. 3). Gene-Xpert, drug susceptibility tests, and culture from the sites were taken but were negative.

After consideration of the radiologic and cytology findings, tuberculous dactylitis was diagnosed and the patient was started on anti-tubercular drugs. After 3 weeks of treatment, the patient showed significant improvement in her symptoms as the swellings decreased and she was able to walk without pain. After three months

of treatment, the swellings were fully resolved and radiology showed marked improvement as well (Fig. 4). A full 12-month course of anti-TB treatment was completed successfully. She was put on a daily regime of isoniazid, rifampicin, ethambutol, and pyrazinamide for two months, followed by daily isoniazid and rifampicin, for ten months in accordance with current national and WHO guidelines for the treatment of osteoarticular TB. The patient was followed up after 1 year of treatment





**Fig. 4** Follow-up evaluation shows marked clinical and radiological improvement after 3 months of Anti-TB

completion and there were no signs of deformity or recurrence.

### Discussion and conclusion

Skeletal TB occurs in 1–5% of children with primary pulmonary TB. Spread to the skeletal system usually occurs through the lympho-hematogenous route and becomes symptomatic in the first few years after the initial infection [3]. In highly endemic areas, skeletal TB manifests in relatively young patients [4]. Around 50% of patients with skeletal TB have concomitant clinically evident primary pulmonary TB, but others might not have a frankly evident primary focus of infection like that of our patient [5].

Tuberculous osteomyelitis, which is a form of skeletal TB, is less common than TB spondylitis and arthritis and has been reported to occur in only 11% of children with skeletal TB [6]. TB dactylitis is even an uncommon manifestation and its incidence among children with TB is reported to be 0.65–6.9% [2]. It is usually common in children less than 6 years old.

The commonest presenting symptom of TB dactylitis is swelling with or without pain [5, 7]. One case series and literature review of 61 patients with TB dactylitis mentioned that swelling was the chief presenting complaint in 96% of cases of which 57% had accompanying pain [8]. Other presenting features include draining sinus tracts and nonhealing ulcers. The duration of symptoms ranges from a few weeks to several months.

TB dactylitis also has a more solitary and localized presentation than a multifocal one [9]. The bones of the hands are more commonly involved than the feet [10], with the metacarpal and proximal phalanx of the index and middle finger being the most commonly affected [11, 12]. Involvement of the feet is very uncommon. A case report mentioned an isolated involvement of the foot with accompanying draining sinus [13].

Multi-focal involvement of both hands and feet is even rare. A case was reported of an adult male with involvement of the right toe and fingers of both hands [14]. Another case report mentioned a 4-year-old with involvement of the first metacarpal of the right hand

and the first metatarsal of the left foot [15]. 4 other cases of multifocal involvement of both hands and feet were reported by a case series [8]. Our patient presented with bilateral involvement of all four of her distal extremities which makes it an even rarer presentation.

The greatest challenge in the diagnosis of TB dactylitis is considering the diagnosis in the first place. This is especially true for patients presenting from non-endemic countries and without typical pulmonary disease. Diagnosis of TB dactylitis is usually suspected when a combination of clinical, radiologic, and histopathologic assessments are done.

The diagnosis can be established by microscopy, culture, and gene-Xpert studies of infected material. The performance of the Xpert MTB/RIF in osteoarticular TB in children is promising. One study tested the performance of Xpert in 102 pediatric patients with osteoarticular TB and found that Xpert confirmed extrapulmonary TB of bone and joints more accurately and faster than culture, providing a sensitivity of 73.9% and specificity of 100% [16]. Culture from infected sites can also be utilized in the diagnosis of osteoarticular TB but results might take time. There are reports where AFB was grown from cultures aspirated from bony lesions [17].

Even though a wide range of radiological features might be seen in TB dactylitis, the commonest features of TB dactylitis are cystic changes in metaphyseal long bones and bony expansions [8].

Histopathologically, the caseous exudative type, which is characterized by bone destruction and caseous necrosis is more often seen in children than the granular type [18].

Anti-TB drugs are the mainstay of treatment and are usually effective in treating this condition but some may require concomitant surgical interventions. Although the optimal duration of therapy for the treatment of musculoskeletal TB is uncertain, we agree with the WHO recommendation of treatment with a 12-month course, with 2 months of four drug regimens (isoniazid, rifampicin, ethambutol, and pyrazinamide) followed by 10 months of 2 drug regimens (isoniazid and rifampicin). Longer therapeutic courses are being favored for musculoskeletal TB because of concerns about poor drug penetration into osseous and fibrous tissues.

Surgical interventions for musculoskeletal TB are usually reserved for those who fail to respond to pharmacotherapy, those with ankylosed and deformed joints, and those with recurrence [12]. Surgery can also be favored when there is significant bony destruction which might necessitate graft placement [19]. In general outcomes of skeletal TB are excellent and respond to anti-TB medications provided that early diagnosis and initiation of treatment are achieved with good compliance for completion of therapy.

In conclusion, our patient had an uncommon disease, TB dactylitis with an even rare bilateral involvement of the small bones of both hands and feet that has never been reported before to the best of our knowledge. Finally, TB dactylitis should always be considered in the differential diagnosis of children from endemic areas who present with swellings of hands or feet even without primary lung involvement or constitutional symptoms.

#### Abbreviations

TB	Tuberculosis
EBTB	Extrapulmonary tuberculosis
ESR	Erythrocyte sedimentation rate
FNAC	Fine needle aspiration cytology
WHO	World Health Organization

#### Supplementary Information

The online version contains supplementary material available at <https://doi.org/10.1186/s12879-024-09871-3>.

Supplementary Material 1

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#### Author contributions

A.W.G. was a major contributor in writing this manuscript. G.M., T.G and A.K.M. were involved with in initial clinical evaluation, setting the diagnosis and starting treatment during inpatient stay. I.O. was involved in taking, analyzing and reporting the histopathology results. A.W.G. and S.B.M. were involved in follow-up evaluation of the patient.

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#### Data availability

Data is provided within the manuscript.

#### Declarations

##### Ethics approval and consent to participate

This study was conducted by the fundamental principles of the Declaration of Helsinki.

##### Consent for publication

Written informed consent was obtained from the patient's mother for the publication of this case report and any accompanying images. A copy of the written consent.

##### Clinical trial number

Not applicable.

##### Competing interests

The authors declare no competing interests.

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