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Case Report

Chronic multifocal tubercular osteomyelitis in a young Nepalese boy: A rare case presentation ☆,☆☆

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ABSTRACT

This study presents a rare case of chronic multifocal tubercular osteomyelitis in a 13-year-old boy from Nepal, a high-burden country for tuberculosis (TB). He presented with chronic pain in the lower extremities and had no pre-existing comorbidities. The patient's clinical presentation, diagnostic process, and treatment plan are described. Multifocal skeletal TB, though infrequent, poses diagnostic challenges due to its variable manifestations. This case emphasizes the importance of considering TB in differential diagnoses, especially in endemic regions, necessitating a high index of suspicion. Early detection and treatment align with WHO's "End TB" strategy and Nepal's TB management guidelines, promoting improved outcomes in high-risk populations.

Introduction

Nepal is among the countries listed by the WHO as having a high tuberculosis (TB) burden, with an incidence rate exceeding 100 cases per 100,000 population [1]. Tuberculosis, caused by the organism *Mycobacterium tuberculosis*, remains one of the significant threats to the health of people in endemic regions [2]. The disease can exhibit a wide range of manifestations, spanning from latent infection to systemic dissemination [3]. Patients may present with both pulmonary and extrapulmonary forms of the disease [4]. Successful management depends equally on the effectiveness of antimicrobial agents and social determinants of health [2]. Skeletal TB refers to tuberculosis affecting the bones and/or joints, which is a form of extra-pulmonary tuberculosis [4]. The most frequently encountered type of skeletal TB is known as Pott's disease, primarily affecting the spine, followed by tuberculous arthritis [5]. Multifocal involvement is uncommon in immunocompetent individuals [6]. We are presenting a rare case of chronic multifocal tubercular osteomyelitis in a 13-year-old boy experiencing persistent symptoms and no associated comorbidities.

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Case details

Over the past year, a 13-year-old boy experienced intermittent episodes of limb pain without swelling or discharge. The most recent episode was 5 months ago, prompting him to limit mobility and seek traditional remedies. However, in the last 2 months, he visited our outpatient department due to new complaints. The left lower leg pain, with an insidious onset, hindered daily activities and was accompanied by progressively increasing swelling. Additionally, he reported discharge from the posterior aspect of his right thigh, described as watery, yellowish, and painless. Similar episodes in the past resolved spontaneously with home NSAID treatment and assistance from traditional healers. They had not identified any specific factors that exacerbated the episodes. However, the recurrence of symptoms, along with the discharge, had prompted the parents to become concerned that it might be something serious, prompting their visit to the hospital. Both the parents and the patient reported no fever, joint pain, weight loss, or loss of appetite. There were no changes in bowel and bladder habits, skin or nail changes, chest pain, shortness of breath, trauma, or abnormal body movements. A systemic review did not reveal any significant findings. He had no known or previously diagnosed medical or surgical conditions, and he had no allergies. There were no significant chronic illnesses or malignancies in his family history. Neither the patient nor any close contacts had ever received a tuberculosis diagnosis. Additionally, he had not traveled recently and had completed all his childhood vaccinations as per the schedule of Nepal.

Upon examination, the patient appeared well-nourished and oriented to time, place, and person but was in mild distress. His temperature was recorded at 99.8 degrees Fahrenheit, with a pulse rate of 88 beats per minute and a respiratory rate of 18 breaths per minute. He maintained an oxygen saturation level of 96% while breathing room air. A non-pulsatile swelling, measuring approximately 2 × 2 cm, was observed on the anterior aspect of the left leg, situated 4 cm proximal to the ankle joint. The swelling exhibited clear margins, and the skin covering it appeared smooth with no signs of erythema or color changes. Palpation did not reveal any fluctuation or tenderness; however, a slight increase in temperature was detected (Fig. 1A). During the examination of his right thigh, a draining sinus, measuring 1 × 1.5 cm, was identified on the posterolateral aspect of his right thigh, located 5 cm proximal to the knee joint. This sinus exhibited no tenderness, and the skin surrounding it felt warm to the touch (Fig. 1B). A watery discharge was observed. The chest examination indicated bilateral equal breath sounds with no additional sounds. The rest of the systemic examination yielded normal findings.

Based on the findings from the patient's history and examination described above, there was a suspicion of chronic osteomyelitis or a bone tumor. Taking these potential diagnoses into account, the following diagnostic tests were ordered: a Complete Blood Count (CBC), C-reactive Protein (CRP) test, Erythrocyte Sedimentation Rate (ESR) test, X-rays of the affected areas, and a chest X-ray. Additionally, an ultrasound was requested to assess the swelling and the draining lesion.

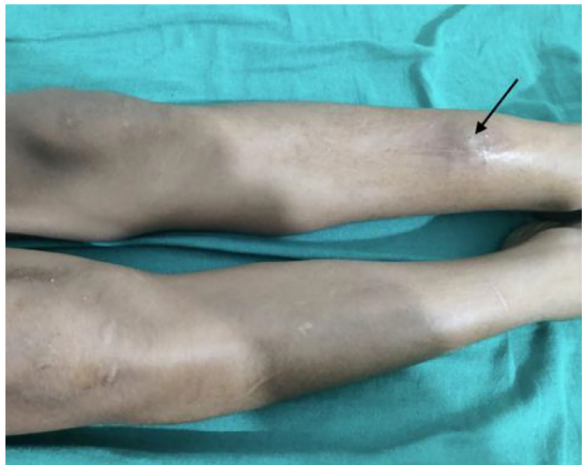

Fig. 1A – Photograph image of the bilateral lower limb of a 13-year-old boy showing the swelling with a tiny opening (black arrow) in the anterior aspect of the left leg.

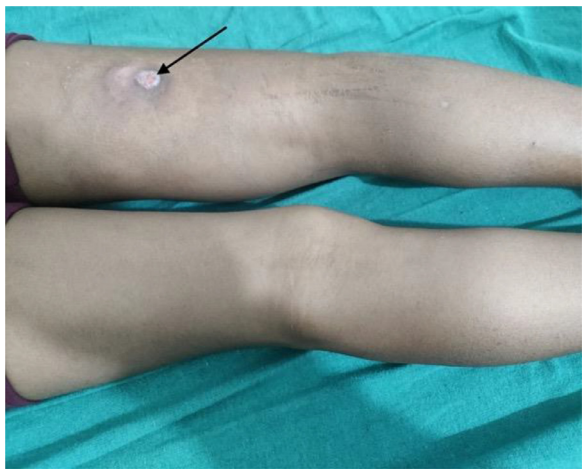

Fig. 1B – Photograph image of the bilateral lower limb of a 13-year-old boy showing the swelling with an opening (black arrow) in the posterolateral aspect of the right thigh.

The lateral X-ray of the left leg revealed cortical irregularity with increased thickness and areas of radiolucency within the meta-diaphyseal region. Adjacent soft tissue swelling is also noted (Fig. 2A). The anteroposterior X-ray of the right thigh showed obliteration of the medullary cavity with increased cortical thickness and foci of radiolucency with fistulous tract and adjacent radiodense sequestrum in the meta-diaphyseal region along with adjacent soft tissue swelling (Fig. 2B). Additionally, the ultrasonography findings were significant for fracture with cortical irregularity of the left distal leg with minimal hypoechoic subperiosteal collection, (Fig. 3A) and right thigh showing the hypoechoic subperiosteal collection with draining sinus tract running via the muscle plane, (Fig. 3B). The discharge however was watery reflecting the possible filtrate of the collection oozing out. Elevated leucocyte count and ESR levels were noted (Table 1). After discussing the findings with

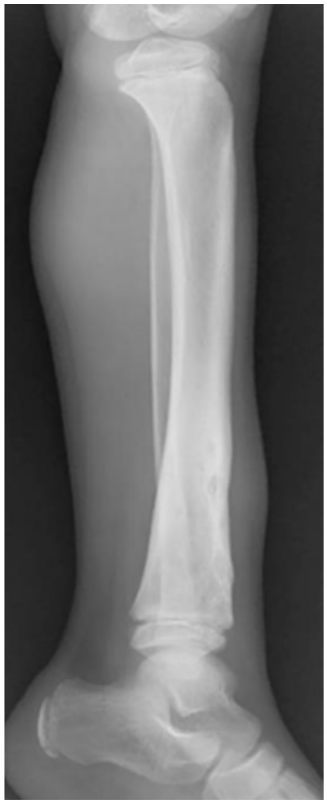

Fig. 2A – X-ray lateral radiograph image of left leg showing the cortical irregularity with increased thickness and areas of radiolucency within the meta-diaphyseal region. Adjacent soft tissue swelling is also noted.

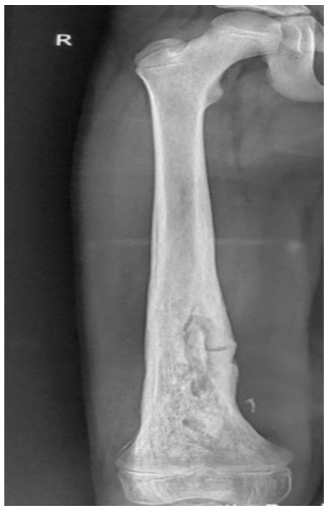

Fig. 2B – X-ray anteroposterior radiograph image of the right hip showing the obliteration of the medullary cavity with increased cortical thickness and foci of radiolucency with a fistulous tract in the meta-diaphyseal region. Adjacent soft tissue swelling is also noted.

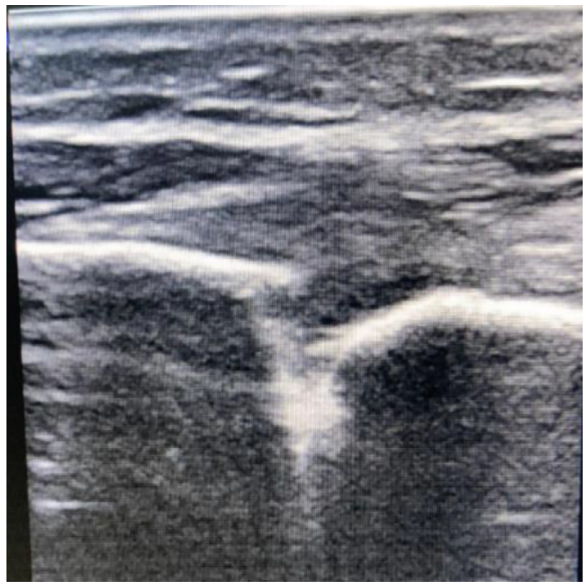

Fig. 3A – Greyscale ultrasound image showing the fracture with cortical irregularity of the right femur with minimal hypoechoic subperiosteal collection.

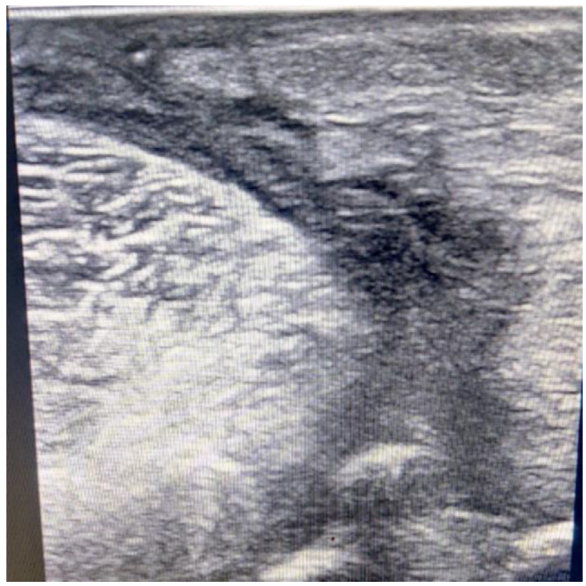

Fig. 3B – Greyscale ultrasound image of the right thigh showing the hypoechoic subperiosteal collection with draining sinus tract running via the muscle plane.

the patient and the parents, it was decided to perform a bone biopsy from the left leg as well as a bone biopsy with a collection of samples from the subperiosteal collection on the right. These samples were then sent for both microbiological and histopathological examination. The microscopic analysis did not reveal the presence of acid-fast bacilli, but the GeneXpert test yielded a positive result with no resistance to Rifampicin from the specimen from both sites. Additionally, the histopathological evaluation ruled out malignancy.

Table 1 – Hematological and biochemical findings of the patient.

Examination	Result	Reference range
CBC		
Total leucocyte counts	10.5 thou/ul	4.5-11.0 × 10^9/L
Neutrophil	35	40%-70%
Lymphocyte	60	20%-45%
Monocyte	3	2%-10%
Eosinophil	2	1%-6%
Hemoglobin	13.4 g/dl	12-14g/dl
RBC count	5.02 mill/cumm	4.5-5.5mill/cumm
PCV	41	35-54
M.C.V	82 fl	80-100fl
M.C.H	27 pg	26-34pg
M.C.H.C	33%	32%-36%
RDW	12.3%	11%-16%
Platelets count	291,000 thou/ul	150,000 -450,000
Serum		
Bilirubin (total)	0.39mg/dl	0.3-1.2mg/dl
Bilirubin (direct)	0.23mg/dl	0-0.3 mg/dl
AST	30 U/L	10-34 U/L
ALT	50 U/L	7-56 U/L
Protein	5.5 g/dL	6-8.3 g/dL
ESR	130mm/hr	<15mm/hr
CRP	25mg/dl	<10mg/dl

Based on the results of the investigations mentioned above, the patient received a diagnosis of chronic multifocal tubercular osteomyelitis. The family was once more asked if there had been any exposure to TB, but they denied any such exposure. Both the parents and the patient were informed about the diagnosis and the causative agent. The healthcare team explained the chronic nature of the condition and the necessity for extended treatment duration. After both the patient and the parents understood the disease additional blood investigations like serology for Human Immunodeficiency Virus (HIV) and Liver Function Test (LFT) were done. The treatment was started with antitubercular therapy consisting of Isoniazid, Rifampicin, Pyrazinamide, and Ethambutol along with Pyridoxine. This therapy was continued for 2 months and was planned to be completed with Isoniazid, Rifampicin, and Ethambutol for 10 months with a total of 12 months of therapy. Due to the presence of subperiosteal collection, debridement was done before the initiation of therapy. Regular follow-up was scheduled and the patient's symptoms and lesions were monitored clinically. Since the patient had a normal chest X-ray and had no cough, sputum examinations were not performed at diagnosis and follow-up. The patient's symptoms improved clinically with a reduction in ESR and leucocytes at 2 months after which the maintenance therapy was initiated. The patient is still under antitubercular therapy and regular follow-up with significant improvement in symptoms.

Discussion

Multifocal tuberculosis osteomyelitis, defined as the involvement of 2 or more noncontiguous skeletal regions, is a rare condition estimated to occur in 10%-15% of musculoskeletal tuberculosis cases [7]. Extraspinal musculoskeletal TB is among the least common manifestations of TB [7].

Multifocal tuberculous osteomyelitis is an uncommon condition and may involve any bone such as the skull, ribs, long bones, spine, and phalanx [8]. The presentation of extrapulmonary forms of TB can be very variable so a diagnosis should be in consideration in high-risk individuals and the endemic areas of the world. Extraspinal musculoskeletal TB and cutaneous TB are among the least common manifestations of TB, with a frequency of about 1%-2% for each [7]. Tuberculous spondylitis is the most common form of musculoskeletal TB and accounts for approximately 50% of cases [9]. Immunocompromised patients have an increased risk of developing extrapulmonary tuberculosis [2]. Prevalence is higher, especially in HIV-infected, hemodialysis patients and patients who are on immunosuppressive therapy [10]. The occurrence of multifocal lesions in an immunocompetent individual is unusual [6].

The pathogenesis mostly includes bacilli from primary M. tuberculosis infection into bone and/or synovial tissue [11]. Active TB can develop immediately or after years of latent infection. Although a pulmonary focus is often presumed, active pulmonary TB is seen in less than 50% of the patients like in our patient [3]. A retrospective review done in Uganda revealed the prevalence of concurrent pulmonary and extrapulmonary TB to be 8.5%.[12] The bone and joint involvement is mostly described in 2 categories. The caseous exudative type is characterized by bone destruction, local swelling, abscess formation, sinus formation, and constitutional symptoms. This type is mostly seen in children [13]. The granular type is more insidious, less destructive, and more common in adults [13]. However, the pattern of disease seen in TB is dynamic and often has mixed patterns. Contiguous spread from the primary site can also be one of the modes of disease pathogenesis [11].

Radiographic imaging can be a useful tool to establish the anatomical locations of musculoskeletal TB but there are no pathognomic radiographic findings [14]. Tubercular osteomyelitis in children can demonstrate cystic changes. Other bacterial osteomyelitis, fugal osteomyelitis and bone tumors are other potential diagnoses to be kept under consideration. Chest radiography is not a sensitive test for diagnosis of skeletal TB however it is obtained to guide the other aspects of disease management like decisions regarding respiratory isolation and collection of sputum samples.

The treatment modality circles around a long course of antimicrobial therapy. Treatment may require surgical interventions like debridement and curettage as required depending upon the extent of bone involvement [14]. Data are limited on the optimal drug regimen and duration of treatment. Nepal has its guidelines for the diagnosis and treatment of TB which aligns with the "End TB" strategy of WHO. The current treatment guidelines for treating musculoskeletal tuberculosis include a 2-month induction therapy with the 4 agents Isoniazid, Rifampicin, Pyrazinamide, and Ethambutol followed by maintenance therapy with Isoniazid, Rifampicin, and Ethambutol for 7 to 10 months [4]. Assessment of immunosuppressed status like HIV infection, malnutrition, and measles infection have to be taken into consideration while treating TB in children. Follow-up is done frequently to assess medication

adherence, adverse drug events, and assessment of clinical symptoms [4].

Conclusion

This case highlights the need for vigilant tuberculosis diagnosis, especially in endemic regions. Emphasizing the importance of considering medical history, physical examination, and socio-demographic factors, such as socioeconomic status and contact history, in diagnosis is crucial. The variable manifestations make it vital for healthcare professionals in endemic areas to consistently consider tuberculosis in their diagnostic approach, regardless of immunocompetence. Knowing that tuberculosis responds well to appropriate antimicrobial agents underscores the critical need for timely diagnosis. Thus, healthcare professionals in endemic regions must maintain a high level of suspicion, even with unusual presentations, to ensure prompt and effective treatment.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Shailendra Katwal: Conceptualization, mentor and reviewer for this case report and for data interpretation; Aastha Ghimire: Contributed in performing literature review, writing the paper and editing; Rhea Bohra: Contributed in writing the paper. All authors have read and approved the manuscript.

Ethical approval

Ethical approval is not required for case reports in my institution (Patan Academy of Health Sciences, Bagmati Lalitpur) so ethical approval was exempted.

REFERENCES

[1] National TB prevalence survey, Government of Nepal Ministry of Heath and Population National TB Control Centre, 2018-19. https://www.who.int/docs/default-source/nepal-documents/policy-brief.pdf?sfvrsn=37b5881c_2. [accessed 14.08.2023]

[2] Tuberculosis, World Health Organization (WHO). https://www.who.int/news-room/fact-sheets/detail/tuberculosis [accessed 02.09.2023].

[3] Gideon HP, Flynn JL Latent tuberculosis: what the host "sees"? 50. Immunologic Research. p. 202–212. [accessed 02.09.2023] https://www.ncbi.nlm.nih.gov/pmc/articles/PMC3788603/.

[4] Government of Nepal Ministry of Health and Population Department of Health Services National Tuberculosis Centre. National tuberculosis management guidelines. 2019 http://nepalntp.gov.np/wp-content/uploads/2019/10/National-Tuberculosis-Management-Guidelines-2019_Nepal.pdf [accessed 04.08.2023].

[5] De Backer AI, Mortelé KJ, Vanhoenacker FM, Parizel PM. Imaging of extraspinal musculoskeletal tuberculosis. Eur J Radiol 2006;57(1):119–30.

[6] Thawani M, Hale E, Habte-Gabr E. Multifocal tubercular osteomyelitis: a case with atypical manifestations. Tuberc Res Treat 2011;2011:e483802.

[7] Nataprawira HM, Ediwan NA, Diana IA, Dwiyana RF, Febrina D. Multifocal osteomyelitic tuberculosis at rare locations with metastatic tuberculosis abscess. Am J Case Rep 2019;20:503–7.

[8] Lynn MM, Kukanesen JR, Khan AW. Troublesome tuberculosis: a case report on multi-focal tuberculous osteomyelitis in an immunocompetent patient. J Clin Med Res 2012;4(1):73–6.

[9] Garg RK, Somvanshi DS. Spinal tuberculosis: a review. J Spinal Cord Med 2011;34(5):440–54.

[10] Centers for Disease Control and Prevention. TB risk factors. 2019 https://www.cdc.gov/tb/topic/basics/risk.htm. [accessed 17.08.2023]

[11] Pigrau-Serrallach C, Rodríguez-Pardo D. Bone and joint tuberculosis. Eur Spine J 2013;22(Suppl. 4):556–66.

[12] Bone and joint tuberculosis. medilib.ir., UptoDate, https://medilib.ir/uptodate/show/7658 [accessed 03.09.2023].

[13] Kyagulanyi E, Mirembe J, Nantaayi B, Nalukenge S, Mukasa D, Tamale J, et al. The prevalence of concurrent pulmonary and extrapulmonary tuberculosis in Uganda: a retrospective study. Ther Adv Infect Dis 2022;9:20499361221107304.

[14] Leonard MK, Blumberg HM. Musculoskeletal tuberculosis. Microbiol Spectr 2017;5(2):1–3. doi:10.1128/microbiolspec.tnmi7-0046-2017.