

A Rare Presentation of Acute Multifocal Skeletal Multidrug-resistant Tuberculosis

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ABSTRACT

Tuberculosis (TB) involving the musculoskeletal system is unusual. An acute and multifocal noncontiguous presentation is diagnostically challenging as it simulates various other infections, hematological malignancies, and secondaries. In this study, we report a rare record of a 14-year-old girl who presented with multiple focal swellings on her body within a short duration of 6 days. The tuberculin skin test was exaggerated and the cartridge-based nucleic acid amplification test (CBNAAT) of the granulomatous aspirate revealed rifampicin-resistant TB. Magnetic resonance imaging (MRI) showed swellings with underlying bone erosions and cultures of pus aspirate from the right shoulder region turned out to be Rifampicin and Isoniazid resistant TB.

Keywords: Acute, Multidrug-resistant tuberculosis, Multifocal.

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BACKGROUND

Tuberculosis (TB) is a significant health concern in India and across the world. In 2020, an estimated 10 million people were diagnosed with TB globally of which 1.1 million were children.¹ However, TB involving the musculoskeletal system is a rare entity. It accounts for 1–2% of all TB cases and 10% of all extrapulmonary TB.^{2,3}

Multifocal tubercular swellings in an immunocompetent child without any pulmonary involvement are uncommon. We report an unusual case of an adolescent girl who presented with multiple noncontiguous mobile swellings on her body within a short duration of 6 days. This aberrant tuberculous manifestation mimics various benign, malignant, and infectious etiologies, spawning diagnostic difficulties and hindrances in treatment resulting in severe deformities and functional deficits.

CASE DESCRIPTION

A 14-year-old girl presented to us with multiple nontender, soft to firm, mobile swellings over her body within 6 days. Swellings were small on the onset and later progressed in size. The swelling was first noticed on the medial aspect of the right clavicle, then on the left side of the neck, near the right shoulder joint, on either side of the spine, and behind the knee. She had no history of fever or evening rise of temperature, cough, or contact with a person infected with TB, dyspnea, fatigue, trauma, or intake of any immunosuppressant drugs. She had a weight loss of 10% over 6 months.

On examination, she weighed 55 kg, was afebrile and her vitals were within normal limits. Multiple nontender, mobile swellings were present on the medial aspect of the right clavicle, posterolateral aspect of the right arm, left lateral side of the neck, paraspinal region, and right popliteal fossa as shown in Figure 1.

Swellings were soft to firm in consistency, and mobile with no visible signs of inflammation. A general physical examination revealed pallor. Systemic examination revealed nontender

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hepatomegaly 4 cm below the right costal margin, and the rest of clinical examination was unremarkable Figure 2.



Fig. 1: Swellings on both sides of the spine



Fig. 2: Swelling near the shoulder joint

Investigations revealed hemoglobin—10.6 gm/dL, white blood cells—11380 cells/cumm, platelet count 4.92 lakhs/cumm, C-reactive protein—51.4 mg/dL, erythrocyte sedimentation rate—86 mm/hour. Peripheral blood smear showed mild normocytic normochromic anemia. Liver function tests, lipid profile, antinuclear antibodies, serum electrolytes, chest X-ray, and ultrasound abdomen were normal. Real-time polymerase chain reaction and rapid antibody test for Coronavirus disease 2019 were negative. X-rays of the shoulder joint and clavicle showed osteolytic lesions. MRI showed multifocal osteolytic lesions in the upper metaphysis of the humerus, right sternoclavicular joint, multiple ribs, and left paraspinal region. Sputum for acid-fast bacilli, GeneXpert, and gram stain showed no organisms.

Tuberculosis (TB) skin test after 48 hours showed an induration of 22 mm. Pus aspirate of the swelling in the right shoulder was negative for bacterial culture within 48 hours whereas CBNAAT showed mycobacterium TB with rifampicin resistance. Pus culture for TB showed rifampicin and isoniazid resistance mycobacterium TB with sensitivity for fluoroquinolones and second-line injectables. She received injection of kanamycin 750 mg, clofazamine 100 mg, ethionamide 750 mg, ethambutol 1200 mg, pyrazinamide 1750 mg, isoniazid 900 mg, moxifloxacin 800 mg, and pyridoxine 100 mg. On follow-up, the size of the swellings remarkably diminished with the disappearance of popliteal swellings and improved general condition.

DISCUSSION

Tuberculosis (TB) involving the musculoskeletal system is unusual and accounts for 5% of skeletal TB in all ages.² Nearly 50% of patients with skeletal involvement have primary pulmonary involvement and are often immunocompromised. Tuberculous osteoarticular lesions arising at two or more locations is multifocal skeletal TB.⁴ Indumathi et al.⁵ has published a case report of a 15-year-old female child who had developed swellings on the right foot in a duration of 1 month and five chest X-rays were suggestive of military TB. In contrast, the patient in our study was immunocompetent with no pulmonary involvement or exposure to TB.

Mycobacterium TB disseminates from the lungs through the lymphohaematogenous course.⁶ These disseminated bacilli lodges in the small terminal arteries of the metaphysis of bones give rise to tuberculous lesions after one year of primary infection.⁴ The bone lesions have four elementary presentations—(1) cystic,

(2) infiltrative, (3) focal erosions, and (4) spina ventosa.⁷ Osteoarticular TB, when disseminated, gives rise to multiple foci, which may be destructive and is frequently confused with sarcoidosis, also known as Jungling's disease, which occurs secondary to primary lung focus. Its onset is subtle, with indeterminate signs and symptoms such as low-grade fever, weight loss, and decreased appetite. Hosalkar et al.⁸ conducted a retrospective study which included 18 children who were treated for tubercular skeletal lesions over a period of 5 years. They found that the average duration of symptoms was 6 months with only four children having a history of contact with active TB patients and five of them had significant weight loss. In our study, the patient had an acute presentation of multiple noncontiguous swellings for 6 days without any history of contact with a TB patient.

Children typically have multiple lesions with involvement of the peripheral skeleton, and primary metaphysis of the long bones. Soft-tissue swellings, tenderness, and limitation of movements are other joint exhibitions. Diverse children's radiological display of tuberculous bone lesions can mimic numerous conditions, including subacute and chronic osteomyelitis, bone cysts, cartilaginous tumors, osteoid osteoma, and granulomatous lesions, hematological disease, and malignant tumors. Rasool et al.⁷ reviewed the clinical records and radiographs of 42 children with osseous lesions of TB diagnosed and treated at King Edward VIII Hospital, Durban, South Africa, between 1984 and 1999. The majority were in the metaphyses of the long bone, the rest were on the diaphyses, the epiphyses, short tubular bones (hands, feet), flat bones (pelvis, clavicle), and in the small round bones (patella, talus, and navicular). Four basic patterns of bony lesions were seen—the most common type was the cystic type. Our patient presented with similar vague clinical signs and symptoms.

Plain radiographs routinely advised may not show any changes in the early stages of the disease. An MRI is a gold standard to determine osteolytic cystic lesions with or without sclerosis or reactive bone formation. Adults typically have sclerotic lesions compared to children.⁹ A study done by Rasool et al.⁷ published that all children showed good healing with minimal sclerosis.

Indeterminate clinical and radiological findings of these multiple lesions imitate hematological malignancies, rheumatological conditions, pyogenic and mycotic osteomyelitis, eosinophilic granuloma, chronic relapsing multifocal osteomyelitis, neuroblastoma, and secondaries. Multifocal tuberculous involvement of the musculoskeletal system is uncustomary and usually overlooked for malignancy. Fine needle aspiration cytology, tissue biopsy, culture, and sensitivity are essential to distinguish from malignancies.^{8,10} In a case report published by Dewan et al.,¹¹ A 3-year-old girl child presented with multiple bony lesions with hepatosplenomegaly and significant pallor, a differential diagnosis of multi-system Langerhans cell histiocytosis, polyostotic fibrous dysplasia, and multifocal TB was considered.¹²

Antitubercular therapy (ATT) for 6–12 months is fundamental and effective in treating 85–90% of cases. Surgery should be considered in 10–15% of cases not responding to ATT. The prognosis is altogether good with precise diagnosis and early management.¹³

Musculoskeletal multidrug-resistant (MDR) TB cases were identified and published more frequently prior to 2013 but such cases weren't published later. Such an acute and rare presentation of MDR TB skeletal lesions should be considered as a differential diagnosis in the upcoming time.

To conclude, we present a rare case of MDR TB acutely involving multiple musculoskeletal sites in an immunocompetent child without a primary pulmonary lesion and previous exposure to TB. Hence, diagnosing and promptly starting treatment is imperative to prevent any disability.

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