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Diaphyseal Tuberculosis of the Tibia in a Sickle Cell Anemia SS: A Case Report

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Abstract

Tuberculous diaphyseal osteitis of the tibia is rare in adults. We report a case of a 26-year-old female patient with sickle cell anemia SS. Physical examination revealed swelling of the left leg in the middle third. Laboratory tests revealed a systemic inflammatory response syndrome. Imaging showed endomedullary osteolytic lesions of the tibial diaphysis. Pathological examination concluded that tibial resection was required, revealing case-follicular tuberculosis, which was bone tuberculosis. Medical treatment was administered to the patient. At 15 months of follow-up, the examination revealed complete healing of the wound. Diaphyseal tuberculous osteitis of the tibia is rare, and its association with sickle cell anemia SS has not yet been described in the literature.

Keywords

Bone, Infection, Antibiotic, Haemoglobin

1. Introduction

Osteoarticular tuberculosis (OAT) accounts for 2% to 5% of all tuberculosis cases and 11% to 15% of extrapulmonary tuberculosis cases [1].

A distinction is made between osteoarthritis and osteitis-osteomyelitis, which are characterised by isolated bone involvement.

Tuberculous osteitis is typically more common in children [1]. Isolated diaphyseal involvement in adults is very rare.

We report one case of tuberculous osteitis involving the tibia in a sickle cell anaemia patient.

2. Patient and Observation (Case)

This is a 26-year-old female patient with sickle cell anaemia SS who was admitted

on September 8, 2024, for inflammatory pain in her left leg, located in the middle third, which had been developing for over a month. Physical examination revealed a deterioration of general condition, left-sided limping, walking with a pair of crutches, and minimal swelling in the anterior-internal aspect of the middle third of her left leg. The swelling was warm, painful on palpation, and the skin was shiny (Figure 1). The left knee and ankle were free and painless.

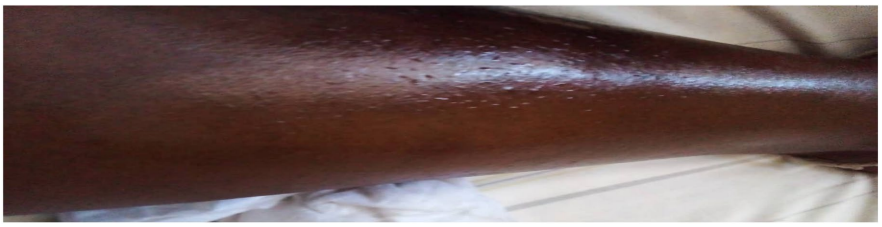

Figure 1. Patient's left leg.

Biology revealed systemic inflammatory response syndrome, an increase in the percentage of lymphocytes (44.4% of white blood cells), an anaemia with a hemoglobin level of 7.3 g/dL and C-reactive protein at 92. Imaging (X-ray and CT scan) revealed osteolytic lesions (lacunae) in the endomedullary region of the middle third of the tibial diaphysis (Figure 2 and Figure 3).

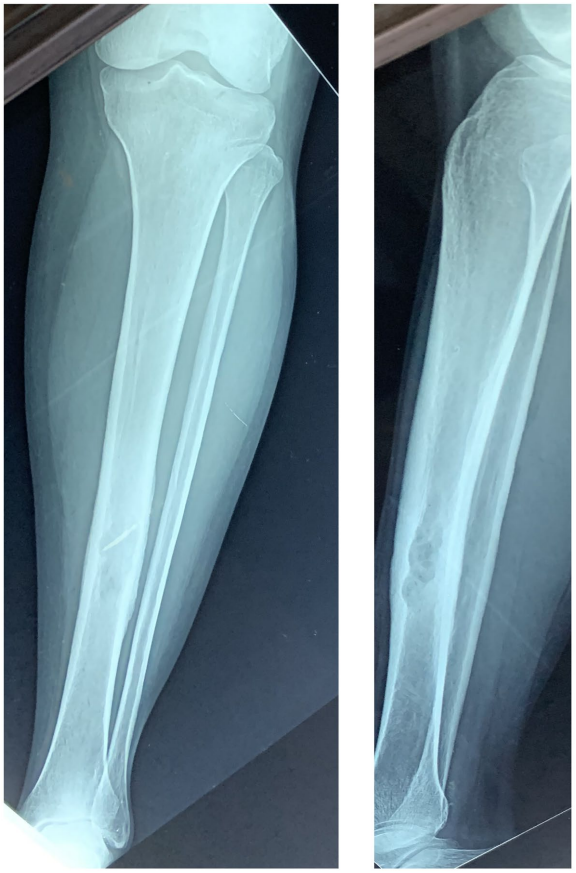

Figure 2. X-ray of the patient's left leg.

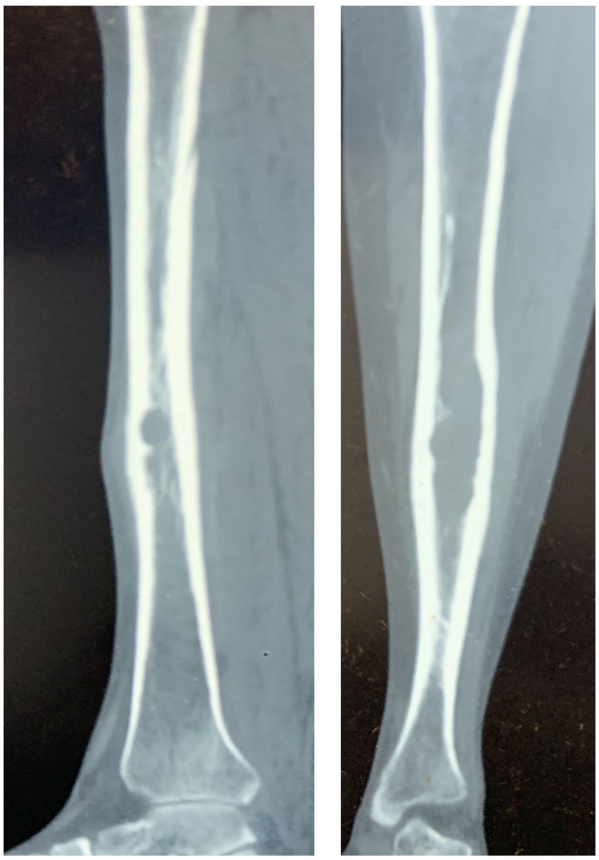

Figure 3. CT scan of the patient's left leg.

A surgical exploration that involved a bone biopsy, debridement, and irrigation following the creation of a cortical fenestration to expose the medullary canal revealing greenish-yellow pus in the intramedullary space with clots and bone remodelling.

Bacteriological tests on standard media were negative.

Three months after surgery (biopsy results not yet available), the condition had worsened, with loss of substance in the leg, exposure of the tibia, and abundant purulent secretions (**Figure 4**).

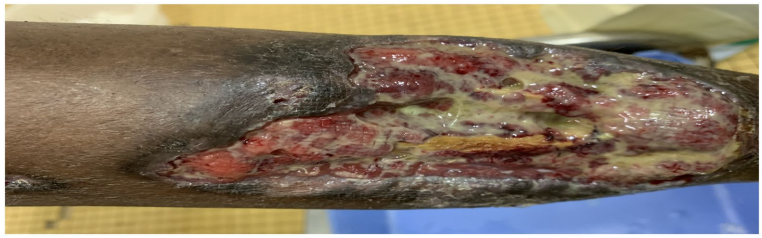

Figure 4. Loss of substance in the left leg and suppuration.

The pathological examination, the results of which were available four months after the biopsy, concluded that a tibial excision was required, showing caseous follicular tuberculosis at the caseofollicular stage (**Figure 5**).

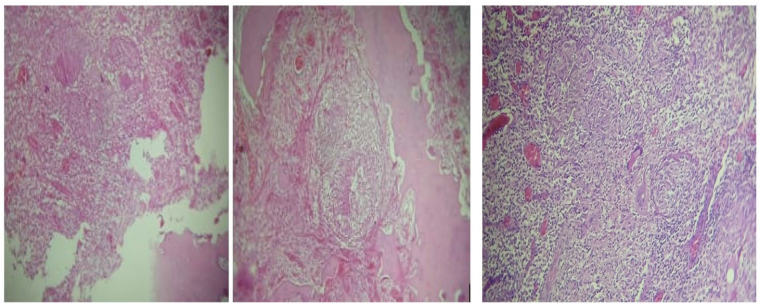

Figure 5. Histological sections showing tuberculous granuloma.

The patient was diagnosed with tuberculous osteitis of the tibia and received antituberculosis treatment (rifampicin, isoniazid, pyrazinamide, and ethambutol) for 12 months. The treatment regimen consists of rifampicin, isoniazid, pyrazinamide, and ethambutol for 2 months, followed by dual therapy (rifampicin and isoniazid) for 10 months.

Six months after the start of treatment, examination revealed complete healing of the wound (**Figure 6**). The X-ray showed no evidence of sequestration (**Figure 7**).

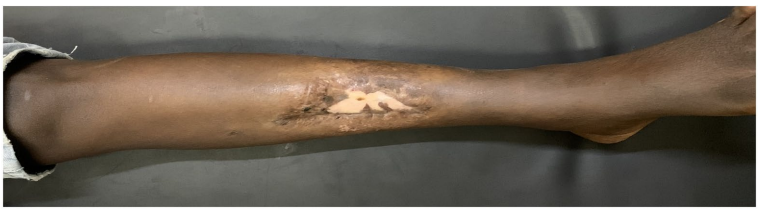

Figure 6. Patient's leg 6 months after starting treatment.

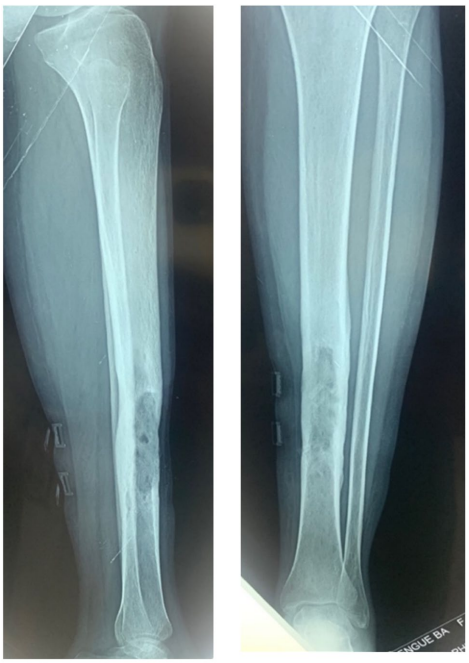

Figure 7. X-ray of the patient's left leg 6 months after the start of treatment.

After more than fifteen months of follow-up, the patient reports no pain in her left leg. The wound has completely healed (**Figure 8**) and the X-ray of the leg is normal, with no evidence of osteitis or bone sequestration (**Figure 9**).

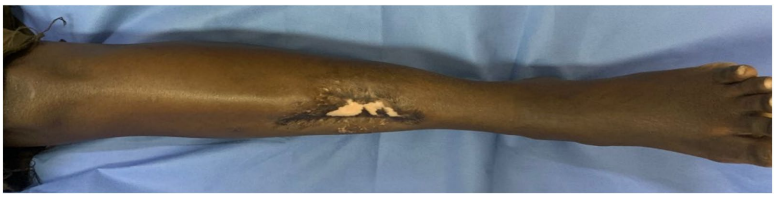

Figure 8. Patient's left leg 15 months of follow-up.

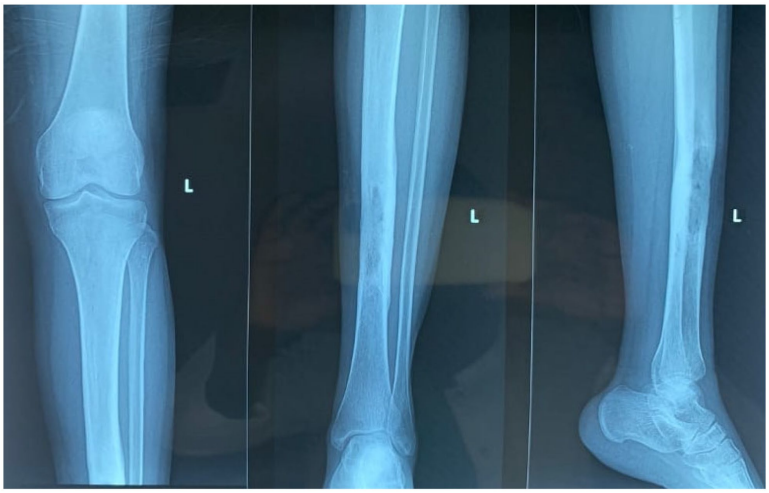

Figure 9. X-ray of the patient's left leg 15 months of follow-up.

3. Discussion

Tuberculous osteitis of the tibial diaphysis is very rare. The authors have described cases of osteoarthritis [2] and osteomyelitis [3] with metaphyseal-epiphyseal localisation [4].

The radiological lesions observed are not specific, hence the pseudo-tumoral appearance described in the literature by several authors [3] [4] which can lead to confusion with other pathologies and dangerous delays in diagnosis [5] [6]. In our patient, standard radiography and computed tomography (CT) revealed osteolytic lesions of the intramedullary lacuna type over a height of 12 cm in the middle third of the tibial diaphysis. These radiological images cannot formally rule out sarcoma. This pseudo-tumoral appearance can be confused with a malignant tumour, especially when cortical breach allows the infection to spread to adjacent soft tissues. Some authors highlight the value of MRI, which is of great interest in tuberculous osteitis, as it can show intra- and extra-osseous extension and the condition of the cortex, through a hyposignal in T1 and a hypersignal in T2 with signal enhancement after gadolinium injection [5] [7] [8]. Nevertheless, MRI remains the imaging modality of choice for the diagnosis and topography of the extent of tuberculous spondylitis and soft tissue tuberculosis [7]. The Ziehl-Neel-

sen stain was not performed, nor was the GeneXpert test. The HIV screening was negative. The diagnosis is based on the results of the histopathology. In a few cases, a combination of a negative BAAR test and a negative culture, along with a tuberculous granuloma on histopathological examination, was observed. [9]-[11].

This diagnosis is supported by the patient's favourable clinical and laboratory response to antituberculosis treatment. Biopsy is the only way to confirm the diagnosis with certainty by demonstrating the presence of a giant cell epithelioid granuloma with caseous necrosis [3] [12].

The femur and tibia are the bones most frequently affected after the spine. The diaphyseal location of tibia has been described by some authors [12]-[15] However, several other locations have been reported in the literature, with varied and sometimes rare topographies, including the fibula [16], patella [11] [17], ribs [18], radius, sternum [19], ulna, iliac wing [20] and metacarpal diaphysis [10]. Isolated diaphyseal localisation of the tibia is very rare, a few cases have been reported in the literature [12] [13] [15] [21]. Primary diaphyseal tuberculosis is rare, over a 27-year period (1955-1982), Richter R. and Krause F.-J. identified three cases of primary diaphyseal tuberculosis of the long bones, including one femoral case and one bilateral tibial case.

Elsewhere, cases of diaphyseal localisation in other long bones have been reported, notably a case of tuberculous diaphyseal osteitis of the radius in a woman in Morocco by Lazrek *et al.* [22] and another case of diaphyseal localisation in the third metacarpal bone of the hand by Bouabid *et al.* [10].

The association of diaphyseal tuberculous osteitis and SS sickle cell disease in adults to the best of our knowledge has not been found in the literature. A case of clavicular and metacarpal tuberculosis in a 2-year-old infant with SAFA2 sickle cell disease was reported by Toure *et al.* in Côte d'Ivoire [9].

The treatment of tuberculous osteitis is medical and surgical. Medical treatment is based on taking anti-bacillary drugs for a period of 12 months. In our case, treatment consisted of a combination of rifampicin, isoniazid, pyrazinamide and ethambutol for two months, followed by dual therapy (rifampicin and isoniazid) for ten months [6] [23].

A very high treatment success rate of around 96% was achieved [3].

4. Conclusion

Tuberculous osteitis of the tibial diaphysis is rare, and its association with SS sickle cell disease has not yet been described in the literature. Diagnosis is based primarily on histology. It should be considered a diagnosis of elimination in our context of endemic tuberculosis. Antituberculosis treatment should be initiated as early as possible.

Informed Consent

Informed consent from the patient for the publication of the case study details has been obtained.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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