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Exposed Tibia: Exceptional Mode of Revelation of Tuberculous Osteomyelitis

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Abstract
Isolated bone localization of osteoarticular tuberculosis is uncommon and their diagnosis is sometimes difficult explaining the delay in management. The authors report a new case of tuberculous osteomyelitis diagnosed late following a pathological fracture with exposure of the tibia in a 6-year-old child. The child initially presented with 8 months of insidious onset pain evolving into a pathologic fracture of the tibia with exposure of the tibial shaft. Standard radiography showed a diffuse osteolytic lesion of the tibial shaft associated with intraosseous abscess images. A biopsy was taken intraoperatively and confirmed the diagnosis of tuberculous osteomyelitis. The evolution of the wound was favorable after surgical debridement and total diaphysectomy followed by antitubercular chemotherapy. Unfortunately, the patient was subsequently lost, preventing the realization of a reconstructive surgery for a better functional recovery of his lower limb.

Keywords: Diaphysectomy, Tibial Osteomyelitis, Diagnostic Delay, Osteoarticular Tuberculosis, Exposed Tibia

1. Introduction
Tuberculosis is a major public health problem, especially in developing countries. Although Madagascar is a tuberculosis endemic area, isolated bone localization is not very frequent and their diagnosis is sometimes difficult explaining the delay in management. However, early treatment is essential to prevent major functional disability [1]. In this study, we report an unusual complication of isolated tuberculous osteomyelitis of the leg in children.

2. Observation
This is a 6-year-old boy who presented for consultation. The first manifestation of the disease was a leg pain that started eight months ago, insidiously, exacerbated by prolonged walking. A sudden deformity associated with a stabbing pain appeared when walking to school in the fifth month of evolution with total functional impotence, then appearance of a fistulization and externalization of a bone fragment in the leg. There was no particularity in his history. The vaccinations according to the current national scheme was up to date, in particular the BCG. On examination, the tibial diaphysis was exposed to a height of about 12cmm and necrotic appearance. The knee, ankle, and foot were edematous, but joint motion remained preserved (Fig 1). There was no vascular or neurological abnormality. A plain radiograph of the left leg revealed a diffuse osteolytic lesion involving the entire tibial shaft associated with intraosseous abscess images (Fig 2). The lung radiograph was normal. Biological examinations were in favor of an inflammatory syndrome with an accelerated globular sedimentation rate of 98 mm at the first hour, an elevated CRP of 26 and a hypochromic anemia of 7.8 g/dl hemoglobin.
Surgical treatment consisted of debridement of the necrotic soft tissue and total diaphysectomy of the necrotic part, leaving the metaphyseal areas in place.
Anatomopathological examination of the bone and soft tissue samples taken intraoperatively confirmed the presence of epithelioid granulomas and Langhans-type giant cells with caseous necrosis, as well as aspects consistent with tuberculous osteitis (Fig 3). Bacteriological examination on deep samples came back sterile.
An eight-month course of specific oral anti-tuberculosis treatment was then prescribed according to the regimen used in Madagascar, with a two-month intensive phase combining four anti-tuberculosis drugs (rifampicin, isoniazid, ethambutol, and

pyrazinamide) followed by a six-month maintenance phase combining two anti-tuberculosis drugs (isoniazid and thiacetazone). The left lower limb was immobilized with a splint for 2 months postoperatively.

The evolution was favorable with wound healing obtained after six months of treatment. Partial weight-bearing protected by English canes was resumed by the third month of treatment. A secondary reconstructive procedure was planned at the end of the medical treatment to minimize the functional impairments caused by the bone loss, but the child was subsequently lost to follow-up.

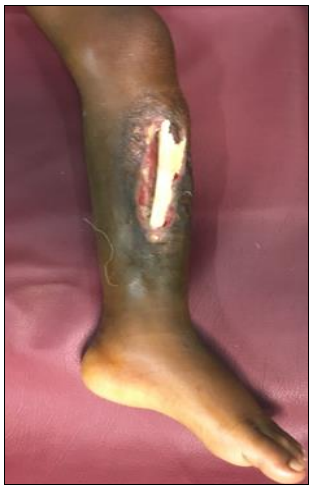

Fig 1: Pathological fracture of tibia; Exposed bone

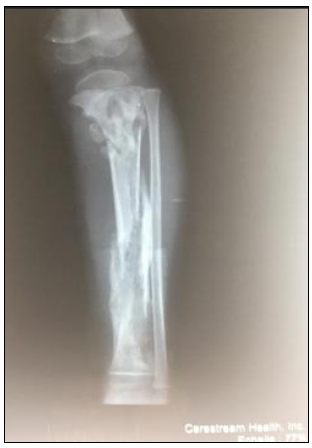

Fig 2: X-ray showing osteomyelitis of the tibial diaphysis

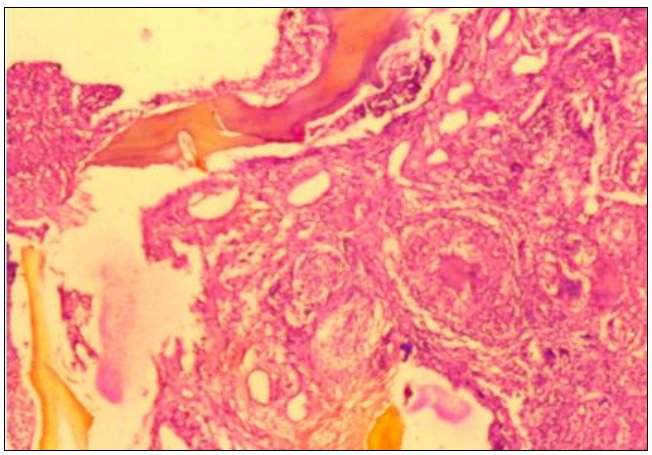

Fig 3: Presence of an epithelioid and gigantofollicular granuloma, histological section (hex100)

3. Discussion

Osteoarticular tuberculosis (OAT) represents up to 35% of extrapulmonary tuberculosis cases. It most commonly affects the spine, followed by tuberculous arthritis and extrarachondrial tuberculous osteomyelitis [2].

Very few studies report cases of pathologic fractures secondary to tuberculosis bone disease. The etiologies of pathologic fractures in children are dominated by benign causes and congenital bone anomalies. A.E. Arfaja, in their series, found one case, i.e. 3.3% of pathological diaphyseal fracture of the femur following tuberculous osteomyelitis [3].

J.L. Rakotoson *et al*, in a study carried out in Fianarantsoa reported a TOA rate of 0.2% [4]. According to the literature, the most common form of osteoarticular tuberculosis mainly affects the spine (50-70%). In the extravertebral forms, localization in the knee and tibia accounts for 10% [5].

Most authors consider that osteoarticular tuberculosis is always secondary to a primary or reactivated infectious focus. However, isolated cases have been reported in apparently healthy patients with no personal or family history of tuberculosis [6]. TOA is encountered in any age group. However, it should be noted that bone tuberculosis in children has started to be important in the developed [7].

The clinical picture includes local symptoms such as pain, tenderness and limitation of movements, with some particularities depending on the topography, sometimes accompanying specific systemic symptoms of tuberculosis and other specific clinical signs such as cold abscesses and fistulas [8].

During its evolution, TOA presents certain general characteristics: it is chronic, slowly progressive and destructive, often leading to walking difficulties and functional impotence [9].

According to WHO guidelines, the diagnosis of extrapulmonary tuberculosis should be based on at least one specimen with confirmation of mycobacterium tuberculosis or histological evidence or strong clinical arguments consistent with active extrapulmonary tuberculosis [10]. Hematologic parameters such as accelerated VSH and hyperlymphocytosis may aid in the diagnosis, but are neither specific nor reliable [11]. Pathological examination of bone biopsy is necessary for diagnostic confirmation of tuberculous osteomyelitis [12]. Being a tuberculosis endemic country, we perform a systematic anatomopathological examination in front of any chronic abscessed or suppurated osteoarticular infection.

X-rays give a good diagnostic clue, but they may mimic pyogenic osteomyelitis, Brodie's abscess, tumors, or granulomatous lesions. MRI, when available, is a valuable adjunct to standard radiographs [13]. However, often these nonspecific clinical and commonplace radiographic features of TOA are the main reasons for their delayed diagnosis [14].

The anti-tuberculosis treatment regimen and duration vary according to well-established protocols in different countries. Most authors have recommended a minimum treatment duration of 12 months, but more recent studies have shown a good result with a short regimen of 6 months [15].

In our case, we had a good clinical course following a treatment regimen in Madagascar of eight months with a combination of four antituberculosis drugs (rifampicin, isoniazid, ethambutol and pyrazinamide) for two months followed by two antituberculosis drugs (isoniazid and thiacetazone) for six months.

Xing Guo *et al*. State in their study that the early use of anti-tuberculosis drugs combined with surgery is the key to the treatment of certain localizations of TOA [16]. According to some authors, the place of surgery remains limited to diagnostic biopsy, evacuation of abscesses or removal of chronic fistulas resistant to specific treatment [17]. Chattopadhyay P *et al* emphasize that curettage of the osteomyelitis lesion offers a chance of early cure [18]. Several works have already emphasized the importance of the fibula in the reconstruction of tibial bone loss. Our observation would have been an indication of this, with the possibility of restoring the fibular splint, given the absence of shortening and axis defect [19]. O. Traore *et al*, in their study on chronic osteomyelitis, also reported the need for total tibial diaphyseal resection treated with a non-vascular peroneal graft in a patient with pandiaphyseitis with a large sequestrum >5 cm [20]. In our case, the patient was lost to follow-up after healing of the operative wound and we were unable to initiate reconstructive surgery for improved limb function.

4. Conclusion

Due to the rarity of the discovery of osteoarticular tuberculosis at the stage of extreme and late complication, our case report does not show long-term results, especially the functional future of the limb. This pathology must be considered in the differential diagnosis of any unexplained lesion of the limbs of chronic evolution in endemic regions. Such a lesion poses a number of management problems, both in terms of bone loss and tuberculosis infection.

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