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Case Report

Primary Chronic Sclerosant Osteomyelitis in Bilateral Tibia Shaft: Case Report

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Abstract

Chronic Sclerosant Osteomyelitis still attracts attention as a disease of unknown etiology. It is more frequently seen in children and adolescents, and is usually self-limiting. It is characterized by occasional recurrence and painful bone lesions settling symmetrically and it constitutes 2-5% of all osteomyelitis. For this disease, that mostly the adult female population is more affected, female/male ratio is 5/1.

It is most frequently seen between ages 4 and 14. There are many different clinical forms of the disease as well as the known classical osteomyelitis.

Key Words

Osteomyelitis; Sclerosis; Surgical treatment; Tibia

Introduction

The sclerosant form of osteomyelitis, defined by Garre in 1893 for the first time, that causes thickening and expansion of the bone, but no suppuration, sequestration or fistula formation is observed, was named by Hardmeier and colleagues as "Primary Chronic Sclerosant Osteomyelitis" [1]. The Primary Chronic Sclerosant Osteomyelitis is a rare and difficultly diagnosed disease [2]. It progresses with insidious onset of pain and recurrent symptoms.

Distention and sclerosis are present in radiologically affected bone. Histologically, the new bone formation with low-grade chronic osteomyelitis appearance is determined [3]. Although the treatment of this disease, that affects the children and young adults, is still controversial we aim to present our clinic's diagnostic and treatment results of the bilateral tibia shaft case.

Case Report

34-year-old male patient admitted with the complaint about pain in the middle part of both tibias. There was no complaint about rash or leakage on both lower extremities but he was describing a pain that restricted his activities. It was detected that the complaints began all of a sudden 5 years ago, he was biopsied at the both tibia shafts, and that was evaluated as the patient had osteomyelitis and he had oral antibiotic therapy. In physical examination, no localized rash or serous discharge was observed instead of dryness on the area fitting bilateral both tibia shafts. In two-way tibia graphs of the patient, with normal neurovascular assessment and no knee or ankle movement limitation, cortical thickening and erratic sclerosis was observed in bilateral tibia shafts (Figure 1a-c). In hematological investigations, erythrocyte sedimentation rate, C-reactive protein and white blood cell count were found to be normal (Figure 2). After scintigraphy revealed the involvement of diaphyseal areas (Figure 3). Magnetic resonance imaging revealed similar findings. Medullary cavity was completely closed bilaterally (Figure 4a,b). Reamerisation was performed on the patient with bilateral medullar reamers under spinal anesthesia. After the reamerisation performed

on left tibia, the procedure was terminated with prophylactic plaque screw osteosynthesis implementation from the medial because of a partial fracture piece in posterior cortex. In the histopathological examination of the material obtained from the lesion, chronic granulation tissue was observed.. After the operation patient was mobilized with partial load only for 4 weeks. The patient returned to his daily activities and work painlessly at the 8th week (Figure 5a,b).

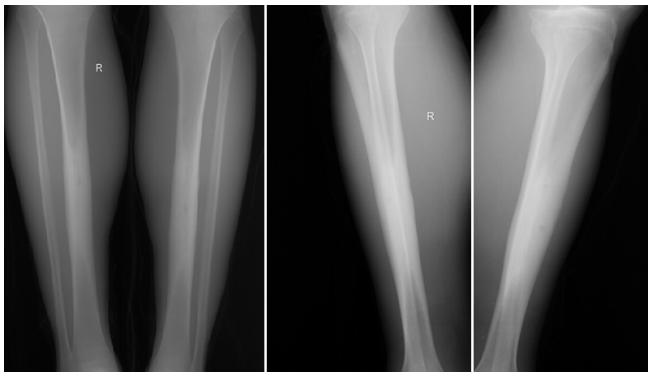


Figure 1a-c: Both AP and lateral radiographs of the tibia; areas shown in the diaphyseal sclerosis.

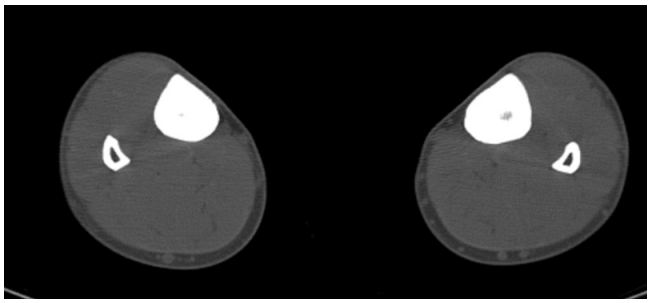
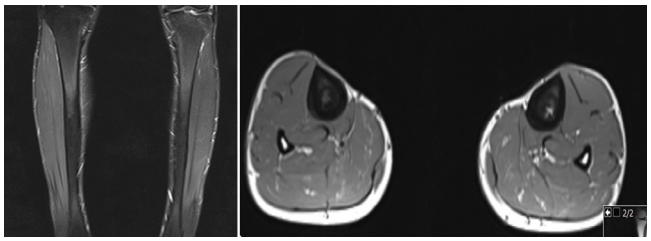


Figure 2: Sagittal CT, tibial diaphyseal medullary fully closed.



Figures 3a,b: MRI scans of patient

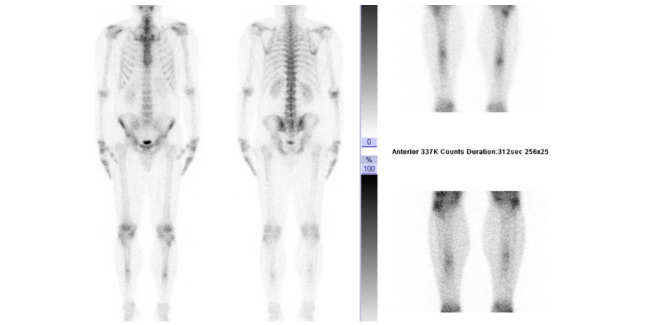


Figure 4a,b: Patient scintigraphy images.

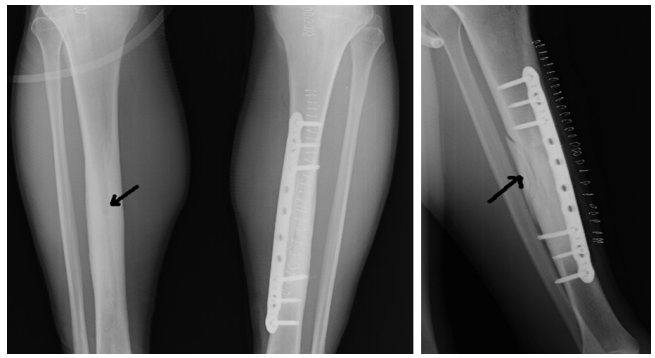


Figure 5a,b: After the operation images, right tibia, the medullary canal open medullary after reamirizasyo cavity (arrow), the left tibia in the fissure lines formed during the reamerizas (arrow) applied to the bridge plate screw application is seen.

Discussion

Chronic Sclerosant Osteomyelitis (CSO), is a disease manifesting itself with fistule formation in bone, thickening, stiffening and expansion without dead bone or apses, and is a disease with exactly unknown cause. This situation is thought to be caused by a microorganism with low infection level. Main complaint is pain but local findings of classical osteomyelitis such as fistule or apses are not seen in this type of osteomyelitis. Expansion and generalized sclerosis in bones stand out in plane graphies. As a laboratory finding, Erythrocyte Sedimentation Rate (ESR) is generally slightly increased in blood. In biopsies low level, non-specific osteomyelitis findings are seen while in cultures microorganism growth is mostly unsuccessful [2,3]. However, in some publications it has been reported that anaerobic *Propionibacterium* or *Actinomyces* species were managed to be grown [4].

The majority of publications in the literature mention chronic sclerosant osteomyelitis developing from maxillary and/or mandibular center after oral or dental infection [5]. However there are case reports of metacarpal, fibular, and even bilateral clavicles involvement [6-8]. For the first time in 1987, synovitis, acne, pustules formation and hyperostosis associated "Sapho syndrome" his syndrome was correlated to Garre's sclerosant osteomyelitis [9]. Collar and Isaacson stated the clinical course of the disease as insidious onset of pain in children and distension in bone [10].

In differential diagnosis pathologies such as osteoid osteoma, osteoblastoma, fibrous dysplasia, and Paget's disease should be kept in mind [2,8,9]. Conservative surgical procedures are reported for the treatment of chronic sclerosant osteomyelitis. In the conservative treatment, long term antibiotic therapy, calcitonin preparations have been tried and temporary reduction in pain was achieved [11,12,14]. Reaming of the medullary canal, opening window in the bone for biopsy, complete or partial resection of the infected bone are also surgical procedures applied for permanent treatment [2,3,6,10,13].

Wright and colleagues reported in a case with femur involvement that after using oral risedronate as the antibiotic therapy didn't remiss the complaints, the complaints ended at the end of two weeks and at the end of six months of use bone turnover markers returned to normal in blood, but there was no observable improvement in plane graphy and bone scintigraphy [14]. Among surgical treatment options, partial or complete resection of the diseased section of bone may provide a permanent healing. Intramedullary reaming, as another method, is one of the surgical procedures performed in long bone chronic osteomyelitis. Pape and colleagues reported that improvement was achieved by increasing local blood circulation in this method [13]. Walenkamp, on the other hand, recommends to open a 1-2cm bone window on medial malleolus in tibia or medial condyle in femur to prevent the accumulation risedronate of debris in the marrow during the reaming [15]. Bettin and colleagues reported that they obtained successful outcomes in five year follow up of a case involving femur that they treated by opening a bone window and reaming through marrow [10]. Radiological and clinical findings, matching sclerosant osteomyelitis like cases mentioned in the literature, were present in our patient.

There was no evidence for infection in physical examination, or laboratory and radiological studies. Sclerotic lesions observed in radiological studies and pain requires surgical treatment of the case. Because of chronic inflammation and sclerosis observed in histological research and negative results in bacteriological cultures, the case received a diagnosis of chronic sclerosant osteomyelitis. In the follow ups of the patient, the pain was detected to be over.

As a result, in patients with the continuing and persistent bone pain and radiological findings of primary sclerosant, eyes should be open for chronic sclerosant osteomyelitis as well as other bone pathologies.

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