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Original Article

A Case of Sclerosing Osteomyelitis of Bilateral Tibia Successively Treated with Open Window Decompression Loaded Antibiotic Slow Release Rods

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Abstract:
The purpose of this study was to report a rare case of sclerosing osteomyelitis (Garré osteomyelitis). The patient was a 48-year-old female who presented 3 months ago with pain in the right lower leg with limitation of movement and significant tibial trunk tenderness, but without fever. The patient was in good general condition, with no localised discharge, and imaging showed disturbed trabecular bone in the tibial stem with periosteal reaction, which was consistent with chronic osteomyelitis of low virulence and slow progression. A curettage of the right tibial stem was performed and secretions were released but resulted in negative cultures. Biopsy was suggestive of infection and inflammation. The etiology was not clear: low virulence bacterial infection was suspected (anaerobes were likely), but cultures were negative. Chronic course maintained by low virulence infection or even after treatment.

Keywords: chronic sclerosing osteomyelitis, tibial osteomyelitis, treatment, bone infection, Garré's disease

Introduction

Sclerosing osteomyelitis, also known as 'Garré's osteomyelitis', is a chronic osteomyelitis caused by a low virulence infection, with a predominantly sclerotic bone [1]. Bacterial cultures are usually negative in the lesions, and the disease is usually caused by increased intraosseous tension in the bone due to osteomalacia [2]. Usually, long tubular bones such as tibia and ulna are the common sites of involvement in this disease, with few systemic symptoms, but often with a history of recurrent episodes [3]. On examination, a previous surgical scar of about 8 cm in length was seen on the anteromedial aspect of the left calf, dry and without exudation, the right calf had no obvious deformity, no obvious local erythema, local skin temperature was normal, and there were multiple compression pains over the whole segment without obvious percussion pains. In this paper, we report a rare case of sclerosing osteomyelitis occurring bilaterally in the tibia, which highlights the diagnostic challenges of Garre osteomyelitis.

Case Data

The patient, a 48-year-old married woman, was admitted to the hospital with 'pain in the right lower leg for 3 months, aggravated for 1 month'. The patient's pain was aggravated by local pain during activities, and the pain was persistent, dull, and vague. After taking oral anti-inflammatory and analgesic medication, the pain was reduced and tolerated, but the pain in the right calf aggravated a month ago without any obvious cause, especially at night. She complained that she had a history of surgery for osteomyelitis of the

left tibia, and there was a surgical scar of about 8cm long on the anterior medial side of the left calf. Her body temperature was normal, and there was no redness or secretion from the right calf. Bacterial cultures were negative, adding to the difficulty of the diagnosis.

Routine laboratory tests were normal (interleukin 6 <1.5 pg/ml; calcitoninogen 0.030 ng/mL; erythrocyte sedimentation rate 43.0 mm/h; fibrinogen 2.49 g/L: C-reactive protein <3.01 mg/L.) X-ray plain films and computed tomography (CT) scans demonstrated thickening of the cortex of the middle tibia bilaterally, with lamellar flocculent and lattice hyperdense shadows visible in the medullary cavity. The borders were indistinct, representing osteosclerosis and narrowing of the medullary cavity in the segment where the lesion was located. The left mid-tibia had localised bony defects and communicated with the medullary cavity, forming a sinus tract (Figure 1).

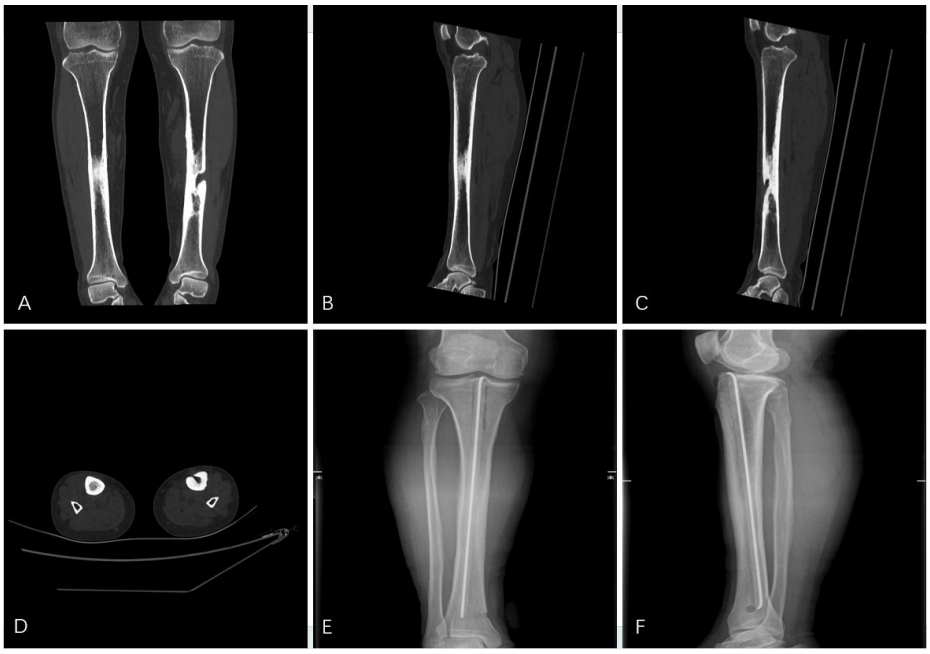

Figure 1:A: CT 3D reconstruction in coronal position; B: CT 3D reconstruction in sagittal position (R); C: CT 3D reconstruction in sagittal position (L) D: CT 3D reconstruction in axial position; E: X-ray postoperative orthopanthesis; F: X-ray postoperative lateral position

There were subcutaneous soft tissue swelling and exudative manifestations on the anterior side of both calves, as well as bilateral calf muscle atrophy. It was consistent with chronic osteomyelitis with low toxicity and slow progression. Microbiological examination was performed under aerobic and anaerobic conditions, and fragments of surgical specimens were collected, none of which revealed microorganisms (Figure 2).

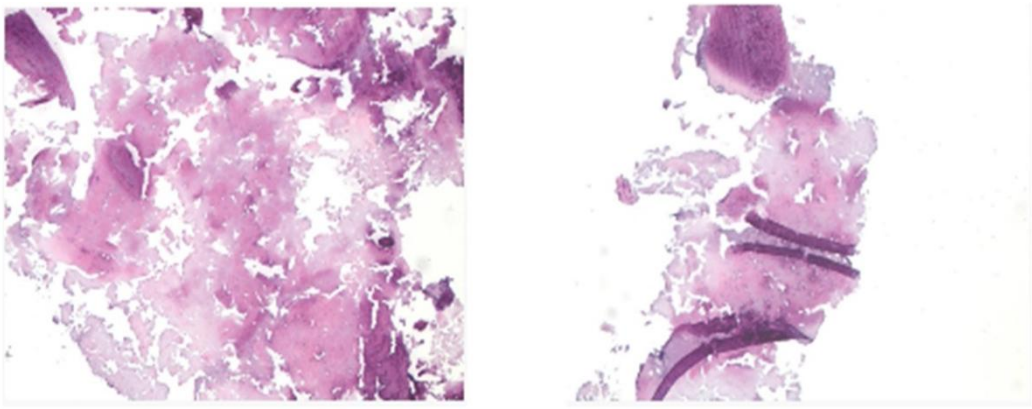

Figure 2 Biopsy staining of pathological tissue

Propionibacterium acnes cultures were negative. The diagnosis was ‘chronic osteomyelitis (sclerosing).

Preoperative treatment: bacterial culture was performed, and the most sensitive antibiotic was selected based on the results of the culture to alleviate the patient's pain and discomfort; the site of the lesion was determined based on X-rays, CT,, and MRI to determine the area of surgical lesion removal. Surgical treatment included right tibial bone marrow lesion debridement, right tibial external fixation, cement filling, and tibial intramedullary nail internal fixation. Localised sclerotic bone was removed using hard expansion with an intramedullary pin, and a small localised opening was made in the medial aspect of the distal tibia after removal. Subsequently, a large amount of sclerotic bone was expelled from the open window. It was removed for bacterial culture under aerobic and anaerobic conditions, and fragments of the surgical specimen were collected and sent for pathological examination. The culture was negative for Propionibacterium acnes. The diagnosis was ‘predominantly chronic osteomyelitis’. However, the culture result was negative.Under the fluoroscopy of C-arm image intensifier, the medullary expansion drill was repeatedly taken to clean the bone of sclerotic diseased bone segments in the medullary cavity until the width of the medullary cavity was restored to the desired width, and a large amount of saline and dilute iodine volts were repeatedly rinsed, and counterpart drainage was carried out through the distal tibia open window hole. Vancomycin-containing PMMA wrapped around Φ2.5mmx30mm elastic intramedullary needles was used to prepare antibiotic slow-release carrier rods, which were solidified, cooled and dried, and then placed in the tibial bone marrow cavity (Figure 3).

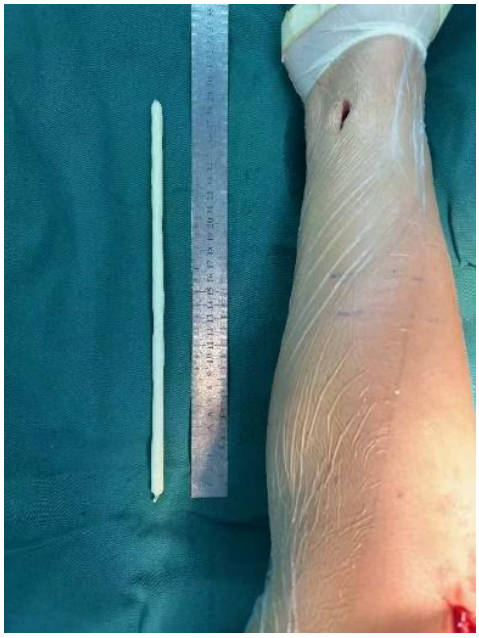

Figure 3 Intramedullary needle slow-release rods

This method delivers an effective concentration of antibiotics to the local infected lesions in a targeted manner, thereby increasing the bioavailability of the drug, improving the efficacy of the treatment while reducing its toxic side effects, and providing structural support for bone regeneration. The tissues obtained intraoperatively were sent for pathological biopsy analysis, which showed the presence of infection and chronic inflammation.

On hospitalisation, Natriuretic Heparin Calcium Injection (4+7) B 4100iu subcutaneously 1/day; Diclofenac Sodium Extended Release Tablets A (base) 100mg orally 1/day; Mazolin Tablets (Horse Chestnut Seed Extract) B 300mg orally 2/day; Dextrose Sodium Chloride Injection (soft) A Nong (base) 500ml IV 1/day. After discharge cefuroxime tablets 0.25g, oral 2/day (oral 3 weeks). Postoperative follow-up for two months, the patient was satisfied.

Discussion

Sclerosing osteomyelitis is mostly caused by long-term chronic infection with blood-borne low-toxicity bacteria, and clinically it mostly manifests local soreness and pain, exacerbated at night, aggravated by exertion, and often recurring, and the symptoms can be alleviated by using antibiotics [1,4]. There is no obvious redness or heat, and the bone shaft can be thickened by palpation after repeated episodes. The diagnosis is supported by imaging (X-ray, CT or MRI) [5], which shows thickening of the bone cortex, narrowing of the medullary cavity and sclerotic bone shadows in the corresponding lesion area. The etiology of this disease remains incompletely elucidated. Bacterial infection is suspected, but cultures are usually negative. The differential diagnosis of medullary stenosis and osteocortical thickening of the long bone diaphysis is most often seen in the following disorders: chronic sclerosing osteomyelitis, intramedullary osteosclerosis, and progressive diaphragmatic dysplasia. Despite the similarity of these diseases from an imaging point of view, with new bone formation in the periosteum and endosteum [6], there are still a number of clinical differences. Intramedullary osteosclerosis usually involves one or both mid tibiae, and the femur and fibula can also be involved [7]. The disease is most common in adult females. It is not associated with infection, trauma or genetic factors. It is usually found on imaging in patients with chronic pain in the leg with physical activity. Increased sclerosis of the medullary bone is seen on imaging. Progressive diaphyseal dysplasia is an autosomal dominant disorder caused by mutations in transforming growth factor-β1 [7]. It usually involves the long bones and skull, with the lower limbs being more severely affected than the upper limbs. However, clinically, it tends to present with limb pain, muscle weakness, and easy fatigue. Thorough diagnosis and comprehensive evaluation are necessary to distinguish sclerosing osteomyelitis from other diseases.

Chronic sclerosing osteomyelitis has a low mortality rate but a high complication rate.Gowda et al[6] found the sensitivity of X-ray and MRI imaging to be 51% and 97% with a specificity of 94% and 96%. In our case, the diagnosis was made on the basis of combination of imaging and clinical and exclusion of other common differential diagnoses. The high rate of diagnosis of osteomyelitis by X-ray and CT at our institution is due to the long period of specialised imaging training we have undergone and our greater experience in imaging osteomyelitis. Our patients have experienced insidious, recurrent pain in the tibia as a common presentation of sclerosing osteomyelitis. Elevated inflammatory mediators, negative tissue cultures, laboratory tests and imaging findings are common features of sclerosing osteomyelitis in our patients. For chronic osteomyelitis, urgent combination of antibiotics is used for combination therapy. The ineffectiveness of antibiotics alone, independent of the way they are administered, is due to the fact that the ‘bone segregation’ in chronically ill patients is made up of necrotic bone fragments, and therefore does not provide a sufficient blood supply for antibiotics to enter the infected tissue.

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Author Contribution: Kai Zhang performed the data analysis and wrote the manuscript. Jia Chang and Zhiqiang Lin planned the study, contributed to the interpretation of results, and contributed to revising the manuscript. Jiahuan Li and Shenghu Zhou generated the data utilized in this analysis. Kai Zhang, Jia Chang , Zhiqiang Lin ,Jiahuan Li , Shenghu Zhou contributed to revising the manuscript. All authors read and approved of the final manuscript.

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Conflicts of Interest Statement: There is no conflict of interest regarding the publication of this paper.

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