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Case Report

Chronic recurrent multifocal osteomyelitis: a case report☆

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ABSTRACT

Chronic recurrent multifocal osteomyelitis (CRMO) is a rare idiopathic inflammatory disease that affects mainly children and young adults. The clinical signs and symptoms are nonspecific, hindering and delaying diagnosis. Radiological and histopathological tests are essential for its definition. A case of CRMO is reported, demonstrating the importance of clinical, laboratory, and radiological data for diagnosis.

Osteomielite crônica multifocal recorrente: relato de caso

Palavras-chave:
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RESUMO

Osteomielite crônica multifocal recorrente (OCMR) é uma doença inflamatória idiopática rara que acomete principalmente crianças e adultos jovens. Os sintomas e sinais clínicos são inespecíficos, dificultam e retardam o diagnóstico. Os exames radiológicos e histopatológicos são indispensáveis para sua definição. Neste relato, os autores apresentam um caso de OCMR que demonstra a importância dos dados clínicos, laboratoriais e radiológicos para o diagnóstico.

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Introduction

Chronic recurrent multifocal osteomyelitis (CRMO) is a rare idiopathic inflammatory disease, the diagnosis of which is made by exclusion, affecting mainly children and young adults. The early identification of this pathology avoids unnecessary examinations and prolonged antibiotic therapy. The clinical signs and symptoms of the disease are nonspecific and hinder the diagnosis based on clinical presentation only. Thus, radiological and histopathological exams are indispensable for its definition.

The objective of this report is, therefore, to present a case of CRMO, and to demonstrate the diagnostic importance of clinical, laboratory and, mainly, imaging tests.

Case report

A 44-year-old female patient, who suffered a car accident 12 years before, with no fractures; but had a laceration on the right thigh. After four years, she presented with continuous pain in the left leg, with nocturnal worsening. Previously healthy, she denied a family history of musculoskeletal conditions, drug reactions, previous surgical history and febrile conditions after the accident.

On September 2005, she sought medical care and underwent computed tomography (CT) (Fig. 1), which showed cortical thickening in the left tibial shaft with areas of bone resorption. She underwent surgical treatment, progressing with tibial fracture three days after the procedure, the reason of which, according to the patient, was the removal of a large bone fragment during the surgical procedure. Treatment followed with cast immobilization and use of non-steroidal anti-inflammatory drugs (NSAIDs).

On October 2013, the patient sought medical care because had pain in the right leg for three months, similar to the previous one in the left leg. Imaging and histopathological exams were performed. Radiographs of the right leg showed diffuse cortical and medullary sclerosis at the tibial shaft. These changes were confirmed in a CT, which also revealed a minimal area of bone neoformation with cortical thickening at the proximal end of the fibula. Surgical biopsy showed a nonseptic lesion, and the medullary canal was recanalized. MDP-Tc^{99m} bone scintigraphy demonstrated hypercaptation of the radiopharmaceutical agent in the middle third of the right leg, without denoting bone involvement due to an infectious process or indicating any other asymptomatic focus.

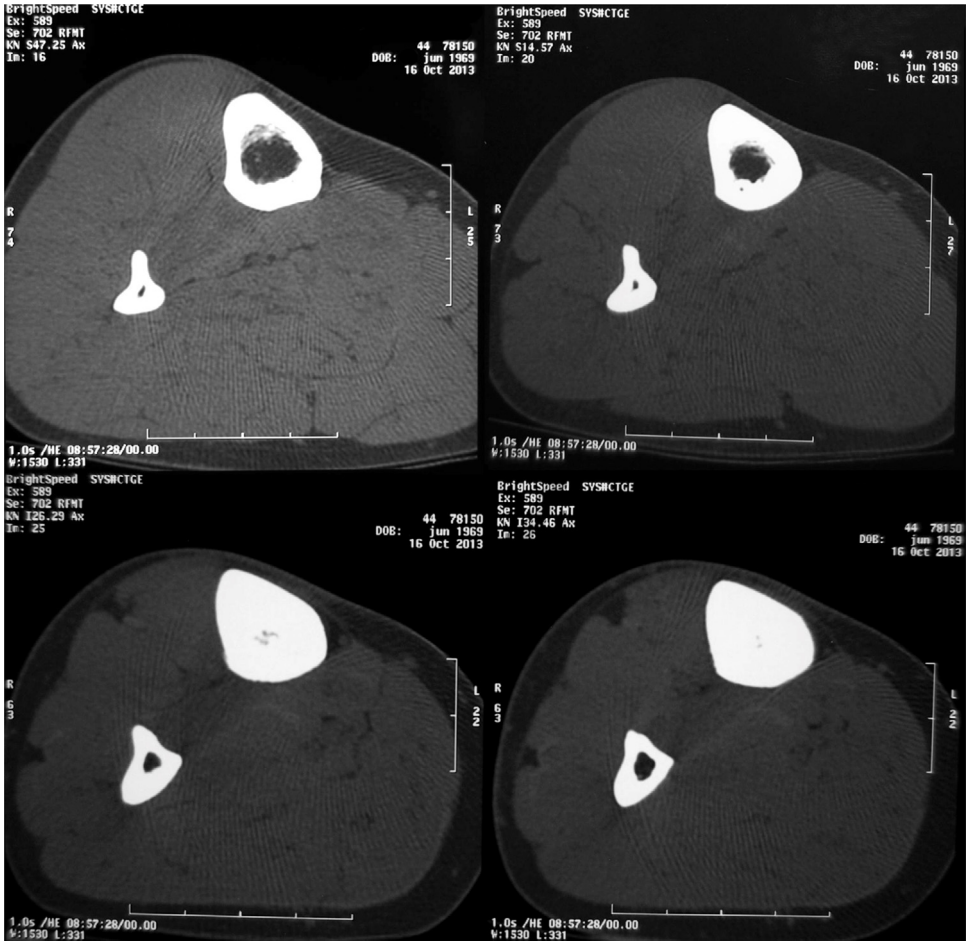

Fig. 1 – Axial computed tomography images of the middle third of the leg bones shaft show periosteal and endosteal bone neoformation that lead to a medullary canal narrowing.

The analysis of the exams allowed the diagnosis of CRMO. Thus, the patient received NSAID therapy, and has not presented any recurrence of pain.

Discussion

CRMO is an idiopathic inflammatory bone disorder seen mainly in children and adolescents[1]; however, it has already been described in older patients up to 55 years.[2,3] This is a rare variant of osteomyelitis, corresponding to 2–5% of the cases.[4] Patients usually present with insidious onset of heat, pain, and soft-tissue edema, restricted to one or more bones.[5] The lower limbs are the most affected,[5] with metaphyses or equivalent sites being the most common locations.[6]

Its diagnosis is made by exclusion, and the main causes to be excluded are neoplasms and infections.[7,8] It is based on the following criteria: (a) bone lesions with images suggesting subacute or chronic osteomyelitis; (b) unusual location when compared to infectious, multifocal osteomyelitis; (c) absence of abscess, fistula or sequestra; (d) absence of an etiological agent; (e) nonspecific histopathological and laboratory tests consistent with subacute or chronic osteomyelitis; (f) typical episodes of prolonged and recurrent episodes of pain, and (g) occasional skin manifestations, such as palmoplantar pustulosis, acne, psoriasis vulgaris and pyoderma gangrenosum.[9,10] The patient presented with all the above criteria, except for cutaneous manifestations.

Image exams reveal nonspecific alterations that require histopathological exams to better define the disease. These tests rule out the possibility of neoplastic or infectious lesions, while the images are useful for assessing the extent of the disease, reveal lytic areas and periosteal reaction, which evolve to hyperostosis and sclerosis, exactly what the patient's images showed (Fig. 1).

Treatment involves specially NSAIDs that aim at symptomatic relief. Antibiotics are considered ineffective. The case patient was treated with NSAIDs and had good progress, with no recurrence of pain.

CRMO progression is unpredictable. Although most cases resolve spontaneously in months to years, there are reports of symptomatic patients for up to 25 years after diagnosis.

Knowledge of the appearance of CRMO lesions and their typical changes is important for an early diagnosis, leading to a reduction in the number and necessity of biopsies, surgeries and antibiotic therapy.

Conflicts of interest

The authors declare no conflicts of interest.

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