Chronic recurrent multifocal osteomyelitis with isolated spinal involvement

Furtado C[1], Amaral R[2], Jacinto T[3], Amaral P[4]

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Chronic recurrent multifocal osteomyelitis (CRMO), also named nonbacterial osteomyelitis (NBO) and being a form of presentation of synovitis, acne, pustulosis, hyperostosis, osteitis syndrome (SAPHO), is an increasingly recognized auto-inflammatory non-bacterial disease, of unknown etiology, which occurs predominantly in children and adolescents[1]. The clinical course is characterized by prolonged, selflimiting recurring episodes. Its differential diagnosis includes infection and malignancy.

Spinal involvement, although less common, has been reported. In a review of 35 cases, lesions of the vertebral bodies accounted for 3%[2]. The thoracic spine is the most affected, followed by the lumbar vertebrae[3]. It may present as spondylodiscitis-like lesions or as predominantly lytic lesions, resulting in the collapse of the affected vertebral bodies and progressive kyphosis[4].

Magnetic resonance imaging (MRI) is the most sensitive and specific imaging modality in assessing local extent, evolution and disease activity[4].

The authors report the image study of a case of spinal CRMO in a 13-year-old female patient presented to the emergency department in April 2014 with insidious, recurrent back pain on the upper thoracic spine for the last 4 months. The pain rated between 6 and 8 in 10 in intensity and had an inflammatory rhythm, worsening at night with morning stiffness, being refractory to analgesics including non-steroidal anti-inflammatory drugs (NSAIDs). She did not recall a precipitating event, particularly trauma, reported no fever, lethargy or weakness. There was no relevant personal or family history.

On examination, painful palpation of the thoracic spinal processes, paraspinal muscles contractures and kyphotic accentuation were noted. Tender or swollen peripheral joints, cutaneous stigma or neurological deficits were absent. Her complete blood count and serum biochemistry were within normal range (NR), erythrocyte sedimentation rate 28mm/h (NR <20 mm) and C-reactive protein 2.86 mg/dl (NR <0.3 mg/dl).

Spinal computed tomography (CT) evidenced lytic destruction of 3th and 4th, and collapse of 5th thoracic vertebral bodies, originating kyphotic deformation (Figure 1). MRI showed that these lesions were hypointense on T1-weighted and hyperintense on T2 (Figure 2). Whole-body scan confirmed radioactivity pooling in 3th to 5th thoracic vertebrae.

She underwent several empirical intravenous antibiotic treatments, including cefotaxime, vancomycin (suspended for toxidermia), tazobactam, teicoplanin (discontinued by secondary agranulocytosis) and a therapeutic trial with tuberculostatics, without clinical improvement. Microbial laboratory assays of blood and bone cultures were sterile for bacteria, including tuberculosis. A CT-guided biopsy of 4th and 5th thoracic vertebral bodies showed fibrotic marrow and non-specific chronic inflammatory lymphoplasmocytic infil-

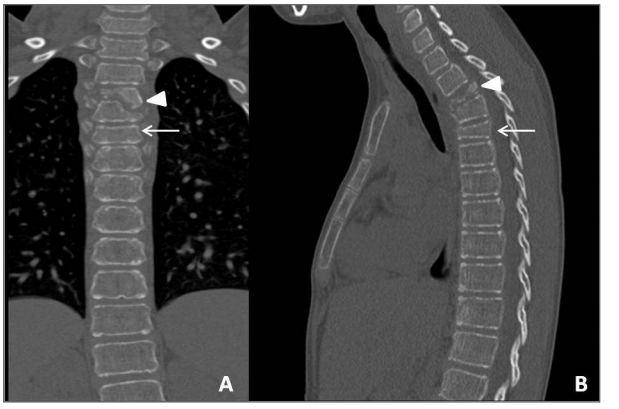

FIGURE 1. Spinal computed tomography (CT) evidenced lytic destruction of 3th and 4th, and collapse of 5th thoracic vertebral bodies, originating kyphotic deformation

1. Department of Rheumatology, Hospital do Divino Espírito Santo – EPE, Ponta Delgada, Azores
2. Department of Radiology, Hospital do Divino Espírito Santo – EPE, Ponta Delgada, Azores
3. Department of Pediatrics, Hospital do Divino Espírito Santo – EPE, Ponta Delgada, Azores
4. Department of Orthopedic Surgery, Hospital do Divino Espírito Santo – EPE, Ponta Delgada, Azores

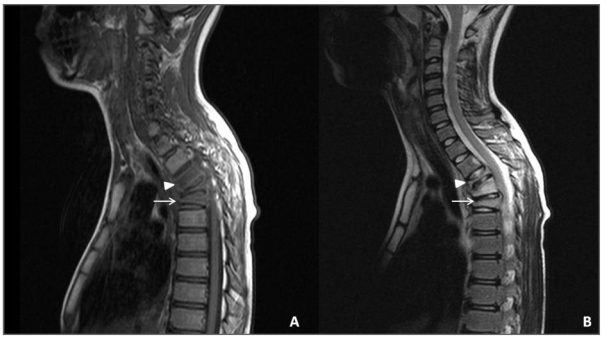

FIGURE 2. MRI showed that these lesions were hypointense on T1-weighted and hyperintense on T2

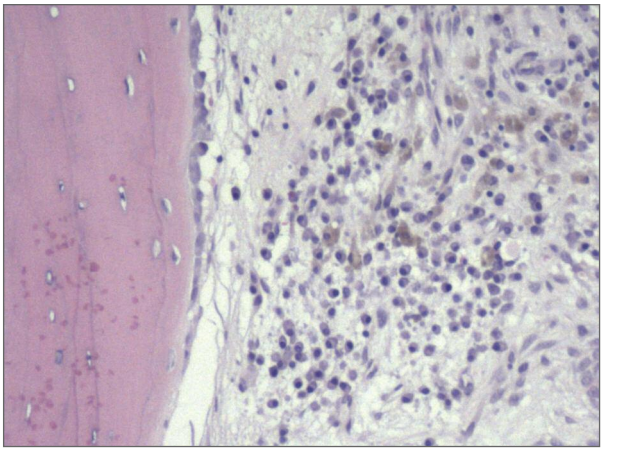

FIGURE 3. CT-guided biopsy of 4th and 5th thoracic vertebral bodies showed fibrotic marrow and non-specific chronic inflammatory lymphoplasmocytic infiltrate

trate (Figure 3). Clinical findings, diagnostic imaging, negative histological and microbiological examinations, and no response to antibiotics led to the diagnosis of CRMO in September 2014. Intravenous pamidronate 1mg/kg was administered for 3 days with rapid pain relief, with no recurrence of symptoms after 12 months of follow-up.

The present case, illustrating the challenge of timely diagnosis, highlights the importance of considering CRMO in adolescents with presumed infective vertebral osteomyelitis, especially if refractory to antibiotics, in order to avoid potentially unnecessary or invasive investigations or treatments.

Pamidronate is recommended in refractory CRMO with extended spinal involvement, with favorable clinical outcome[5].

CORRESPONDENCE TO
Carolina Furtado
Department of Rheumatology, Hospital do Divino Espiríto Santo – EPE
Ponta Delgada, Azores
Portugal
E-mail: carolfurtado633@hotmail.com

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