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Case report

Non-bacterial osteitis: Chronic Recurrent Multifocal Osteomyelitis or pediatric SAPHO?

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Abstract

Chronic Recurrent Multifocal Osteomyelitis (CRMO) and SAPHO syndrome represent the group of autoinflammatory bone disease responsible for recurrent non-bacterial osteitis (NBO). both are considered as defects of innate immunity. The most common clinical presentation is recurrent episodes of bone pain with or without fever.The clinical and imaging features are non-specific.This usually leads to late and confusing diagnosis.
We hereby report a case of CRMO in a 12-year-old patient. The aim is to highlight the confusing overlap of clinical features between CRMO and SAPHO syndromes.

Keywords:
multifocal osteomyelitis, non-bacterial osteitis, SAPHO, bone pain.

Observation

A 12-year-old female patient with a history of recurrent metatarsalgia in the past year presented with right thigh pain of three weeks duration. The pain was related to exertion in the beginning and became permanent later. A fever of three days duration preceded the onset of the pain. The examination revealed a small painful swelling in the right thigh. Joint and skin examinations were normal.
X-rays of right femur showed multilamellar inflammatory reaction of the shaft (Figure1). Magnetic resonance imaging (MRI) showed heterogeneous mass withT1 hyposignal and T2 hypersignal which was diffusely infiltrating the muscles (Figure 2).
The abdominal ultrasound and chest CT scan were normal. Laboratory exams revealed high CRP and ESR. The hemoglobin rate was normal. The X-rays of both feet showed multilamellar reaction of the 2nd and 4th left metatarsal, bones and also of the right 2nd metatarsal bone(Figure 3). MRI of the whole body showed multiple lesions in the proximal and distal metaphysis of the left humerus, right humeral shaft, left acetabulum, pubic rami, the right side of the sacrum and the right femoral neck (Figure 4). Bone biopsies were performed from the femoral mass to rule out Ewing's sarcoma.
Histopathology examination showed non-specific chronic inflammatory cell reaction and fibrosis. There was no sign of malignancy and the culture was sterile. Majeed syndrome was considered as differential but not retained due to the absence of anemia and similar familial history.
The patient was treated by non-steroidal anti-inflammatory drugs and showed good response. At 2-months follow-up there were satisfactory biological and radiological improvements.

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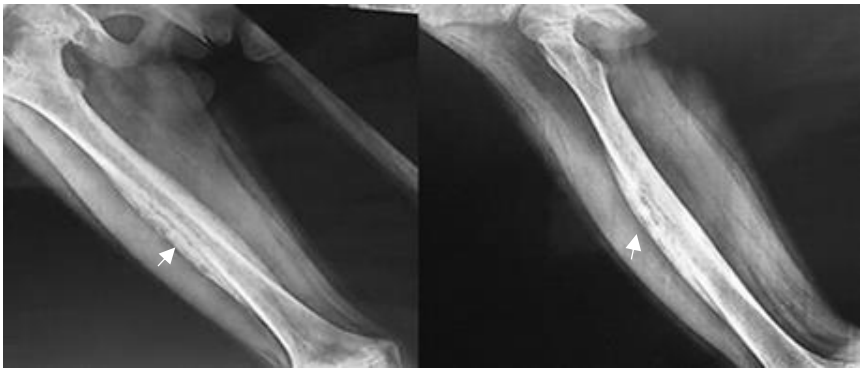

Figure1

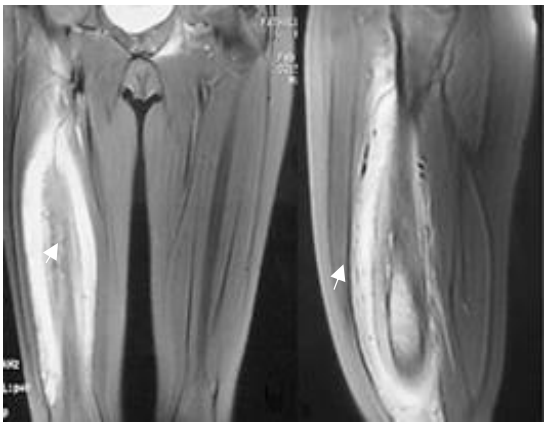

Figure 2

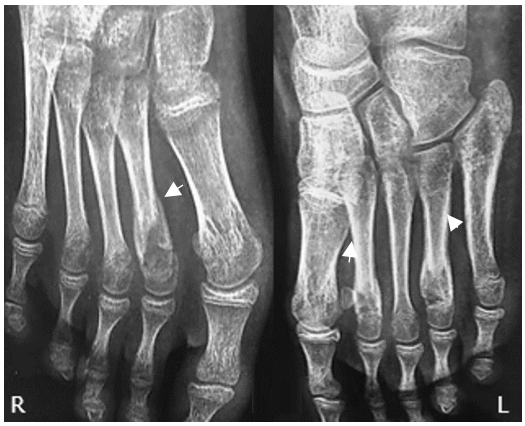

Figure 3

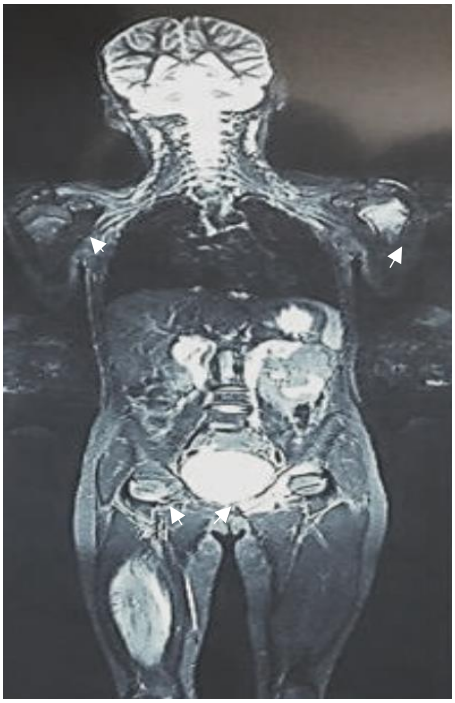

Figure 4

Figure 1: X-ray of the femur showing multilamellar reaction

Figure 2: MRI aspect of infiltrating right thigh mass

Figure 3: X-rays of both feet showing multiple metatarsal inflammatory processes.

Figure 4: cartography of the different CRMO sites.

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Discussion

Typically, CRMO presents as recurrent bone pain with or without low grade fever [1]. Episodic exacerbations and remissions are characteristic. The bone pain has usually insidious onset. Objective signs of arthritis may involve one or more joints [2]. The skin findings include psoriasis, palmoplantar pustulosis, pyoderma gangrenosum, and cystic scarring acne. The average age of onset is 10 years (4-55years). Bone involvement in CRMO has usually an asymmetric distribution with predilection for long bones of lower extremities. This can mimic infectious osteomyelitis or malignant tumors in children. During exacerbations, high CRP and ESR are found in more than half patients. However, it is not usual to find objective anemia and Rheumatoid factor is negative. Only ten percent of patients are positive for HLA-B27[3]. Comorbid conditions found in CRMO may include spondyloarthropathy, psoriatic arthritis as well as Inflammatory Bowel Disease (Crohn's disease or ulcerative colitis). Comorbid conditions can be absent in the onset of the disease and appear after 1 to 5 years of evolution[4,5].

SAPHO is also characterized by episodic recurrent bone pain due to non-bacterial osteitis [6]. The most commonly affected bones are located in the chest wall [7]. The osteitis can be unifocal or multifocal and asymmetric. One of the distinguishing SAPHO features is the finding of severe skin manifestations. Severe scarring acne, psoriasis or palmoplantar pustulosis are commonly present on examination. SAPHO syndrome has an older age of presentation. The onset is usually at 30 years(12-65 years) [6,7]. Moreover, SAPHO has predilection for different bones.

In our case most of the lesions were located in long bones and the chest wall was free. The diagnosis of CRMO was more plausible.

In all cases, both CRMO and SAPHO should be diagnosis of exclusion. It is always mandatory to rule out infection, malignancy, and systemic autoimmune diseases. The diagnosis is usually made on both clinical and radiological arguments. The MRI whole body could be useful. It detects infra-clinic osteitis sites. The cartography of the lesions could evoke the diagnosis [8]. The treatment is almost always based non-steroidal anti-inflammatory molecules for mild cases. Otherwise corticosteroids and interleukin-1 receptor antagonist (IL-1Ra) may be required. The indications for surgery are rare [9,10].

Conflict of Interest: None

Acknowledgments

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This report does not contain any personal information that could lead to the identification of the patient. A parent's consent was obtained.

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