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Chronic recurrent multifocal osteomyelitis with psoriatic skin manifestations in a 12-year-old female

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Introduction

Chronic recurrent multifocal osteomyelitis (CRMO) is a rare, noninfectious, inflammatory bone disease, which occurs mainly in childhood [1]. We present a case of CRMO and palmoplantar psoriatic skin lesions in a 12-year-old girl.

Case Presentation

A 12-year-old girl presented with recurrent erythematous palmoplantar plaques and pustules. She also complained about pain in her left ankle that started 7 months earlier. Previous magnetic resonance imaging (MRI) had consistently revealed a multifocal bone edema of the left foot. Symptoms of weakness, fever, and morning stiffness were absent. The family history was unremarkable.

Physical examination revealed well-demarcated erythematous plaques with remnants of dried pustules in a palmoplantar distribution (**Figure 1**). The active range of motion of the left upper ankle joint was painfully decreased by 50%.

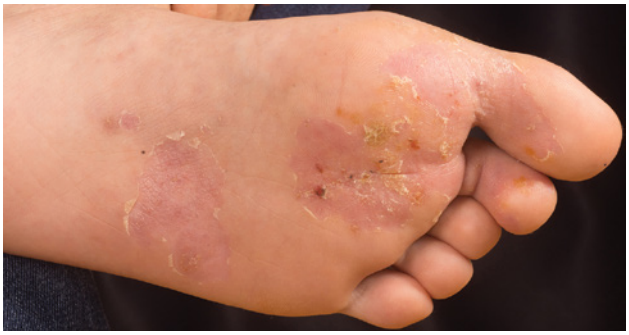

Figure 1. Well-demarcated erythematous scaly plaques with remnants of dried pustules on the left sole of the patient. [Copyright: ©2018 Epple et al.]

Laboratory results showed a slight increase of inflammation parameters, including c-reactive protein level and erythrocyte sedimentation rate. Antinuclear antibody level, rheumatoid factor, HLA-B27, and Lyme disease testing were negative.

Serial MRIs revealed fluctuating T2 hyperintensities and T1 hypointensities involving the left talus, calcaneus, and

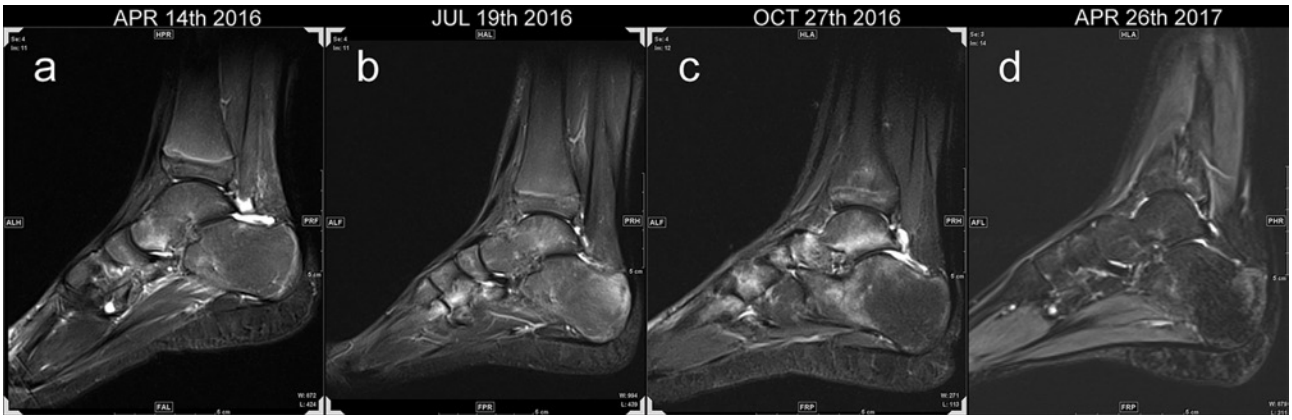

Figure 2. Serial MRIs over the course of 12 months (a-d) with fat suppressed T2-weighted images showing the fluctuating hyperintensities of the left ankle involving the calcaneus, talus, and metatarsal bones with an undulating joint effusion in the left upper ankle joint. The date of each MRI is indicated in the top row of images. [Copyright: ©2018 Epple et al.]

metatarsal bones with undulating discrete joint effusion in the left upper ankle joint (Figure 2a-d). Additionally, synovial thickening of the left talocalcaneonavicular joint was noticed. T1-weighted images after contrast application were acquired at several MR-measurements over the course of 1 year, mainly reflecting the edema seen as T2 hyperintensities.

Based on these results, the diagnosis of CRMO accompanied by palmoplantar pustular psoriasis (PPPP) was made. Treatment with oral nonsteroidal anti-inflammatory drugs (NSAIDs) and topical mometasone furoate 0.1% cream was initiated.

CRMO was first described by Giedion et al in 1972. It primarily occurs in the distal metaphyses of long tubular bones [1]. The involvement of the calcaneus, as described herein, was rarely reported. PPPP is found in approximately 15% of CRMO patients. The pathophysiology of CRMO is not well understood. Recent studies of CRMO patients described a reduced production of interleukin (IL) 10 by monocytes. This impairment may result in an increased activation of the Nod-like receptor family pyrin domains containing protein 3 inflammasome (NLRP3) leading to an enhanced expression of IL-1β, which has a role in osteoclast activation via receptor activator of nuclear factor kappa-B ligand (RANKL) stimulation. Bissonnette et al [2] described high levels of IL-1β and IL-17A in patients with PPPP leading to a secondary chemokine production of keratinocytes with accumulation of neutrophils. Thus IL-1β seems to play a key role in both CRMO and PPPP.

Conclusion

CRMO should be treated interdisciplinarily, and NSAIDs should be the medication of first choice. Skin lesions may be alleviated by topical steroids. Moreover, bisphosphonates, TNF antagonists, IL-1-inhibitors, sulfasalazine or methotrexate have been described as effective. Fortunately, our patient showed a complete remission 2 years after the onset of symptoms, which is also observed in 30% to 40% of reported cases.

In summary, we describe a rare case of CRMO initially presenting as PPPP with joint pain. Dermatologists and pediatricians should be familiar with the association of CRMO and PPPP in children to lead the way to correct diagnosis and treatment.

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