Radiology Case Reports 13 (2018) 485–489

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: http://Elsevier.com/locate/radcr

Diagnostic Imaging

The role of whole-body magnetic resonance imaging in diagnosing chronic recurrent multifocal osteomyelitis

Hannah C. Chen MD, Marc F. Wuerdeman MD, James H. Chang MD, Neris M. Nieves-Robbins MD*

Department of Radiology, Madigan Army Medical Center, 9040A Jackson Ave, Tacoma, WA 98431, USA

ARTICLE INFO

Article history:
Received 2 July 2017
Received in revised form 25 November 2017
Accepted 30 November 2017
Available online 9 January 2018

Keywords:
CRMO
DWI

ABSTRACT

Chronic recurrent multifocal osteomyelitis (CRMO) is an uncommon idiopathic inflammatory disorder. The diagnosis is often delayed because a variable clinical presentation and limited awareness among care providers. We present an 11-year-old female diagnosed with CRMO and her imaging workup. In particular, this case highlights the role of whole-body magnetic resonance imaging to enhance detection and diagnosis of CRMO.

Introduction

Chronic recurrent multifocal osteomyelitis (CRMO) is an uncommon idiopathic inflammatory disorder that is characterized by recurrent episodes of noninfectious osteomyelitis. Giedion et al. reported the first case of CRMO in 1972, describing it as a subacute and chronic recurrent symmetric osteomyelitis. The authors also described the disease process as having multiple bone lesions that predominantly affect the metaphyseal regions [1,2]. The varied clinical presentation of CRMO contributed to it being reported by many different names in the literature. It was not until 1978 that Probst et al. firmly established the disease's name as CRMO [1].

CRMO manifests as remitting and relapsing musculoskeletal pain with a protracted course. It primarily affects children and adolescents, with a female-to-male ratio of 2-4:1 [1,3]. The initial presentation typically consists of swelling and pain over the affected bone. Associated radiographic findings suggestive of osteomyelitis are also noted at the time of presentation.

Unfortunately, the diagnosis of CRMO is often delayed because of its variable clinical presentation and limited awareness of this condition among care providers [2]. To avoid delays in diagnosis, it is imperative that both clinicians and radiologists understand the presentation and the radiological findings of CRMO.

This case report describes the clinical presentation of an 11-year-old female with CRMO and her diagnostic imaging

Competing Interests: The authors have declared that no competing interests exist.
The views expressed in this article are those of the authors and do not reflect the official policy or position of the Department of the Army, Department of Defense, or the U.S. Government.

* *Corresponding author.*
E-mail address: Hannah.c.chen.mil@mail.mil (H.C. Chen).

https://doi.org/10.1016/j.radcr.2017.11.014

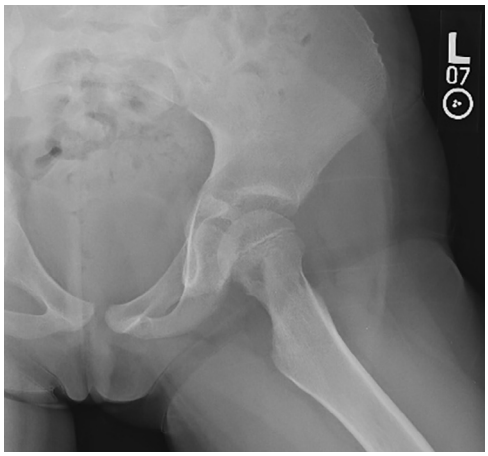

Fig. 1 – Left hip radiograph, frog-leg view. Eccentric lytic lesion at the medial aspect of the proximal femoral metaphysis. L, left.

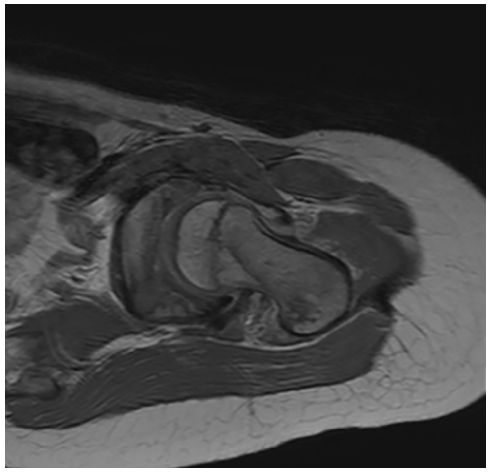

Fig. 2 – Left hip magnetic resonance imaging, oblique axial proton density sequence; curvilinear low signal intensity line abuts the physis in the proximal femoral metaphysis consistent with an occult fracture.

workup. In particular, this case report highlights the role of the sequences performed during a whole-body magnetic resonance imaging (MRI) to enhance detection and diagnosis of this previously unrecognized condition in this patient.

Case report

An 11-year-old girl presented to our facility with chronic multifocal joint pain. Her medical history is significant for a left hip injury sustained after doing cartwheels at the age of 7 years and complaints of chronic multifocal joint pain since the time of injury. Radiographs of the left hip obtained at the time of injury demonstrated an eccentric lytic lesion abutting the medial aspect of the proximal femoral metaphysis (Fig. 1). These plain radiograph findings prompted further evaluation with a left hip MRI. The initial hip MRI demonstrated a fracture involving the left femoral epiphysis, metaphysis, and diaphysis with surrounding marrow edema (Fig. 2). The fracture line was not evident, even in retrospect, on plain radiograph. Despite treatment, the patient continued to have pain in her left hip. A repeat MRI was performed 1 month later to evaluate for resolution. The second MRI showed unchanged left proximal femoral marrow edema surrounding a stable-appearing fracture line and no evidence of callus (Fig. 3A-C). The lack of healing raised concerns for an underlying pathologic process predisposing to a fracture. At the time, differential diagnostic considerations included osteomyelitis and Langerhans cell histiocytosis. Although typically epiphyseal in location, chondroblastoma was also included in the differential, given the extensive marrow edema. Ewing sarcoma, lymphoma, or leukemic involvement of the bone were considered to be less likely differential diagnostic considerations because there was no associated soft tissue mass. The patient was referred for a left hip curettage. Evaluation of the pathology sample showed bone fragments and hematopoietic elements but no evidence of either acute inflammation or malignancy.

Over the course of the next 4 years, the patient continued to experience waxing and waning pain in her left hip. She also complained of intermittent pain in her knees and right shoulder. The patient subsequently presented with a 1-month history of right ankle pain that was associated with warmth and swelling, findings that were concerning for osteomyelitis. Radiographs of the patient's right ankle showed a mottled appearance of the lateral aspect of the distal right tibial metaphysis with mixed areas of sclerosis and lucency (Fig. 4).

The patient was referred to a pediatric rheumatologist for further evaluation because of the multifocal joint involvement. Laboratory studies revealed a mildly elevated C-reactive protein and erythrocyte sedimentation rate. No other laboratory abnormalities were found. A whole-body MRI was ordered after consultation with a pediatric radiologist. The following MRI sequences were obtained: axial diffusion-weighted imaging (DWI), axial apparent diffusion coefficient (ADC), and coronal whole-body short-tau inversion recovery (STIR). The MRI study showed a hyperintense signal within the left femoral head and neck, and proximal diaphysis on the STIR sequence, with corresponding hyperintensity on DWI and ADC sequences (Fig. 5A-C). Confluent STIR hyperintensity was also noted in multiple areas to include the right proximal humerus, acetabular roofs, knees, and distal tibiae without restricted diffusion.

The diagnosis of CRMO was made based on a combination of the clinical presentation, chronicity of the complaints, the multifocal involvement, history of a negative bone biopsy, and the most recent MRI findings.

Discussion

CRMO is characterized by an insidious onset of vague pain, swelling and tenderness over an affected joint [4]. The course of CRMO consists of intermittent periods of exacerbations and improvement in musculoskeletal pain. The mean onset of symptoms has been reported between 8-14 years of age [1,4,5]. Several studies have demonstrated that the symptoms can last

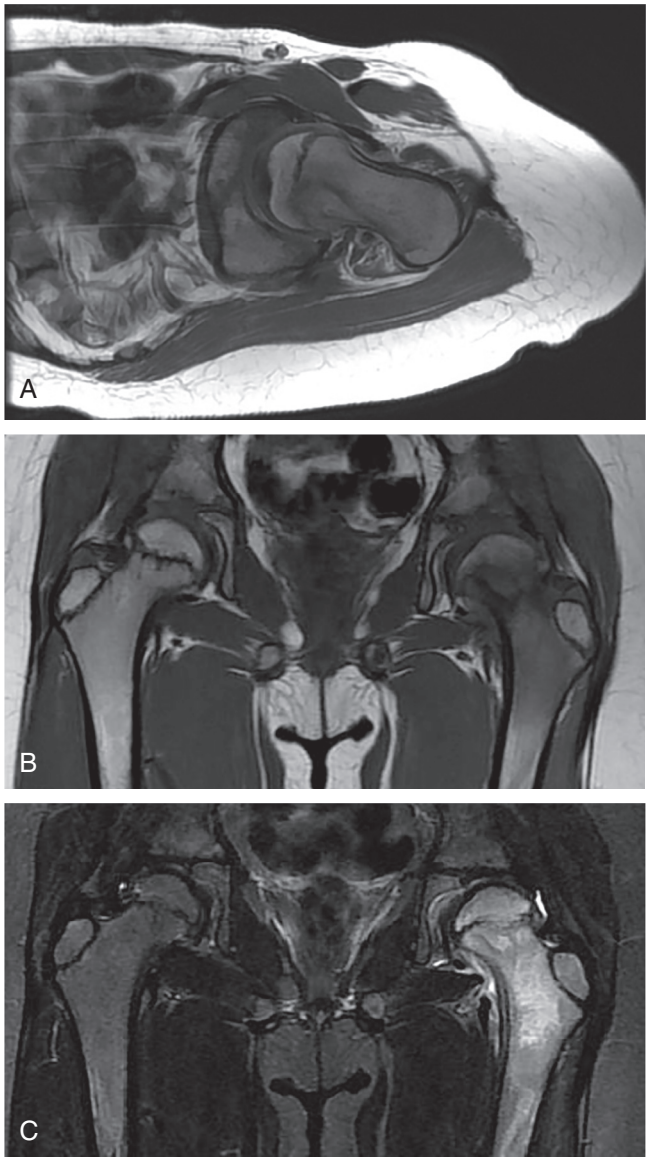

Fig. 3 – (A) Left hip MRI, oblique axial proton density sequence; curvilinear low signal intensity line abuts the physis in the proximal femoral metaphysis without significant interval callus formation. (B) Left hip MRI, coronal T1 sequence; curvilinear low signal intensity line abuts the physis in the proximal femoral metaphysis with surrounding hypointense marrow. (C) Left hip MRI, short-tau inversion recovery sequence, curvilinear low signal intensity line with surrounding hyperintense signal in the marrow of the proximal femur. MRI, magnetic resonance imaging.

anywhere from 2.5 years to as long as 25 years after the initial diagnosis [1]. Laboratory findings at initial presentation or during an exacerbation are often nonspecific, showing an elevated erythrocyte sedimentation rate and C-reactive protein with a normal white blood cell count [5,6].

Initial imaging typically consists of radiographs, which may be normal or may demonstrate lesions near the metaphysis and growth plates. Lesions can range from purely osteolytic,

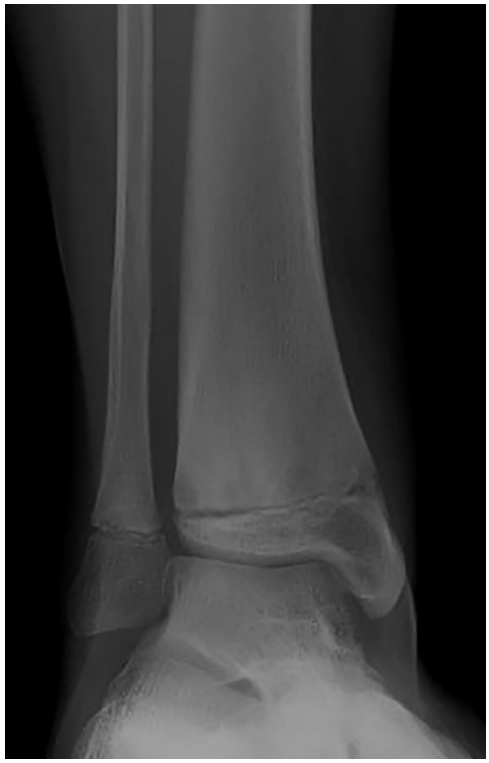

Fig. 4 – Right ankle radiograph, oblique view. Mottled appearance of the right tibial metaphysis adjacent to the physeal plate.

mixed lytic, and sclerotic to purely sclerotic, depending on the chronicity [1]. The lesions are characteristically symmetric in morphologic appearance with involvement of the metaphyses and the epiphyses of long bones. However, lesions often lack a clinical and temporal symmetry [1,5]. The most commonly affected long bones are the femur and the tibia. The spine, pelvis, mandible, hands, and feet can also be involved [1]. Involvement of the clavicle is unique to CRMO and helps distinguish it from other processes [1].

The degree of osseous involvement present on imaging that is not clinically apparent is a characteristic finding in CRMO [2]. Therefore, whole-body imaging is often obtained to help identify asymptomatic lesions, as well as to establish a baseline of disease burden. Whole-body imaging can be been done with Tc-99 bone scintigraphy or with MRI. Radionuclide studies demonstrate areas of increased uptake, with early uptake suggesting inflammation and late uptake bone sclerosis [5]. Bone scans are not performed as frequently because of the uptake in the growth plates, as well as patient exposure to radiation. Whole-body MRI has become the study of choice as it does not expose the patient to radiation and provides better evaluation of the anatomy and soft tissues [1,2,5,7]. MRI can also demonstrate marrow edema, periostitis, soft tissue inflammation, transphyseal disease and joint involvement. Acute findings on MRI are hyperintense on fluid-sensitive sequences and enhance with contrast. On the other hand, chronic lesions are hypointense on both T1 and T2 because of sclerosis of the lesion [1,6]. Potential disadvantages of MRI include limited evaluation of the ribs and the skull because of a large slice thickness.

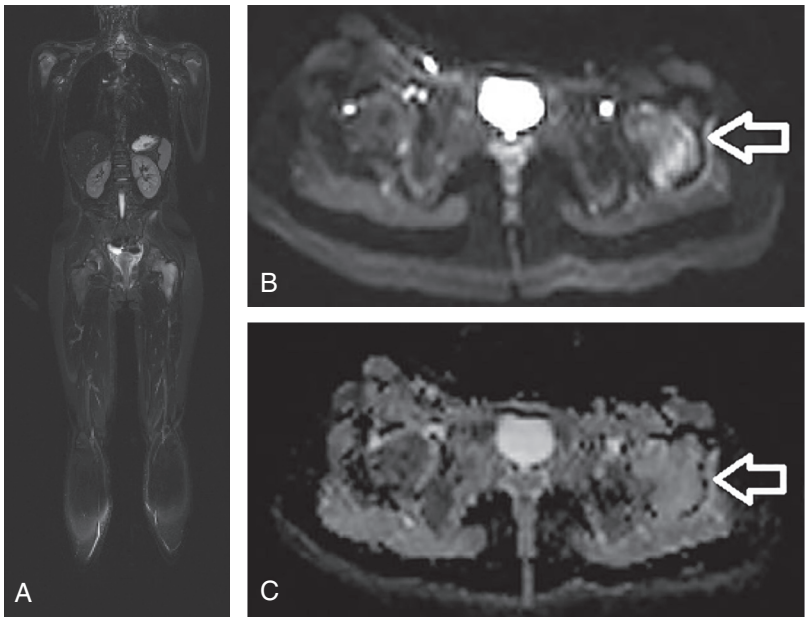

Fig. 5 – (A) Whole-body MRI, coronal short-tau inversion recovery sequence demonstrating a hyperintense signal in the right proximal humerus and in the left proximal femur. (B) Whole-body MRI, axial diffusion-weighted sequence through the pelvis demonstrating a hyperintense signal in the proximal left femur (arrow). (C) Whole-body MRI, axial apparent diffusion coefficient sequence through the pelvis demonstrating a hyperintense signal in the proximal left femur (arrow). MRI, magnetic resonance imaging.

Additional drawbacks are increased cost and general anesthetic, which is often required in younger children.

Although whole-body MRI has become one of the mainstays in making the diagnosis of CRMO, the findings of CRMO may overlap with those of bacterial osteomyelitis and bone tumors, such as lymphoma, Ewing sarcoma, and Langerhans cell histiocytosis [5,6,8]. DWI is now used as an aid in narrowing the differential diagnosis. DWI measures Brownian motion of water in tissues in the body, and the ADC maps yielded from DWI provide a quantitative measure of the Brownian motion [9]. In highly cellular tissue, there is restricted water motion and therefore a lower ADC value. The low ADC value corresponds with a hypointense signal, whereas a high ADC has a hyperintense signal. Thus, DWI and ADC sequences are useful for assessing the cellularity within a given lesion [10–13]. Leclair et al. recently evaluated the role of DWI and ADC values in patients with CRMO [7]. Leclair et al.'s study found that bone lesions in CRMO restricted diffusion and had increased ADC values. Thus, benign inflammatory lesions could be differentiated from some malignant bone lesions based on the DWI and ADC patterns. Douis et al. also found that there was a statistically significant difference in benign and malignant skeletal lesions in the pelvis based on DWI findings, but not in ADC values. Instead, Douis et al. concluded that, in differentiating benign from malignant skeletal lesions, a minimum ADC value was more reliable than a mean or maximum ADC value [10].

The role of DWI and ADC in distinguishing infectious osteomyelitis and CRMO is still controversial. Several studies have demonstrated restricted diffusion with a corresponding low ADC signal associated with infectious osteomyelitis. The study by Douis et al. found the mean ADC values associated with osteomyelitis to be lower than chronic inflammatory cells but higher than adjacent muscles [10]. However, Herneth et al. found the ADC values to be variable, depending on the amount of pus vs watery content [11]. However, whole-body MRI is still useful in identifying secondary findings, such as abscess, associated with infectious osteomyelitis.

In the case of our patient, there was hyperintense signal on DWI and ADC. These findings were indicative of a benign process and helped the radiologists arrive at the appropriate diagnosis. The addition of DWI could provide considerable diagnostic information in cases of suspected CRMO. This approach could ultimately prevent unnecessary medical interventions such as invasive procedures or higher-risk treatment options. Additionally, the use of whole-body MRI with DWI in patients with CRMO may help establish baseline disease burden and response to treatment.

REFERENCES

[1] Khanna G, Sato TSP, Ferguson P. Imaging of chronic recurrent multifocal osteomyelitis. Radiographics 2009;29(4):1159–77. doi:10.1148/rg.294085244.
[2] Fritz J. The contributions of whole-body magnetic resonance imaging for the diagnosis and management of chronic recurrent multifocal osteomyelitis. J Rheumatol 2015;42(8):1359–60. doi:10.3899/jrheum.150676.
[3] Huber AM, Lam P-Y, Duffy CM, Yeung RS, Ditchfield M, Laxer D, et al. Chronic recurrent multifocal osteomyelitis: clinical outcomes after more than five years of follow-up. J Pediatr 2002;141(2):198–203. doi:10.1067/mpd.2002.126457.
[4] Roderick MR, Shah R, Rogers V, Finn A, Ramanan AV. Chronic recurrent multifocal osteomyelitis (CRMO)—advancing the diagnosis. Pediatr Rheumatol Online J 2016;14(1):47. doi:10.1186/s12969-016-0109-1.

[5] Wipff J, Adamsbaum C, Kahan A, Job-Deslandre C. Chronic recurrent multifocal osteomyelitis. Joint Bone Spine 2011;78(6):555–60. doi:10.1016/j.jbspin.2011.02.010.
[6] Ract I, Storey J, Travers JY, Pastural G, Gayon P, Balu M. Chronic recurrent multifocal osteomyelitis. Diagn Interv Imaging 2015;96(3):313–6. doi:10.1016/j.diii.2014.12.004.
[7] Leclair N, Thörmer G, Sorge I, Ritter L, Schuster V, Hirsch FW. Whole-body diffusion-weighted imaging in chronic recurrent multifocal osteomyelitis in children. PLoS ONE 2016;11(1):e0147523. doi:10.1371/journal.pone.0147523.
[8] Wintrich S, Horneff G. Characteristics and outcomes of chronic non-bacterial osteitis in children. Eur J Rheumatol 2015;2(4):139–42. doi:10.5152/eurjrheum.2015.0020.
[9] Oh E, Yoon YC, Kim JH, Kim K. Multiparametric approach with diffusion- weighted imaging and dynamic contrast-enhanced MRI: a comparison study for differentiating between benign and malignant bone lesions in adults. Clin Radiol 2017;72:1–8. doi:10.1016/j.crad.2017.02.017.
[10] Douis H, Davies MA, Sian P. The role of diffusion-weighted MRI (DWI) in the differentiation of benign from malignant skeletal lesions of the pelvis. Eur J Radiol 2016;85(12):2262–8. doi:10.1016/j.ejrad.2016.10.014.
[11] Herneth AM, Ringl H, Memarsadeghi M, Fueger B, Friedrich KM, Krestan C, et al. Diffusion weighted imaging in osteoradiology. Top Magn Reson Imaging 2007;18(3):203–12. doi:10.1097/RMR.0b013e3180cac61d.
[12] Khoo MMY, Tyler PA, Saifuddin A, Padhani AR. Diffusion-weighted imaging (DWI) in musculoskeletal MRI: a critical review. Skeletal Radiol 2011;40(6):665–81. doi:10.1007/s00256-011-1106-6.
[13] Yao K, Troupis JM. Diffusion-weighted imaging and the skeletal system: a literature review. Clin Radiol 2016;71(11):1071–82. doi:10.1016/j.crad.2016.07.007.