CASE REPORT

Chronic recurrent multifocal osteomyelitis in a 3.5-year-old boy

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We report an extremely rare case of multifocal bone disorder in a 3.5-year-old boy who appeared for left forearm and arm pain and multiple periods of fever with an unusual presentation of lymphoma/leukemia and highlight diagnostic challenges leading to a misdiagnosis, which was then diagnosed and treated for chronic recurrent multifocal osteomyelitis (CRMO). Based on a left arm biopsy and whole-body scans, he was eventually diagnosed with CRMO. Taken together, in this case, we noticed a notable amelioration after a 5-month treatment with nonsteroidal anti-inflammatory drugs on multiple bone pains.

Key words: Bone, chronic recurrent multifocal osteomyelitis, eosinophilic granuloma, lymphoma

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INTRODUCTION

Chronic recurrent multifocal osteomyelitis (CRMO), or chronic nonbacterial osteomyelitis, is an extremely scarce condition in children with low-incidence, noninfectious inflammatory bone disease.[1] CRMO which occurs fundamentally in children and adolescents may be misdiagnosed as osteomyelitis, lymphoma/leukemia, arthritis, or cancer.[2-4] Furthermore, CRMO has unknown etiology, but agents or factors containing the disorders of inflammatory, genetic, autoimmunity, juvenile seronegative spondyloarthropathy, and infectious organism have been shown to play a major role as well as CRMO epidemiological data are sparse and contain little case series and zonal cohorts plus, clinical symptoms presented are various, such as bone pain, local swelling, scarcely skin redness, and heat.[1-5] In addition, in the early stages of diagnosis, complete body imaging using magnetic resonance imaging (MRI) (turbo inversion recovery magnitude) techniques should be performed to diagnose silent clinical lesions, exclusively in the vertebral column, and also to assess CRMO activity during follow up, the exploration and monitoring the consequences of CRMO, which may include fractures, inflammatory involvement, and tissue damage to surrounding structures and histopathology findings show the chronic inflammation together with granulocytes, lymphocytes, plasma cells, and monocytes in later stages.[1-7] We hereby describe the clinical and radiological data on a rare case of CRMO treated with nonsteroidal anti-inflammatory drugs (NSAIDs).

CASE REPORT

A 3.5-year-old male patient with left forearm pain and fever was admitted to the pediatrician. After initial examinations, including hemoglobin: 8, hematocrit: 32.3, white cell count (white blood cell): 8100, lymph: 51.7%, platelet count: 462,000, erythrocyte sedimentation rate: 72 and X-ray [Figure 1a], lymphoma/leukemia was suggested. The patient was also examined by MRI, biopsy of left radius, flow cytometry, and bone marrow aspirate. The pediatrician, due to the significance of the issue of initiating treatment, suggested the patient's

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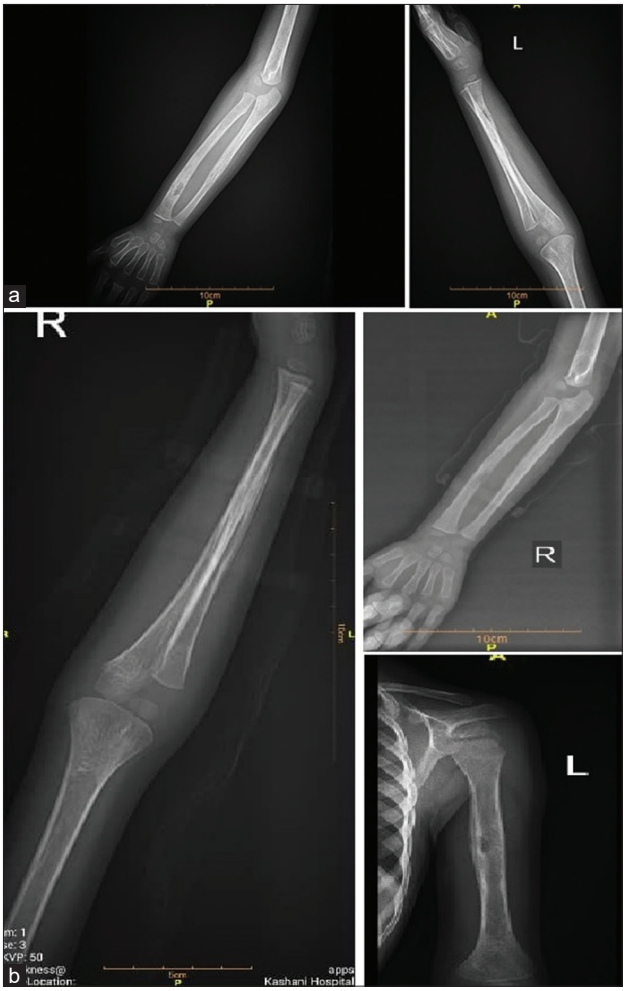

Figure 1: (a) Increased density in medulla of both ulna and radius bones associated with permeative lytic lesions in radius is seen. Cortical irregularity is also evident in some areas. No obvious soft tissue. The wrist seems to be intact. (b) There is a lytic lesion with a wide zone of transition in midshaft of the left humerus bone. Adjacent cortical irregularity and disruption is also noted associated with lamellated periosteal reaction. No obvious soft tissue is evident

referral to the orthopedic department. With the suspicion of eosinophilic granuloma, the patient underwent a biopsy of the forearm, in which many necroses were reported without a decisive diagnosis, but was not referred due to his recovery. The probability of eosinophilic granuloma was considered and the patient did not receive treatment for lymphoma/leukemia and only took the medication and becomes a follow-up. Five months later, the patient returns with the right forearm pain and left arm (and the previous location of the lesion in the left forearm X-ray showed that the lesion was removed) [Figure 1b]. The patient was re-examined for supplementary lymphoma/leukemia, and a whole-body scan was taken from the patient (metabolically active bony lesion of the left humerus and right forearm); as a result, diagnosis of eosinophilic granuloma was rejected. Finally, as a result of the left arm biopsy, CRMO was diagnosed. Taken together, the patient was treated with NSAIDs and recovered quickly and his graphs ultimately became normal.

DISCUSSION

Similar to our diagnosis approach, Girschick *et al*., 2007, reported the major role of pathologic biopsy in ruling out other diagnoses such as cancer and infectious diseases, so biopsy must be considered, particularly since it is often hard to create a definite detection among oncological bone lesions and those related to CRMO.[8] Gicchino *et al*., 2018, concluded that the most common differential diagnosis of CRMO is shown by infections, malignant bone tumors, and Langerhans cell histocytosis.[9] In addition, a study by Fritz *et al*., 2015, revealed that the use of the whole-body MRI was beneficial in reaching the diagnosis,[10] which is consistent with our case. Furthermore, our study revealed that a whole-body MRI scan seems to have even better diagnostic potential and sensitivity. A number of studies have been reported that that whole-body MRI is very valuable, although expensive and this method may become a diagnostic standard in the work-up of doubtful CRMO in future.[11] Moreover, a review evaluation by Schultz *et al*. of case reports of CRMO showed that 79% of patients had a good response to NSAIDs.[12] In addition, Walsh *et al*. recommend that CRMO signs are generally well controlled by NSAID therapy; therefore, in our experience, NSAIDs are the first choice for the initial treatment in our case.[13] Furthermore, a report by Pham *et al*. examined a 38-year-old man who presented with lower extremity musculoskeletal pain with the presentation of Hodgkin lymphoma as a misdiagnosis instead of CRMO.[14]

CONCLUSION

This case report demonstrates the diagnostic difficulties of a musculoskeletal presentation of lymphoma, challenges of nondiagnostic bone pain, and yields challenges of diagnostic delays in providing a potential therapeutic treatment for CRMO. We consider that whole-body scans and bone biopsy can be reliably performed in children for the diagnosis of CRMO. Therefore, we propose that children with bone pain due to CRMO involvement be considered for NSAIDs therapy primary after diagnosis.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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