Bone scintigraphy in diagnosing chronic recurrent multifocal osteomyelitis

Case Report

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(Received 8 June 2016, Revised 1 September 2016, Accepted 2 September 2016)

ABSTRACT

A 10-year-old boy was referred to us for evaluation of FUO accompanied with bone pain in both calves. Three hours after intravenous injection of 13 mCi of ^{99m}Tc-MDP, whole body scan in multiple spot views was performed. The scan showed symmetrical areas of diffusely increased tracer uptake in multiple long bones. Histopathologic evaluation confirmed osteosclrosis and fibrotic changes without any bacterial growth in the specimen culture. Based on patient's history, lab results, bone scan and histopathologic findings, chronic recurrent multifocal osteomyelitis (CRMO) was considered as the most likely diagnosis. Dramatic response to NSAIDs and pamidronate therapy confirmed the diagnosis of CRMO.

Key words: Chronic recurrent multifocal osteomyelitis; Bone scintigraphy; ^{99m}Tc-MDP

Iran J Nucl Med 2017;25(Suppl 1):66-69
Published: February, 2017
http://irjnm.tums.ac.ir

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INTRODUCTION

Chronic recurrent multifocal osteomyelitis (CRMO) is an unusual form of osteomyelitis which is more common in females [1]. This auto-inflammatory bone disease primarily occurs in children and adolescents and has alternative exacerbation and remission cycles [2, 3]. Metaphysis of the long bones particularly tibia, femur and clavicle are the most involved bones; however lesions can be observed in the epiphysis and diaphysis and flat bones such as ribs, mandible and sternum [3]. Although CRMO is a multifocal symmetric bone disease with unknown etiology, in recent years, some researchers have proposed relations between this disease and autoimmune disorders. It has also been established that symmetrical pattern is not necessary for diagnosis of CRMO [1]. Initial presentation is often limited to a single bone which mimics acute osteomyelitis with a negative culture. Involvement of other parts of the skeleton is always seen over time when older lesions are in resolving phase [1, 4].

Clinical manifestations are non-diagnostic with sudden onset of fever and localized tenderness of the involved bone. The imaging modalities for detection and localization of abnormalities include plain radiography, CT scan, MRI and bone scintigraphy [5].

Plain radiography is the first step in evaluation of CRMO. It can detect lytic and sclerotic lesions with periosteal reactions, although lots of lesions are not detectable in plain radiographs [6].

CT images can determine extension of lesions. If there is any erosive pattern or periosteal reaction, CT images can detect it. Usual pattern of CRMO in CT scan includes multiple focal zones of lytic areas surrounding with sclerosis [7].

During the active phase of the disease, MR imaging shows typical findings of marrow edema, which appears hypointense on T1-weighted images and hyperintense on T2-weighted images. MR imaging can demonstrate associated periostitis, soft-tissue inflammation, and transphyseal disease—findings.

Bone scintigraphy is a non-invasive method with no adverse effects which can detect multiple foci of involved bones and determine the most appropriate spot for biopsy.

Histopathologic findings are not specific, but inflammatory changes followed by sclerosis and fibrotic tissue can be observed [8]. Diagnosis can be confirmed based on clinical picture, findings of imaging modalities and pathology results. Differential diagnosis includes neoplastic diseases with hyperactive bone marrow or metastatic lesions from Ewing osteosarcoma [5].

CASE REPORT

Our case is a 10-year-old boy who was referred to our nuclear medicine department with a 5-month history of fever and bone pain which was more prominent in both legs and forearms. He had been admitted to the hospital a month earlier. Lab tests showed a white blood cell count (WBCs) of 10200 per mm^3, an erythrocyte sedimentation rate (ESR) of 67 mm and a 2 plus C-reactive protein, all indicating an inflammatory process. In plain radiographs of both legs and forearms, only diffuse cortical sclerosis was reported. (Figure 1) Bone marrow aspiration and biopsy from the iliac bone showed mild dysplastic changes with a blast cell count less than 5%. Rheumatologic tests such as Anti-nuclear antibody (ANA) and HLA B27 were negative. ASO titer was more than 400.

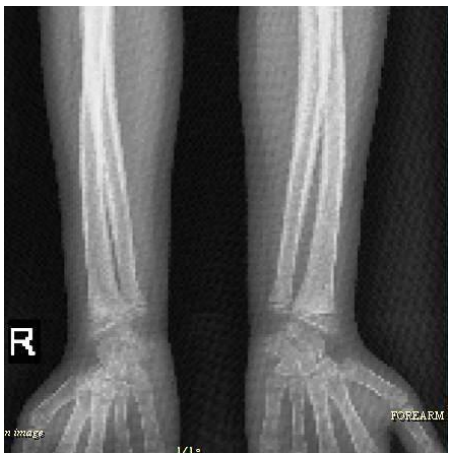

Fig 1. Diffuse cortical sclerosis in plain radiography of the patient's legs and forearms.

We performed a whole body bone scan to localize any abnormality in the skeletal system (Figure 2). Three hours after intravenous injection of 13 mCi of ^{99m}Tc-MDP, scanning was performed in multiple anterior and posterior spot views (5 minutes per view time) using a dual head gamma camera equipped with low energy high resolution collimator.

The spot images showed symmetrical areas of diffusely increased tracer uptake throughout both humeri, ulnas and tibias. In addition, increased tracer uptake due to osteoblastic reaction was observed in mid and distal parts of both femurs. The scan also showed increased tracer uptake in the mandible (Figure 3).

According to the patient's history and lab results, an inflammatory process was considered. Bone scan pattern suggested multifocal bone involvement and it determined the appropriate site for biopsy.

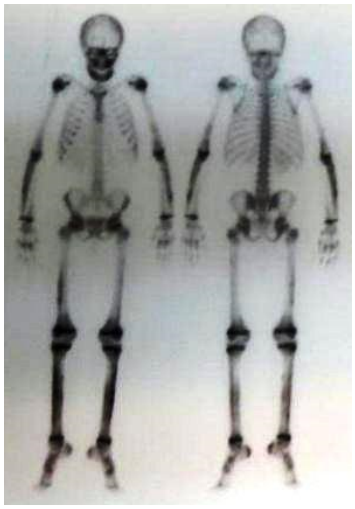

Fig 2. Whole body bone scan (left: anterior view and right: posterior view) three hours after intravenous injection of 13 mCi of ^{99m}Tc-MDP.

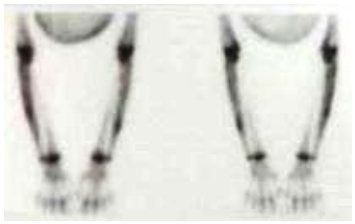

Fig 3. Anterior (left) and posterior (right) views of the spot images of bone scan.

Surgical open biopsy was performed on the proximal part of the right tibia and the pathology report showed sclerosis with fibrotic changes and no malignant neoplastic tissue. Based on these findings, the diagnosis of chronic recurrent multifocal osteomyelitis was presumed and medical therapy started. Patient was successfully treated with NSAIDs and pamidronate and a relief of pain over the following weeks was observed.

DISCUSSION

Chronic recurrent multifocal osteomyelitis is an uncommon type of osteomyelitis with auto inflammatory process and has characterized by multiple sites of bone involvement, cyclic exacerbations and remissions [9]. It occurs more frequently in children and adolescence with female to male predominance. Unlike acute bacterial osteomyelitis, bacteriologic evaluation of biopsy specimen is always negative pointing that inflammatory process is not due to bacterial infection [8]. Thus, antibiotic therapy is not recommended for this disease and typically response to treatment with NSAIDs is reported. In recent years, bisphosphonate drugs especially pamidronate are proposed for medical therapy and were associated with good response to treatment [5, 10].

The most common locations are long bones include tibia, femur and clavicle. However, involvement of ribs, sternum and vertebrae may be seen in some rare cases [3]. Diagnosis of this condition is of utmost importance to decrease unnecessary antibiotic therapy and costly invasive procedures [1, 11]. Clinical presentation and radiographic changes are appropriate clues for diagnostic suspicion, but sometimes radiography cannot be helpful [3, 5]. In this situation, MRI or bone scintigraphy play an important role in diagnosis. The bone scan reveals multiple sites of increased tracer uptake, particularly in the extremities which are compatible with involved bones. Bone scan can also help clinician to determine the best site for biopsy [5].

CONCLUSION

In conclusion, bone scan is an available non-invasive method for localization of skeletal abnormalities in chronic recurrent multifocal osteomyelitis.

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