ABSTRACT

We report a case of chronic recurrent multifocal osteomyelitis in a 9-year-old girl. She presented with a 9-month history of gradually worsening pain and swelling in her left foot. Non-steroidal anti-inflammatory drugs were prescribed but the symptoms persisted. She underwent curettage through a small oval corticotomy window on the first metatarsal bone. The pain and swelling improved promptly and she was able to walk without pain 2 weeks later. Curettage enabled rapid symptomatic relief and induced remission, with little risk of complications.

Keywords: anti-inflammatory agents, non-steroidal; curettage; osteitis; osteomyelitis

INTRODUCTION

Chronic recurrent multifocal osteomyelitis (CRMO) is also known as chronic sclerosing osteomyelitis, condensing osteitis, sclerosis and hyperostosis, primary chronic osteomyelitis, pustulotic arthroosteitis, and lymphoplasmacellular osteomyelitis. It is an inflammatory disorder of unknown origin. It is part of the spectrum of non-bacterial osteitis (NBO) that is classified into acute or chronic non-bacterial osteomyelitis or CRMO. CRMO is the most severe form of NBO (Table 1).

CASE REPORT

In November 2007, a 9-year-old girl presented with a 9-month history of gradually worsening pain and swelling in her left foot (Fig. 1), with no history of any trauma. No surrounding soft tissue inflammation was noted. The pain was continuous, deep, and dull and intensified during walking. She also complained of occasional low fever but no loss of weight or appetite. She had no history of skin lesions, rashes, arthralgia, arthritis, myalgia, or myositis. Two years earlier, a similar episode of pain and swelling had occurred in her right foot and symptoms resolved after curettage. No other organs were involved. Non-steroidal anti-inflammatory drugs (NSAIDs) were prescribed but...
the symptoms persisted.

Her erythrocyte sedimentation rate (ESR) was mildly raised. Her total white cell counts and other blood tests were within normal limits. Radiographs revealed mixed centrally placed lytic lesions and eccentrically located sclerotic lesions in the first metatarsal bone. The cortices were thickened and the diaphysis appeared wider than normal (Fig. 2).

An open biopsy and curettage was performed through a small oval corticotomy window on the superior aspect of the first metatarsal bone (Fig. 3). A thick yellowish substance mixed with blood, dead bone, debris, slough, and pus was curetted and the bone was irrigated with saline. Cultures did not yield any organisms; the inflammatory changes were consistent with acute and chronic infection. The oval window on the bone was left open for drainage and prevention of further collection. The pain and swelling improved promptly and the patient was able to walk without pain 2 weeks later. Her ESR returned to normal a month later.

**DISCUSSION**

CRMO mainly affects young girls (peak age of onset usually at 10 years), with a female to male ratio of 4:1.\(^9\) It is characterised by chronic, multiple, focal inflammatory lesions of the bone with periods of exacerbation and remission. The involved areas are often symmetrical, unlike infectious osteomyelitis. It usually affects the clavicle and sternum, followed by the long bones, tarsal bones, ribs and spine.\(^10-13\)

It usually causes mild to moderate bony pain, swelling, malaise, and fever; arthritis of the adjacent and distant joints occurs occasionally. It is often associated with other inflammatory conditions, including peripheral arthritis, sacroiliitis, psoriasis, pustulosis palmoplantaris (synovitis, acne, pustulosis, hyperostosis, and osteitis), pyoderma gangrenosum, inflammatory bowel disease, and Sweet syndrome.\(^14\)

The aetiology of CRMO is unknown; suggested causes include *Propionibacterium acnes*, atypical mycobacteria, anaerobic streptococci, or coagulase-negative staphylococci,\(^15\) as well as a viral aetiology and an autoimmune process.\(^10\)

Cultures usually fail to grow any organisms.\(^16,17\) The histopathologic features are consistent with non-specific acute or chronic inflammation,\(^18\) and cannot

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**Table 1**

Classification of non-bacterial osteitis (NBO)

<table>
<thead>
<tr>
<th>NBO</th>
<th>Presentation</th>
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<tbody>
<tr>
<td>Acute NBO</td>
<td>Symptoms not longer than 6 months with at least one bone lesion</td>
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<tr>
<td>Chronic NBO</td>
<td>Persistent symptoms (without remission) lasting longer than 6 months with at least one bone lesion</td>
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<tr>
<td>Chronic recurrent multiple osteomyelitis</td>
<td>Multiple bone lesions, or one bone lesion with palmoplantar pustulosis, recurrent flares with remissions</td>
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**Figure 1** Swelling over the dorsum of the first metatarsal.

**Figure 2** Radiographs showing cortical expansion, mixed lytic and sclerotic lesions, and widening of the diaphysis of the first metatarsal.
differentiate the condition from other types of chronic inflammatory bone lesions in children. CRMO can begin as an acute inflammation with polymorphs and microabscesses. Then, infiltration with lymphocytes predominates, with granulomatous foci and reactive new bone formation. The granulation tissue contains lymphocytes, plasma cells, neutrophils, connective tissue, and multinucleate giant cells.

The diagnosis of CRMO is mainly based on the clinical history, physical examination, and radiological findings. The differential diagnosis includes malignancy (osteosarcoma, Ewing’s sarcoma, neuroblastoma), osteoid osteoma, and subacute bacterial osteomyelitis. Classic CRMO is diagnosed when the following criteria are met: (1) multifocal bone lesions, (2) prolonged clinical course over several years with remissions and exacerbations, (3) lack of response to antimicrobial therapy, and (4) radiographic examination showing multiple foci of osteolysis surrounded by sclerosis.

Four major and 6 minor criteria for classifying and diagnosing NBOs have been reported. Patients with at least 2 major criteria or one major and 3 minor criteria are diagnosed as having NBO. Patients with at least 3 major and 4 minor criteria are diagnosed as having CRMO (Table 2).

<table>
<thead>
<tr>
<th>Diagnostic criteria of non-bacterial osteitis (NBO)*</th>
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<tbody>
<tr>
<td><strong>Major</strong></td>
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<tr>
<td>Radiologically proven osteolytic/sclerotic lesion</td>
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<tr>
<td>Multifocal bone lesions</td>
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<tr>
<td>Palmo/plantar pustulosis or psoriasis</td>
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<tr>
<td>Sterile bone biopsy with signs of inflammation and/or fibrosis, sclerosis</td>
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<tr>
<td><strong>Minor</strong></td>
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<tr>
<td>Normal blood count and good general health</td>
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<tr>
<td>C-reactive protein and erythrocyte sedimentation rate mildly to moderately elevated</td>
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<td>Observation time longer than 6 months</td>
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<td>Hyperostosis</td>
</tr>
<tr>
<td>Associated with other autoimmune diseases apart from palmoplantar pustulosis or psoriasis</td>
</tr>
<tr>
<td>Grade I or II relatives with autoimmune or autoimmune inflammatory disease, or with non-bacterial osteitis</td>
</tr>
</tbody>
</table>

* Patients with at least 2 major criteria or one major and 3 minor criteria are diagnosed as having NBO. Patients with at least 3 major and 4 minor criteria are diagnosed as having chronic recurrent multifocal osteomyelitis

The treatment goals for CRMO are maintaining normal bone growth and function of the adjacent joint. Antibiotic treatment is considered ineffective, NSAIDS are the treatment of choice for CRMO, NSAIDS can induce remission in up to 85% of patients with NBO. Remission is defined as no pain, a decrease in inflammatory markers back to normal values, and no radiological progression. In patients with frequent relapses and those who do not respond to NSAIDs, the use of steroids may be necessary. Bisphosphonates, interferon gamma, and TNF blockers have all been reported as useful for treating CRMO. In patients resistant to NSAIDs or steroids, curettage enables rapid symptomatic relief and induces remission, with little risk of complications. Cultures and biopsy specimens needed for making the diagnosis can be obtained during the procedure. Curettage should be performed through a small oval corticotomy window to minimise the risk of iatrogenic fracture.

REFERENCES