J Pediatr Res 2023;10(2):151-5
DOI: 10.4274/jpr.galenos.2023.80148

Case Report

Garre's Osteomyelitis of the Jaw in a Child: Report of a Case Treated with Colchicine

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ABSTRACT

Garre's osteomyelitis is a rare chronic inflammatory disease with reactive peripheral bone formation due to low-grade local infection. Here, we present a 12-year-old female with chronic osteomyelitis and proliferative periostitis with no definite source of infection, such as caries or periodontitis. The patient had a history of 4-5 hospitalizations with the same symptoms intermittently over the previous two years at the hospital which referred her to our hospital. The patient had undergone a biopsy at the referring hospital, and she was referred to our hospital with a histopathological diagnosis of osteoid osteoma. Physical examination showed a unilateral swelling in the right mandible at admission to our hospital. Since we could not exclude the diagnosis of bacterial osteomyelitis, antibiotics were continued. Periapical radiography, magnetic resonance, computed tomography, and clinical features supported the diagnosis of Garre's osteomyelitis, so antimicrobial therapy was discontinued. Her biopsy materials were re-examined by the pathologist at our hospital, and Garre's osteomyelitis was considered. Non-steroidal anti-inflammatory drugs were started. We added colchicine treatment because she failed to achieve remission, and normal facial symmetrical morphology was not achieved in the two-month follow-up period. However, the symptoms regressed within one year, and the swelling disappeared.

Keywords: Garre's osteomyelitis, mandibular, children

Introduction

Chronic osteomyelitis with proliferative periostitis is a rare osteomyelitis characterized by periosteal reaction and new bone formation and it is traditionally known as Garre's osteomyelitis (1). The first cases affecting the jaw were reported in 1948 by Berger, and Pell described them in 1955. It generally originates from a low-virulence infection and it is characterized by chronic non-suppurative proliferative osteomyelitis associated with new bone formation (2).

Common sources of jaw infection include periapical periodontitis, periodontitis, fractures, and dental caries associated with non-odontogenic infections (3). Garre's osteomyelitis mainly occurs in young patients and often affects the mandibular trunk unilaterally (4,5). The premolar and molar regions of the mandible are most affected.

The lesion is characterized by local thickening of the periosteum with a reactive deposition of new cortical bone and periosteal osteoid. Clinically, this reactive process can be seen with a firm swelling on the jaw followed by facial

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Received: 19.12.2022 **Accepted:** 02.04.2023

asymmetry. Although its clinical symptoms are variable, the lesions are usually asymptomatic (6).

Imaging findings of the jaws in Garre's osteomyelitis are bone hyperplasia and destruction. Histological examination showing new bone formation under the periosteal layer is a characteristic disease feature. Extraction of the offending tooth, antibiotics, and non-steroidal anti-inflammatory drugs (NSAIDs) were reported as the most common treatment options (4,7).

Here, we present a case of chronic osteomyelitis with proliferative periostitis in a child.

Case Report

A 12-year-old female was referred to our department in February, 2021 due to persistent pain, jaw swelling, and difficulty in eating. The patient was previously healthy with no known systemic illnesses/allergies and had not received any medications apart from frequent antibiotics and NSAIDs for painful episodes of jaw swelling. No trauma/dental history was reported. There was no significant family history.

She had reported recurrent painful episodes with swelling in the right mandible over the prior two years. She had been hospitalized with similar symptoms 4-5 times in the past, and intravenous antibiotics had been given for suspected acute osteomyelitis. The ultrasonography performed in the hospital she was referred from showed soft tissue involvement, edematous, and hyperemia in the subcutaneous area and the right buccal region. Magnetic resonance imaging (MRI) showed expansion of the intramedullary bone, which extended slightly towards the body part of the bone, and bone marrow edema accompanied by cortical irregularities in the ramus of the mandible on the right and an inflammatory process surrounding the soft tissues. Intravenous ceftriaxone and clindamycin treatment were administered due to radiological findings indicating osteomyelitis. A biopsy was performed because of persistent symptoms despite receiving appropriate treatment for 15 days, and the histopathological diagnosis was reported as "osteoid osteoma" at the hospital which the patient was referred from.

On the first examination at the pediatric infectious diseases department of our hospital, the patient was afebrile, and neither tachycardic, nor tachypneic. Physical examination showed unilateral swelling on the right mandible, and facial asymmetry was noticed (Figure 1); however, no tenderness, erythema, or intraoral signs of gingival or periodontal infection were observed. The following were the results of the patient's laboratory tests: total leukocyte counts $9.09x10^3/\mu L$ (neutrophil 60%, lymphocyte 33%, monocyte 4.1%), hemoglobin 12.5 g/dL, platelet count: $284x10^3/\mu L$, C-reactive protein: 0.6 mg/L, lactate dehydrogenase: 171 U/L, erythrocyte sedimentation rate: 21 mm/hour, and her liver and kidney function tests were normal. Ampicillin/sulbactam treatment was also initiated due to the suspicion of bacterial osteomyelitis. The basic immunological tests and oxidative burst activity were normal. Viral markers showed past cytomegalovirus and rubella infections. Blood cultures were negative.

Periapical radiography demonstrated no findings of periapical periodontitis or fractures (Figure 2). The mandibular radiography (Figure 3) showed an "onion skin" appearance of the distended cortical plate buccal and inferior tooth. Computed tomography imaging showed bony expansion, diffuse sclerosis, cortical thinning, and periosteal reaction involving the right mandibular body, angle, and ramus (Figure 4). MRI revealed an abnormal bone marrow signal compatible with edema. Increased

Figure 1. Pre-treatment extra-oral photographs; (A) lateral view; (B) Frontal view; (C) 2 months later

signal on T2-weighted images and contrast enhancement on fat-saturated T1-weighted images were present in the surrounding soft tissue, indicating edema and inflammation (Figure 5). These imaging findings were highly suggestive of Garre's sclerosing osteomyelitis. Biopsy material testing performed in the previous hospital was re-evaluated by the pathologist in our hospital and it showed new bone formation with periosteal reactivity, a significant inflammatory process.

Regarding the histopathological and radiological findings, Garre's osteomyelitis was considered. Antibiotic therapy was discontinued. Non-steroidal anti-inflammatory treatment was started. The pain disappeared on the 16th day of hospitalization, and the patient was discharged. Since the swelling continued through to the follow-up two months later, colchicine was added. Six months after her discharge, the swelling had regressed by 60-70%. At the 1-year follow-up, the patient was asymptomatic with no

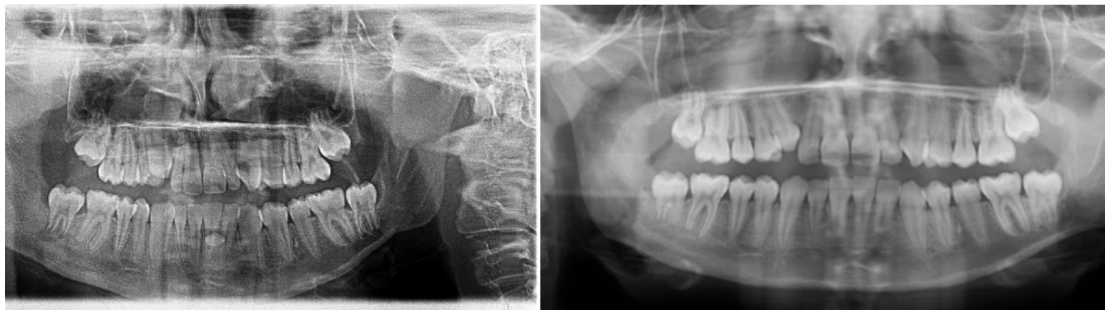

Figure 2. Panoramic radiograph showing the no findings of periapical periodontitis or fractures

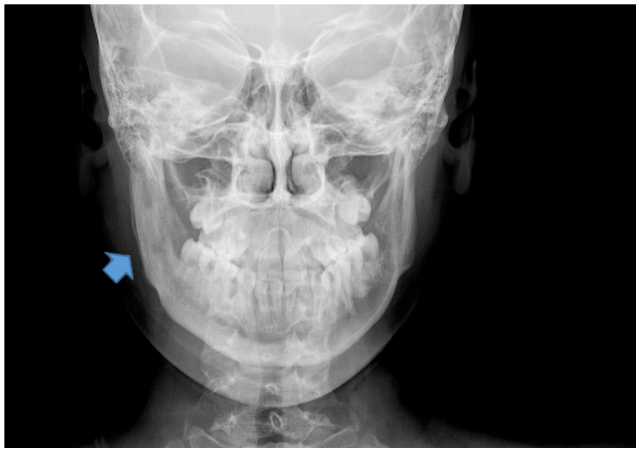

Figure 3. No findings of periapical periodontitis or fractures appearance of the distended cortical plate buccal and inferior tooth

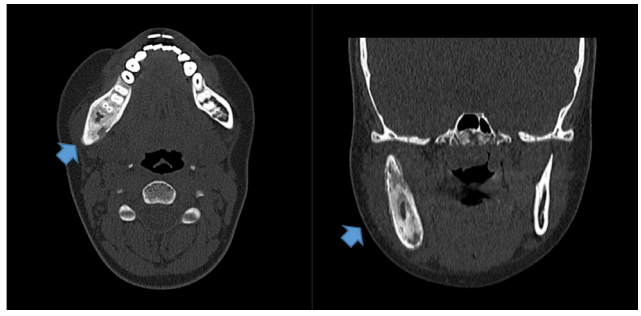

Figure 4. Axial (a) and coronal reformatted (b) CT images show bony expansion, cortical thinning, and periosteal reactions at the right mandibular body, angle, and ramus

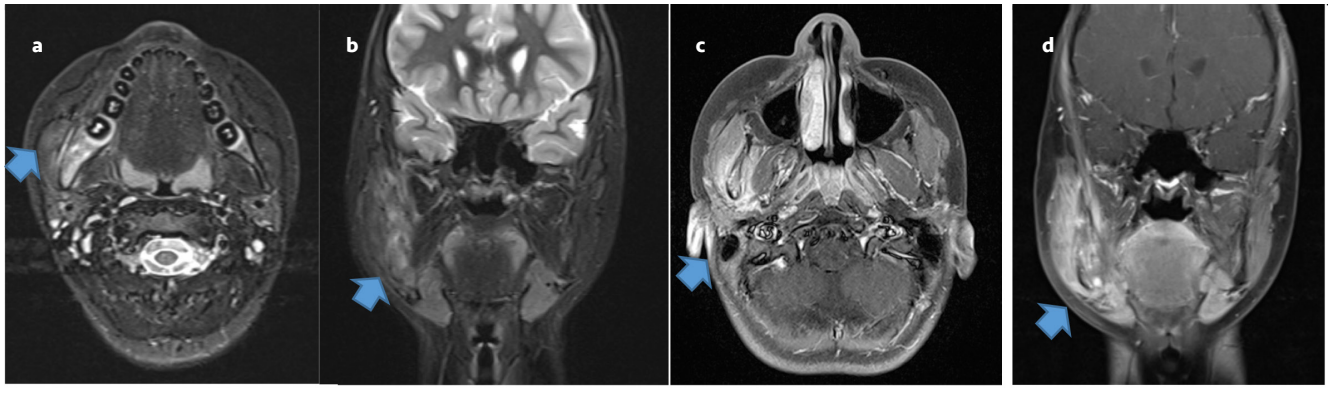

Figure 5. Fat-saturated T2-weighted axial (a), coronal (b) images and contrast-enhanced fat-saturated T1-weighted axial (c), coronal (d) images show bony expansion, bone marrow edema, and soft tissue inflammation adjacent to the lesion

palpable swelling. Written informed consent was obtained from the parents to publish their child's pictures.

Discussion

We report a case of Garre's osteomyelitis in a 12-year-old female whose age was in the range where this disease is common. Garre's osteomyelitis affects the mandible more than the maxilla and involved the unilateral mandible (5). Consistent with the literature, unilateral mandibular involvement was detected in our case.

Dental problems and inflammatory diseases have been primarily reported in the etiology. The pathogenesis of Garre's osteomyelitis has been suggested as being because the periosteum has more osteogenic potential during the growth phase of the jawbone in response to a stimulation of an inflammatory process in very young patients (especially <15 years). The severity and variability of symptoms are determined by various factors, including the organism's virulence, the host's immunity, and the person's underlying systemic condition (4,8). No periapical or periodontal pathology, history of trauma, or underlying systemic diseases were noticed in our case.

The clinical features are episodic non-progressive, insidious onset, and localized pain. Its duration is highly variable, lasting several months or several years. The lesions may be asymptomatic, persistent, or intermittent. In general, the function of the affected bone is preserved (9). In the present case, multiple episodes were reported with an adverse effect on the quality of life. There was a long-term history in our case. The function of the bone was preserved.

Radiologic studies in Garre's osteomyelitis have shown the presence of bony laminations, called "onion skin" appearance (10). In our case, radiological studies supported the diagnosis of Garre's osteomyelitis, showing bony expansion, diffuse sclerosis, cortical thinning, and periosteal reaction.

The diagnosis of Garre's osteomyelitis is usually based on clinical and radiological findings. However, a biopsy can be used for a definitive diagnosis when clinical and radiological findings are not pathognomonic. In our case, a biopsy was performed because of initial non-specific findings.

Treatment options include antibiotics, anti-inflammatory drugs, colchicine, steroids, conservative therapy, and surgery (4,7). Indications for antibiotic treatment include fever of more than 100 °F (37.8 °C), malaise, lymphadenopathy, trismus, cellulitis, and rapid soft tissue swelling associated with infections. Since the patient presented here was referred to our hospital with a preliminary diagnosis of osteomyelitis, antibiotics were administered until the diagnostic studies were completed. When Garre's osteomyelitis was diagnosed, this treatment was discontinued because the patient had no dental pathology, fever, cellulitis, or erythema. As the swelling did not regress with NSAID treatment, colchicine was added, and the patient recovered without surgery.

In conclusion, in this case, radiological findings and the recovery period of Garre's osteomyelitis, which is rare, are discussed. It should also be kept in mind that remission may occur with colchicine treatment without surgery when there is no response to NSAID treatment.

Ethics

Informed Consent: Written informed consent was obtained from the parents to publish their child's pictures.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: S.Y.A., Z.S.B., G.G.O., N.M.B., Z.K., M.S., A.C., M.O.Y., F.O., Concept: S.Y.A., Z.S.B., G.G.O., N.M.B., Z.K., M.S., A.C., M.O.Y., F.O., Design: S.Y.A., Z.S.B., G.G.O., N.M.B., Z.K., M.S., A.C., M.O.Y., F.O., Data Collection or Processing: S.Y.A., Z.S.B., G.G.O., N.M.B., Z.K., M.S., A.C., M.O.Y., F.O., Literature Search: S.Y.A., Z.S.B., G.G.O., N.M.B., Z.K., M.S., A.C., M.O.Y., F.O., Writing: S.Y.A., Z.S.B., G.G.O., N.M.B., Z.K., M.S., A.C., M.O.Y., F.O.

Conflict of Interest: The authors declared that there were no conflicts of interest.

Financial Disclosure: The authors declared that this study has received no financial support.

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