Journal of Dental Sciences (2015) 10, 450–455

Available online at www.sciencedirect.com

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CASE REPORT

Chronic osteomyelitis with proliferative periostitis in the lower jaw

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Final revision received 1 March 2011; accepted 18 March 2011
Available online 23 October 2012

KEYWORDS
chronic osteomyelitis;
computed tomography;
Garré's osteomyelitis;
proliferative periostitis

Abstract Chronic osteomyelitis with proliferative periostitis (Garré's sclerosing osteomyelitis) is a distinctive type of chronic osteomyelitis that mainly affects children and young adults. Here we report on a 9-year-old girl in whom the condition arose following a pulpoperiapical infection in a mandibular right primary secondary molar. Clinically, it manifested as a bony, hard, mildly tender swelling. Radiography revealed a pathognomonic patchy thickening with radiolucency and radiopacity. The dental inflammation and infection were eliminated and conservative therapy followed. The patient was otherwise asymptomatic. Remission of the disease process and reappearance of a normal-looking mandible was observed with computed tomography imaging, three-dimensional reconstruction and a bone scan at a 10-month follow up visit.

Introduction

Garré's sclerosing osteomyelitis was first described by Carl Garré in 1893 as irritation-induced focal thickening of the periosteum and cortical bone of the tibia.[1] It is a type of chronic osteomyelitis that primarily affects children and adolescents.[2] In the orthopedic literature, chronic osteomyelitis with proliferative periostitis (COPP) of the tibia is a well-known syndrome. Berger described the first cases of proliferative periostitis affecting the jaw bones. Reports of COPP in the body of the mandible are relatively common.[3] Most reported cases are unilateral, but one case involving all four quadrants of the jaw has been reported.

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1991-7902/$36
http://dx.doi.org/10.1016/j.jds.2012.09.020

COPP is a more accurate description of the pathogenesis of this condition than Garré's osteomyelitis, as mild irritation and infection affect the jaw and lead to peripheral subperiosteal bone deposition.[4] This process, related to periosteal osteoblastic activity formation of subperiosteal bone, represents a periosteal reaction to inflammation. The histopathology of COPP is actually distinctive. The affected periosteum forms several layers of vital bone that are parallel to each other and to the surface of the affected bone. An intact cortex is present below the new bone formation.

COPP commonly occurs in young patients with a mean age of 13 years. Sporadic cases have been reported in patients in their 20s and in infants as young as 2 years. COPP is a non-suppurative inflammatory process. The most common provoking factors in the jaw region are a previous tooth extraction, tooth eruption, dental caries with associated periapical inflammation, periodontal infections, fractures and non-odontogenic infections. Most cases arise in the molar/premolar area of the mandible. Clinically, patients may present with a hard swelling of the jaw and facial asymmetry caused by this reactive process.

Radiographic examinations typically show bony laminations parallel to each other and to the cortical surface of the involved bone. Areas of small sequestra or osteolytic radiolucencies can be found within the new bone. Appropriate radiographic angulation can highlight a radiolucent zone of soft tissue between the original bony cortex and the newly-formed reactive bone. COPP presents a special radiographic feature, especially evident in computed tomography (CT) with three-dimensional reconstruction, with new periosteal proliferation located in successive layers parallel to the condensed cortical bone.[5] This phenomenon is also correlated with typical radiographic features such as an "onion skin" appearance. Other lesions that must be considered in differential diagnosis of COPP are Ewing's sarcoma, fibrous dysplasia, osteogenic sarcoma, infantile cortical hyperostosis, callus, exostosis, calcifying hematoma, and osteotomas.

In this case, a 9-year-old girl was evaluated based on her clinical, radiographic and histopathological features. Clearly, in lesions of this type, histopathology plays a major role in determining the final diagnosis and subsequent course of treatment.

Case report

A 9-year-old girl presented with extra-oral swelling in the right inferior border of the mandible. Extra-oral examination of the right side revealed a diffuse, mildly tender swelling, which was hard in consistency with no lymphadenopathy. The skin color was normal. An intraoral photograph showed a postextraction (85) wound in relation to area 44–46 (extracted by a general practitioner 20 days previously). Periodontal probing around the tooth revealed no deep pocketing. There was a functional opposing tooth. The patient's history revealed she had been seen by several practitioners and had been treated without resolution for an assumed dentoalveolar abscess with various courses of antibiotics.

At the patient's first presentation at our outpatient department, plain radiographs (Fig. 1A) and CT scans demonstrated enlargement of the affected mandible. They showed patchy areas of sclerosis and relative radiolucencies together with thickening of the overlying soft tissues (Fig. 1B and C). Increased tracer uptake in the region of the right mandible was evident on a bone scan; there was no significant scintigraphic activity except for the epiphyseal plates (Fig. 2A). A full blood count, serum levels of urea, electrolytes, calcium, and phosphate were within normal limits. A raised erythrocyte sedimentation rate (24 mm/h) and alkaline phosphatase (221 U/L) were consistent with the clinical examination. We made a provisional diagnosis of neoplastic bone, with inflammatory-infectious and fibro-osseous changes to the mandible.

Histopathology of a bone biopsy examination revealed a chronic nonspecific inflammatory lesion without any evidence of granulomatous inflammation; a microbiological culture was also negative. The biopsy exhibited trabeculae of bone with osteoblastic rimming and reversal lines (Fig. 2B). Based on the patient's clinical presentation, radiographic characteristics and histopathology, we diagnosed COPP. We administered a non-steroidal anti-inflammatory drug and the symptoms and signs subsided in 7 days. We closely followed this patient and at a 10-month follow-up visit, we found the patient to be asymptomatic with no signs (Fig. 3A and B). Occlusal, panoramic radiographs (Fig. 3C) and CT scans showed remodeling of the jaw (Fig. 4A). The previous increased tracer uptake in the region of the right mandible had returned to normal on a bone scan (Fig. 4B).

Discussion

The patient's symptoms and radiographic and clinical features were consistent with several different disease categories, including neoplastic, fibro-osseous and inflammatory-infectious diseases.

The lack of fever, limited response to antibiotic therapy, enlargement of the lesion, lack of suppuration and radiographic features of a loss of the mandibular border were consistent with a malignancy. The possible specific neoplasms included mesenchymal tumors such as an osteosarcoma, a chondrosarcoma, and Ewing's sarcoma. This variation in radiographic presentation is caused by the varied forms of osteosarcoma (lytic, sclerosing, or mixed).[6] Osteolytic mandibular lesions are typically bulky, with irregular radiolucency including both expansion and destruction of the cortical plate. For chondrosarcomas, a "sun-ray" and "onion skin" appearance have also been described.[7,8] For Ewing's sarcoma, onion skinning is also a typical radiographic feature.[9] However, the histopathology in this case revealed a chronic nonspecific inflammatory lesion without malignant changes (Fig. 2B).

A fibrous dysplasia needed to be considered because this type of lesion is frequently seen in the craniomaxillofacial region as a painful enlargement of the periosteal bone, and its characteristic radiographic features could be consistent with those seen in this patient. Although more frequently found in the maxilla, fibrous dysplasia can occur in the mandible of young patients as a focal enlargement.[10]