

Proliferative periostitis of the mandibular ramus and condyle: a case report

Hyun Seok¹, Seong-Gon Kim¹, Ji-Young Song²

¹Department of Oral and Maxillofacial Surgery, College of Dentistry, Gangneung-Wonju National University, Gangneung,

²Department of Oral and Maxillofacial Surgery, Jeju National University Hospital, Jeju National University School of Medicine, Jeju, Korea

Abstract (J Korean Assoc Oral Maxillofac Surg 2015;41:198-202)

Proliferative periostitis is a rare form of osteomyelitis that is characterized by new bone formation with periosteal reaction common causes of proliferative periostitis are dental caries, periodontitis, cysts, and trauma. While proliferative periostitis typically presents as a localized lesion, in this study, we describe an extensive form of proliferative periostitis involving the whole mandibular ramus and condyle. Because the radiographic findings were similar to osteogenic sarcoma, an accurate differential diagnosis was important for proper treatment.

Key words: Periostitis, Osteomyelitis, Mandibular condyle

[paper submitted 2014. 11. 18 / revised 2015. 4. 8 / accepted 2015. 5. 3]

I. Introduction

Proliferative reactions of the periosteum have been described using various terms, including Garre's osteomyelitis, proliferative periostitis of Garre, periostitis ossificans, osteomyelitis with proliferative periostitis, nonsuppurative ossifying periostitis, osteomyelitis sicca, and perimandibular ossification¹. Proliferative periostitis is a rare disease, and represents new bone formation with periosteal reaction. The common causes of periosteal reaction are dental caries with associated periapical lesions and secondary periodontal infections, fracture, and nonodontogenic infections². The most prevalent areas are the premolar and molar areas of the mandible³. Radiographically, radiopaque lamination is observed parallel to the surface of the cortical bone⁴. The number of

laminations varies from 1 to 12, and radiolucent separation is seen between the newly formed bone and the original cortical bone². Less frequently, osteolytic radiolucency and sequestra are observed in the newly formed bone². Most lesions are localized around the buccal cortex of the teeth which the periostitis originates³⁻⁶.

The most common treatment for this disease involves eliminating the origin of infection. Extraction and endodontic therapy around the involved tooth have been described as successful methods to treat the odontogenic origin of proliferative periostitis^{5,7}. The newly formed bone is consolidated within 6 to 12 months after eliminating the origin of infection and is remodeled by overlying muscle². A broad differential diagnosis is necessary for determining the treatment plan and may include fibrous dysplasia, Ewing's sarcoma, osteogenic sarcoma, ossifying hematoma, exostosis, and osteoma⁸.

We present a case of extensive proliferating periostitis that resembled osteogenic sarcoma.

II. Case Report

An 11-year-old girl was referred from a local clinic for swelling of the left mandibular area that began one month prior.(Fig. 1) She presented with trismus and facial asymmetry due to swelling of the left mandibular angle and condyle

Ji-Young Song

Department of Oral and Maxillofacial Surgery, Jeju National University Hospital, Jeju National University School of Medicine, 15 Aran 13-gil, Jeju 63241, Korea

TEL: +82-64-717-1845 FAX: +82-64-717-1097

E-mail: 2460song@naver.com

ORCID: <http://orcid.org/0000-0002-9500-6557>

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This work was supported by a grant from the Next-Generation BioGreen21 Program (No.PJ01121404), Rural Development Administration, Republic of Korea.

area; however, there was no tenderness, fever, or redness. She had no history of trauma to the left facial area. On intra-oral examination, there were no dental caries or periodontal lesions. However, there was mild swelling and tenderness of the distal gingiva on the left mandibular second molar. The



Fig. 1. Clinical photograph shows the swelling of the left mandibular ramus and angle.

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third molar was impacted.

A panoramic view showed the unerupted third molar on the left mandible and a radiolucent appearance of the ascending ramus. A sclerotic appearance was observed over the left mandibular body, ramus, and condyle. Because of the bony sclerotic change, the cortical bone of the left mandibular canal was faint compared to the corresponding area on the right side.(Fig. 2) This feature can be observed in detail on the computed tomography (CT) image.(Fig. 3) There was an osteolytic and sclerotic bone lesions in the left mandibular



Fig. 2. Panoramic radiographs show the radiolucent appearance around the unerupted third molar on the left mandible.

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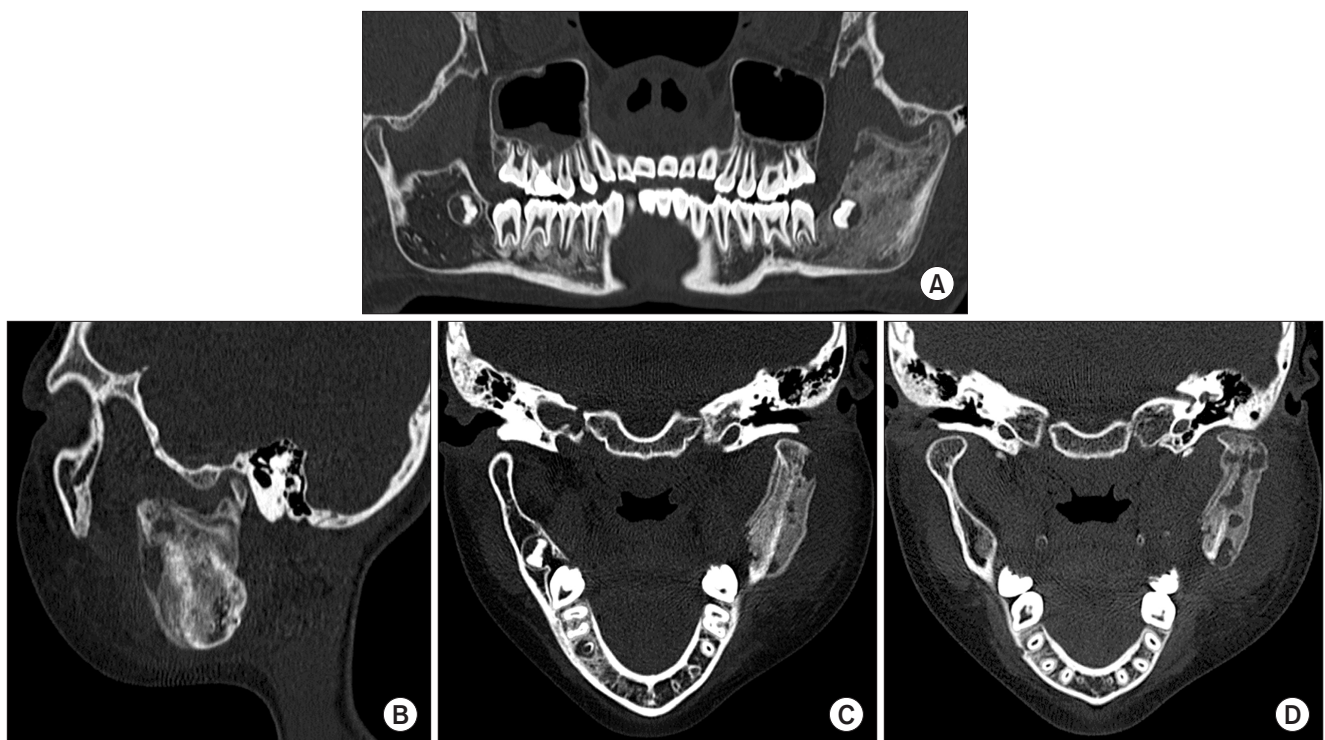


Fig. 3. Computed tomography image. Osteolytic and sclerotic bone lesion. A. The ramus and condyle. B. Mandibular angle. C. Periosteal reaction in the left mandibular ramus and condyle. D. Osteolytic change inside the newly formed bone.

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body, ramus, and condyle area.(Fig. 3. A, 3. B) New bone formation with periosteal reactions was observed in the medial and lateral portion of the condyle.(Fig. 3. C) It was more dominant in the lateral portion. Osteolytic change was also observed inside the newly formed bone.(Fig. 3. D) The left masseter muscle showed swelling and contrast enhancement. On laboratory testing, the C-reactive protein (CRP) level was 2.0 mg/dL, white blood cells (WBC) count was $11.2 \times 10^3/\text{mm}^3$ and erythrocyte sedimentation rate (ESR) was 104 mm/hr. These values were greater than the normal ranges.

From these clinical and radiological features, the initial diagnosis was osteogenic sarcoma with an initial treatment plan of hemimandibulectomy with costochondral graft. To obtain a proper diagnosis, we performed an incisional biopsy on the left mandible area under local anesthesia. Histologically, acute and chronic inflammation with reactive new bone formation was observed. Based on the pathological findings, the initial diagnosis was revised to an extensive form of proliferative periostitis.

Under general anesthesia, a vestibular incision was made starting from the left mandible outer oblique ridge area. Subperiosteal dissection was performed inferiorly and posteriorly to expose the mandibular angle. After adequate exposure of the coronoid process, a coronoidectomy was performed to properly approach to the condylar area. The newly formed bone overlying the cortical bone of the ramus was identified, and decortication was performed. The impacted third molar was extracted, and the removed specimens were sent for biopsy.

Histological examination showed new trabecular bone

formation with osteoblastic rimming. The individual bony trabeculae of the reactive woven bone were arranged perpendicular to the bone surface.(Fig. 4. A) The intertrabecular space was occupied by a dense fibrous stroma with chronic inflammatory infiltrates and necrosis.(Fig. 4. B) From these features, the mandibular lesion was diagnosed as osteomyelitis with proliferative periostitis.

Amoxicillin was prescribed following surgery. Ten days after surgery, the patient did not feel any particular discomfort, and the surgical wound showed was in a good healing state. The trismus also showed improvement. The laboratory tests returned to normal ranges. The CRP level was 0.13 mg/dL, WBC count was $6.7 \times 10^3/\text{mm}^3$, and ESR was 39 mm/hr. One month after surgery, the left mandibular angle area showed normal facial contour, and the trismus had disappeared. At 11-month follow-up, there was no sign of recurrence or facial contour abnormality.(Fig. 5. A) On CT imaging, the newly formed bone and sclerotic change of the mandibular ramus and condyle area had disappeared.(Fig. 5. B, 5. C) Additionally, the extraction socket of left mandibular third molar completely healed.(Fig. 5. D)

She agreed to have her clinical photographs being published on this paper.

III. Discussion

Proliferative periostitis is a form of chronic osteomyelitis that involves new periosteal bone formation over the cortical bone. The most common cause of proliferative periostitis is odontogenic infection². Most proliferative periostitis occurs

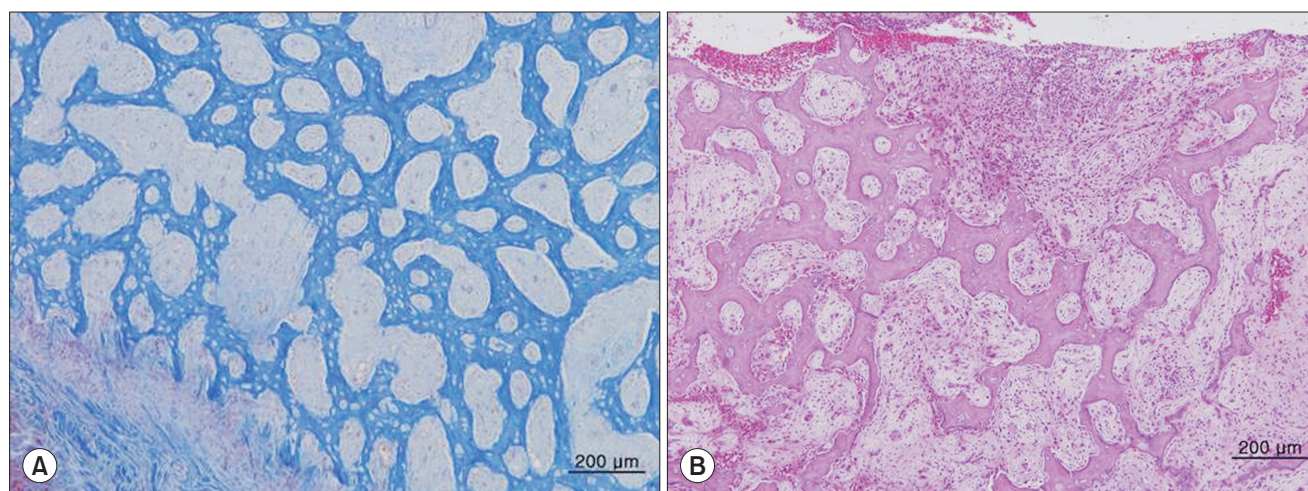


Fig. 4. Histological examination. A. Bony trabeculae of the reactive bone arranged perpendicular to the bone surface (Masson's trichrome staining, $\times 100$). B. Dense fibrous stroma with chronic inflammation in the intertrabecular space (H&E staining, $\times 100$).

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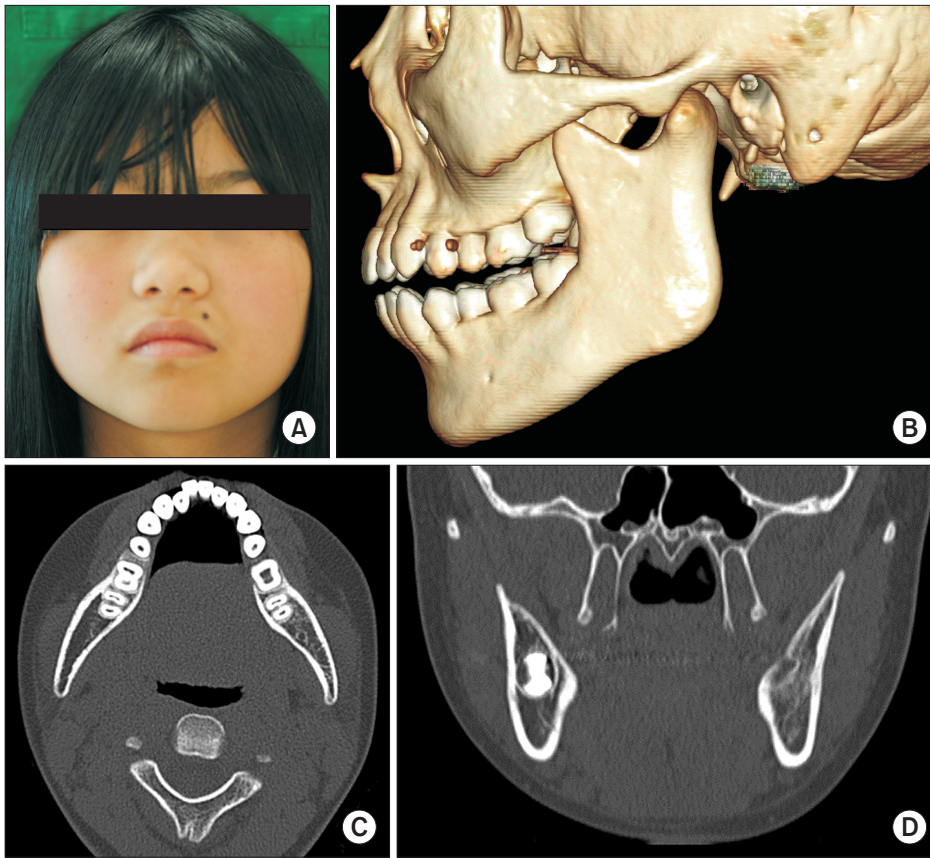


Fig. 5. At 11-months follow-up. A. Clinical photograph shows symmetrical face contour. B, C. Disappearing of newly formed bone and sclerotic change on mandibular ramus and condyle. D. Extraction socket of the third molar completely healing.

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over the cortical bone and the area of the involved tooth but in a localized form³. Some reports used only occlusal radiography to make a diagnosis of this disease without biopsy or further radiological examination^{3,4}. A case of extensive bone formation over the involved area has never been reported. In this case, periosteal bone formation extended beyond the involved areas into the left mandibular ramus and condyle.

Usually, proliferative periostitis originates from a periapical lesion associated with dental caries or periodontal infection³. Rarely, proliferative periostitis has been reported to occur due to an unerupted third molar⁶. According to Neville et al.², periosteal hyperplasia can be caused by an unerupted tooth near the lesion. In this patient, the developing third molar was suspected to be the source of infection, and proliferative periostitis was observed adjacent to the unerupted third molar.(Fig. 2)

In the CT image, osteolytic change was observed inside the newly formed bone.(Fig. 3) Generally, proliferative bone forms layers of radiopaque lamination, like “onion skin”. Cortical bone lamination should be arranged in parallel layers³. Less frequently, osteolytic radiolucencies are found in the new bone⁷. In our patient, the outer surface of the newly formed bone was smooth and continuous (Fig. 3. C);

however, a bony, destructive and sclerotic appearance was observed inside the new bone, which was dominant on the lateral portion.(Fig. 3. D) This bony change also appears in Ewing’s sarcoma and osteogenic sarcoma⁸, but the bony destruction involved in those diseases is more irregular and has a moth-eaten appearance^{2,8}. Additionally, the periosteal reaction is typically seen as a sun burst or sun ray appearance as opposed to lamination of cortical bone^{2,8}.

Histologically, parallel rows of trabeculae in reactive, woven bone formed perpendicularly to the cortical bone surface, and inflammation and necrosis were observed in the intertrabecular space. In proliferating periostitis, the individual trabecular bone is sometimes interconnected and widely distributed², which is similar to the pattern of immature fibrous dysplasia⁹. However, in fibrous dysplasia, the trabeculae of woven bone are irregularly shaped, and a fibrous stroma is seen. Furthermore, in osteogenic sarcoma, anaplastic tumor cells and osteoid produced by malignant mesenchymal cells can be observed². However, there was no osteoid production or tumor cells in this case.

Treatment options for proliferative periostitis typically include removal of the infected tooth or endodontic treatment^{3,4,6}. After eliminating the source of the infection, the

enlarged bone is remodeled back to its original form by the action of overlying muscle². If the osseous enlargement is too severe and remodeling to the original form is unlikely, surgical intervention is needed to contour the enlarged bone¹⁰. In this case, the source of infection was removed by extraction of the left mandibular third molar, and an outer decortication was performed to remove sclerotic bone and for aesthetic reasons. The patient's face recovered, and regained normal symmetric contours one month after surgery. Eleven months after surgery, new bone formation on the ramus and condyle completely disappeared.(Fig. 5) Although the sclerotic change extended broadly from the mandibular ramus to the condyle, we could easily treat this case by simple tooth extraction and decortication without radical resection.

In conclusion, this case demonstrates that proliferative periostitis can occur due to infection around an unerupted tooth. In this patient, the periosteal reaction was seen extensively from the mandibular ramus to the condyle. The sclerotic and osteolytic bony change presented inside the newly formed bone. We used CT imaging and incisional biopsy to make an initial diagnosis. Based on these results, we performed extraction of the unerupted tooth and subsequent decortication.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

ORCID

Hyun Seok, <http://orcid.org/0000-0002-5278-3807>

Seong-Gon Kim, <http://orcid.org/0000-0001-5088-2732>

Ji-Young Song, <http://orcid.org/0000-0002-9500-6557>

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