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Case report

Osseous choristoma of the buccal mucosa: A case report with immunohistochemical study of bone morphogenetic protein-2 and -4, and a review of the literature

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ABSTRACT

Osseous choristomas are benign lesions that are characterized by ectopic bone formation in the soft tissue of the head and neck region. However, the pathophysiological mechanism responsible for ossification remains unclear. In this paper, we report a case of an osseous choristoma arising in the buccal mucosa and present a review of the relevant literature. We also carried out an immunohistochemical study of bone morphogenetic protein (BMP)-2 and -4, the potent inducers of ectopic bone formation, to identify the characteristics of bone-forming cells in the lesion. An 86-year-old woman was referred to our department because of an asymptomatic mass in the buccal mucosa. The lesion was 7 mm in size, and was excised under general anesthesia. The postoperative course was uneventful, with no signs of recurrence during the 24 months after surgery. Histologically, the lesion contained a lobular-shaped osseous tissue with a fibrous capsule, and the pathological diagnosis of osseous choristoma was made in consideration of the ectopic bony tissues that were localized away from the maxillo-mandibular bone. Immunohistochemical examination showed the first evidence of expression of BMP-2 and -4 in the osseous choristoma. BMPs were expressed in the osteoblasts surrounding the ectopic bone. Our results indicated that BMP-2 and -4 was associated with the ossification of the osseous choristomas.

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1. Introduction

Osseous choristomas are rare benign lesions that are characterized by ectopic bone formation in the soft tissue of the head and neck region [1,2]. The lesions occur almost exclusively in the tongue, but they have been occasionally reported in the buccal mucosa [3–13]. However, the pathophysiological mechanism responsible for the ossification in soft tissue remains unclear [1,2]. In this paper, we report a case of an osseous choristoma arising in the buccal mucosa and present a review of the relevant literature. To identify the characteristics of bone-forming cells in the lesion, we also carried out an immunohistochemical study of bone

morphogenetic protein (BMP)-2 and -4, the potent inducers of ectopic bone formation [14]. We present the results and discuss the mechanism in relation to the BMP expression.

2. Case report

An 86-year-old Japanese woman was referred to the Department of Dentistry and Oral Surgery, University of Fukui Hospital, for an evaluation of a mass in the buccal mucosa in November 2010. The patient's medical history revealed the presence of hypertension, diabetes mellitus and aortic regurgitation; however, there were no conditions that were relevant to the oral complaint. Physical examination revealed a non-tender mass covered with normal mucosa in the left buccal region (Fig. 1). The lesion was approximately 7 mm in diameter and had developed over a period of 3 years. The mass was elastic and was not very hard on palpation. The mass was movable, and there was no evidence of adhesion to the surrounding tissues. The patient wore a denture and had no history of inflammation or trauma in the left buccal region. The movement of the lips and cheeks was normal, and sensory disturbance was not evident. Significant swelling of the cervical lymph nodes was not found. The laboratory examination revealed that

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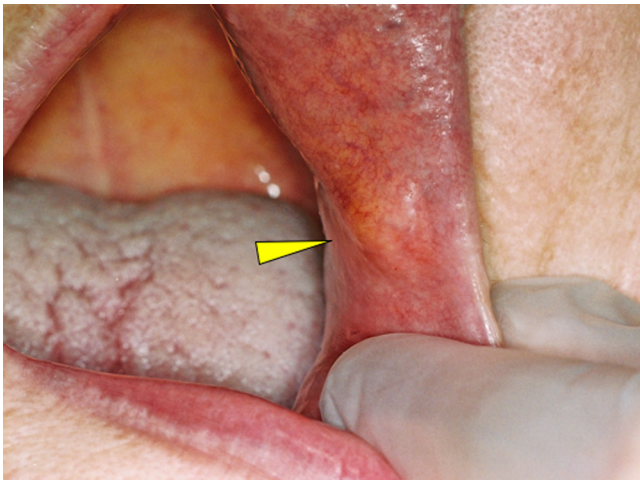


Fig. 1. Clinical appearance. An intraoral view showing a slight swelling of the left buccal mucosa (arrowhead). The sessile mass was covered with normal mucosa.

the serum calcium value was within the normal range. Magnetic resonance imaging (MRI) revealed a 7 mm, well-circumscribed lobular mass in the buccal region (Fig. 2A–C). The mass showed heterogeneous, low signal intensity on T1-weighted images, with heterogeneous enhancement after an intravenous administration of gadolinium, and heterogeneous, intermediate signal intensity on T2-weighted images. The clinical diagnosis was a benign tumor in the left buccal mucosa. The lesion was excised totally with the surrounding normal tissue under general anesthesia. Histologically, the mass consisted of well-circumscribed loose cancellous bone, and it was surrounded by a layer of collagenous fibrous connective tissue (Fig. 3A). Adipose tissue was observed adjacent to the mass. Osteoblasts and osteocytes were found on the bone surface and in the bone lacunae, and a few osteoclasts were observed in the Howship's lacunae (Fig. 3B). There was no evidence of inflammation or malignancy in the specimen. The histological diagnosis of an osseous choristoma was made in consideration of the ectopic bony tissues that were localized away from the maxillo-mandibular bone [3].

We performed an immunohistochemical examination of BMP-2 and -4. Three-micrometer sections of formalin-fixed, decalcified specimen were prepared, and the sections were deparaffinized, rehydrated and microwaved in citrate buffer for antigen retrieval. The sections were preincubated with a blocking reagent (including 3% hydrogen peroxide to block endogenous peroxidase activity). Then, the sections were incubated with goat anti-human BMP-2 polyclonal antibody (N-16; Santa Cruz Biotechnology, Inc., Santa Cruz, CA, USA; dilution, 1:50) and with goat anti-human

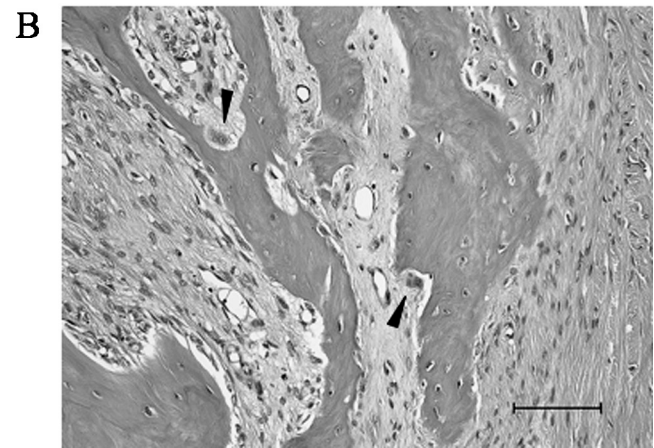
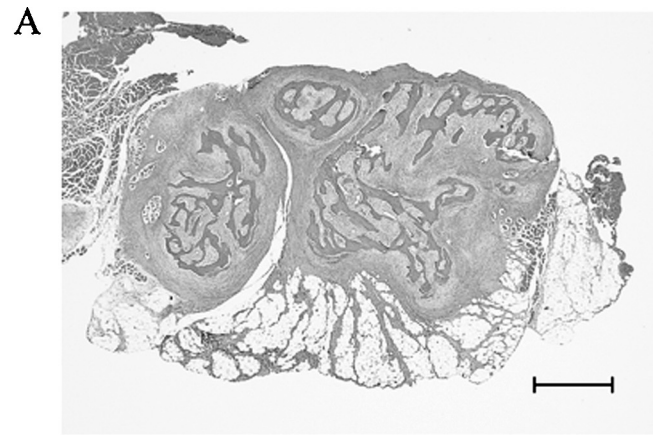


Fig. 3. Microphotograph. (A) The mass consisted of well-circumscribed loose cancellous bone and was surrounded by a layer of collagenous fibrous connective tissue (hematoxylin and eosin, scale bar 1000 μm). (B) Osteoblasts were found on the bone surface, and osteocytes were found in the bone lacunae. A few osteoclasts were observed in the Howship's lacunae (arrowheads) (hematoxylin and eosin, scale bar 100 μm).

BMP-4 polyclonal antibody (N-14; Santa Cruz Biotechnology; dilution, 1:50) as the primary antibodies. The antigen antibody complex was reacted with streptavidin–biotin–peroxidase complex (Histofine® Simple Stain MAX-PO (G); Nichirei, Tokyo, Japan) and visualized with 3,3'-diaminobenzidine (DAB) solution. The sections were counterstained with Meyer's hematoxylin. The sections exposed to normal goat serum were used as negative controls, and none of these sections showed staining. A section of peripheral cemento-ossifying fibroma was used as a positive control [15],

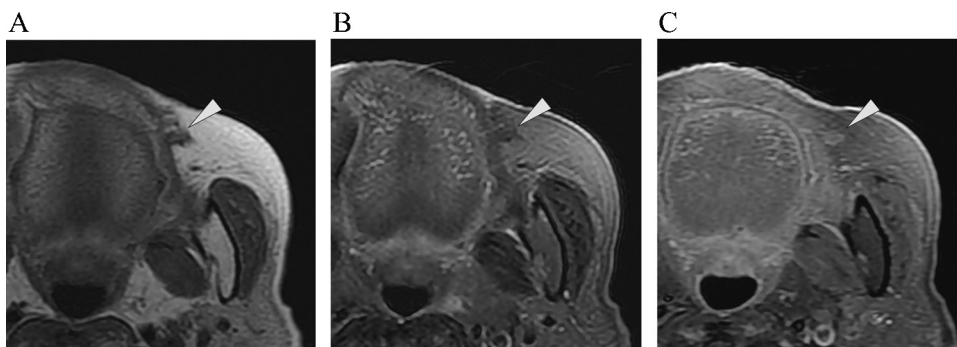


Fig. 2. MRI findings. The axial MR image showing a well-circumscribed lobular mass of 7 mm in diameter in the buccal region (arrowhead). (A) The lesion showed heterogeneous, low signal intensity on T1-weighted images. (B) The lesion showed heterogeneous, intermediate signal intensity on T2-weighted images. (C) The lesion showed heterogeneously enhanced signal intensity after an intravenous administration of gadolinium on T1-weighted images.

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