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Case Report

Intravascular papillary endothelial hyperplasia associated with hemangioma of the mandible: A rare case report

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ABSTRACT

Intravascular papillary endothelial hyperplasia (IPEH) is unusual, benign lesion of vascular origin caused by exuberation of endothelial cell proliferation. It is rarely seen in the oral region. Previous studies on oral IPEH revealed that the lower lip is the most frequent site, followed by tongue, upper lip, buccal mucosa and mandibular vestibule.

We report a case of IPEH associated with hemangioma of the mandible of a 75-year-old male. Embolization of the inferior alveolar artery was performed in this case and the lesion was removed under general anesthesia the day after embolization.

Histopathologically, intravascular proliferation of papillary processes within hemangioma was found. Immunohistochemically, the endothelial cells were positive for CD34. In this case, IPEH was observed in preexisting varices, hemangiomas, pyogenic granulomas, or lymphangiomas, known as a secondary (mixed) form.

Continuous and careful observation is needed, because recurrence of the mixed form has been mostly reported due to incomplete excision or regrowth of the underlying lesion.

1. Introduction

Intravascular papillary endothelial hyperplasia (IPEH) is unusual, benign, non-neoplastic, vascular lesion caused by the exuberation of endothelial cell proliferation. It was first described by Masson in 1923 as a 'vegetant intravascular hemangioendothelioma' [1]. The more descriptive term, "intravascular papillary endothelial proliferation", which was more frequently used in the English literature, was coined by Clearkin and Enzinger in 1976 [2]. It is thought that this lesion occurs in any blood vessel in the body but especially has a propensity for the skin and subcutaneous tissues of the head and neck region, extremities, fingers and trunk. This lesion is thought to be caused by trauma or stimulation. And this lesion seems to grow asymptomatically and slowly by discontinuous stimulation such as chewing, clenching, or suction. We report a case of intravascular papillary endothelial hyperplasia associated with hemangioma of the mandible, which was preoperatively evaluated the extent and property of the lesion carefully. Then, the embolization of the inferior alveolar artery was performed and the entirety of lesion was removed successfully.

2. Report of a case

The patient was a 75-year-old male. Early in March 2013, he felt pain in the left side of the mandible and consulted a family dentist. The panoramic radiograph showed the radiolucency from the right to left side molar region of the mandible. In mid-March 2013, he was referred to our department desiring a more intensive examination and treatment of radiolucent lesion of the mandible as a chief complaint. The intraoral findings showed the slightly bony hard swelling from the number 32 to 44 tooth region and was covered with normal mucosa (Fig. 1). Panoramic radiograph showed a multilocular radiolucent image from the number 34 to

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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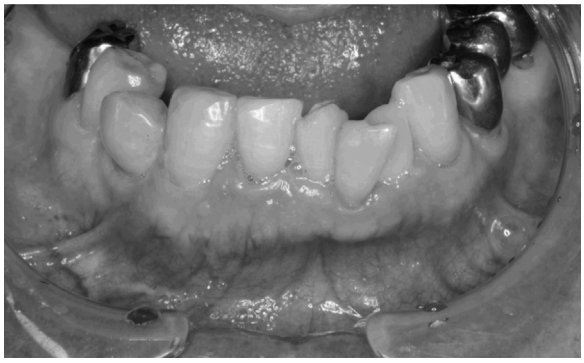

Fig. 1. Photograph of the intraoral findings at first clinical presentation showing the slightly bony hard swelling from the number 32 to 44 tooth region.

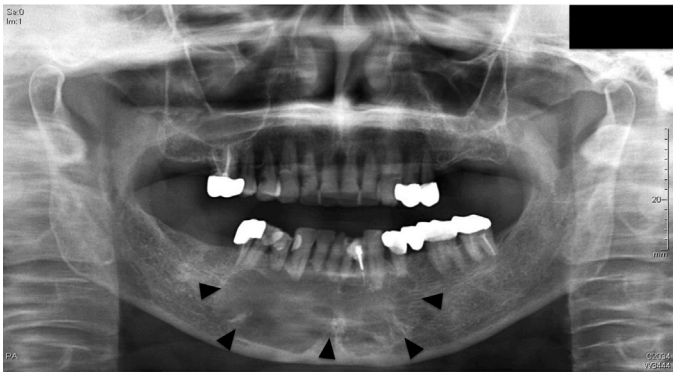

Fig. 2. Panoramic radiograph examination showing multilocular radiolucent image (arrowhead) from the number 34 to 44 tooth region. Note the partially honeycombed appearance.

44 tooth region and a partially honeycombed appearance (Fig. 2), while axial computed tomographic (CT) examination revealed the destruction of the labial cortical bone and intraosseous bone of the mandible (Fig. 3). In addition, partial bone defect of the labial side was found. We performed a tissue biopsy for definite diagnosis. Because of persistent bleeding at biopsy, we discontinued the

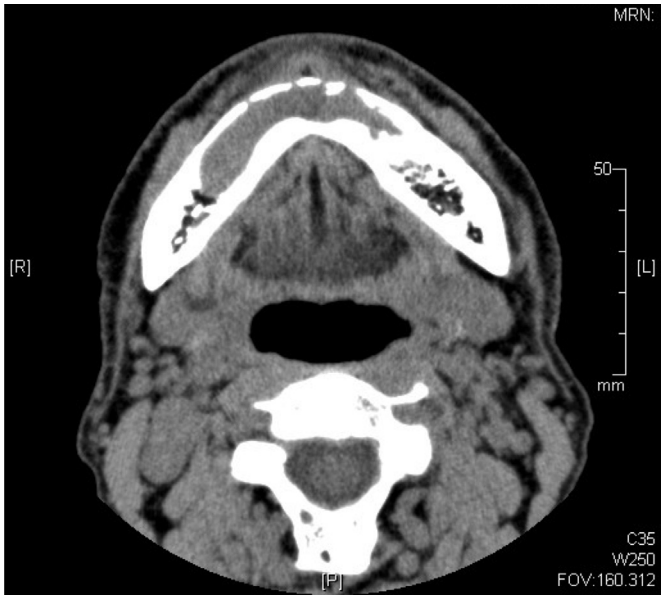

Fig. 3. Axial CT of the mandible showing intraosseous lesion from the number 35 to 46 tooth region, demonstrating destruction of the labial cortical bone and partial extraskeletal extension of intraosseous lesion.

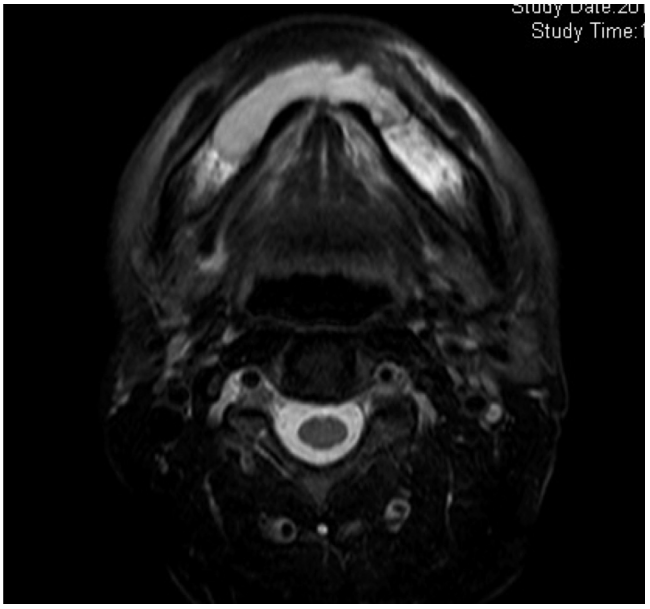

Fig. 4. T2-weighted MRI showed intraosseous lesion of the mandible: low flow pooling of blood was observed in the same site.

procedure. Therefore, Magnetic Resonance Imaging (MRI) was carried out. T2-weighted imaging also showed an intraosseous lesion, and low flow pooling of blood was observed at the same site (Fig. 4). Because a vascular lesion was suspected, pool scintigraphy was performed. Radioactive substance was accumulated in the mandible at the site of the intraosseous lesion which was observed by CT scan (Fig. 5). To screen for the presence of artery flow, right side transfemoral angiography of both sides of the external carotid artery was performed. There was no artery blood flow into the intraosseous lesion. Based on these examination findings, we suspected a clinical diagnosis of venous malformation of the mandible. As the

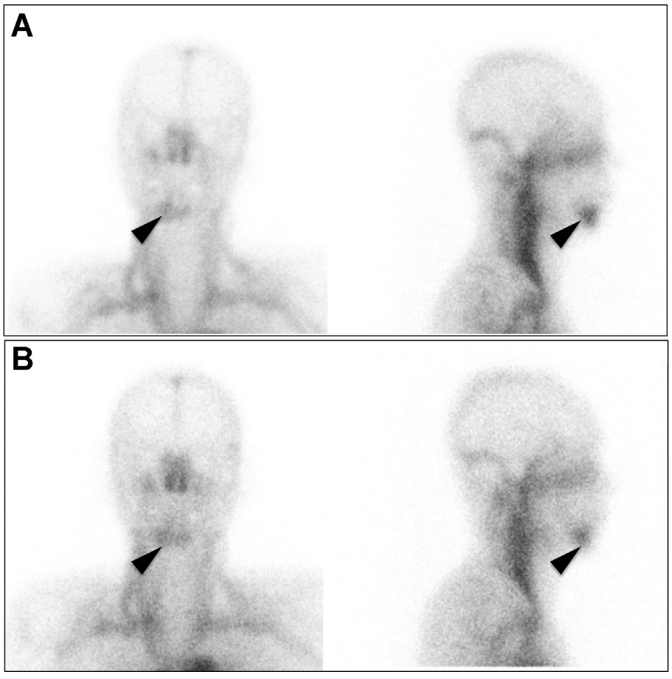

Fig. 5. (A) Pool schintigraphic images at 30 min. (B) Schintigraphic images at 4 h. Radioactive accumulation was confirmed in the mandible at the site of intraosseous lesion (arrowhead) which was observed by CT scan.