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Case report

Mandibular simple bone cyst in a patient with first and second branchial arch syndrome

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ABSTRACT

Simple bone cysts are known as pseudocysts and are thought to arise due to growth or developmental abnormalities, but few reports have been published on this phenomenon. We here report a rare case of a left-sided simple bone cyst in a patient with first and second branchial arch syndrome. This 13-year-old girl had regularly visited the orthodontic department for follow up of a jaw deformity and malocclusion since the age of 4 years. She reported to our department for examination of a radiolucent area in the computed tomography images of the middle to left side of the mandible, which had been found incidentally during orthodontic follow-up. Her facial features were asymmetric due to hypoplasia of the jaw. There was a bone-like bulge, without pain, at the labial gingiva around the left lower canine. She had not experienced trauma. We diagnosed this as a simple bone cyst after biopsy. Under general anesthesia, we performed curettage and promoted organizing. At 6 months after the operation, the radiolucent area had been partially replaced with new bone. This is a rare case indicating that a simple bone cyst can arise from a hypoplastic area of the jaw.

1. Introduction

A simple bone cyst is an intraosseous pseudocyst that lacks an epithelial lining. The cavity is sometimes empty and sometimes filled with serous or sanguineous fluid. Most simple bone cysts occur in the long bones (90%) [1], and occurrences in the jaw bones appear to be rare [2]. The disease is categorized with bone-related lesions, including ossifying fibroma, fibrous dysplasia, osseous dysplasias, central giant cell lesion, cherubism, and aneurysmal bone cyst in the Classification of Head and Neck Tumors of the World Health Organization 2005. Simple bone cysts are often diagnosed before the age of 25 years [3] and are also termed solitary bone cysts, traumatic bone cysts, hemorrhagic bone cysts, hemorrhagic cysts, unicameral bone cysts, or idiopathic bone cysts.

Generally, these cysts are asymptomatic and are found incidentally on radiographs, where they appear as a radiolucent area with well-defined, unilocular or scalloped, irregular margins. In some cases, expansion of the cortical bone can be observed on computed tomography (CT) images. Root absorption is rare, and it sometimes causes disappearance of the hard lamina. Multilocular cysts, and cysts associated with impacted teeth have also been reported [2].

Most simple bone cysts in the jaw are located in the pre-molar or molar regions of the mandible, although some cases cross the midline [4], and some have been reported in the ramus, condyle, or maxilla.

Histologically, the cavity in the cancellous bone has no or minimal liquid content, and is not lined, or is lined with non-epithelial fibrous connective tissue. Small amounts of newly formed bone and collagen deposits may be present, often described as having a fibrin-or cementum-like appearance.

The etiology of simple bone cysts is still unclear, although Harnet et al. stated that there are three predominant theories [5]. Clinical observations, cyst features, the age of diagnosis, and the location near osseous remodeling areas support the theory that it is an osseous growth abnormality. The primary ossification area is located near the mental foramen in the mandible, which is also where simple bone cysts commonly occur. The second theory holds that it is a degenerating tumoral process, similar to osteodystrophic pathological conditions, such as fibrous dysplasia. Hara et al. stated that simple bone cysts may occur as a complication of fibrous dysplasia due to fibrous degeneration [6]. Third, it may be triggered by hemorrhagic trauma, where thrombosis, ischemia, and necrosis may lead to cyst formation [5]. Vascular alterations may also

☆ AsianAOMS: 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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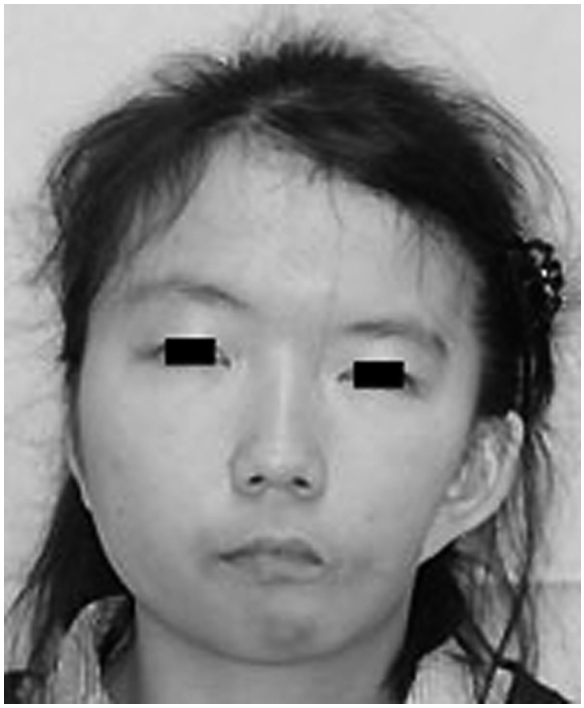

Fig. 1. Facial photograph of the patient at the age of 13.

be related to the resorption phenomena. However, the presence of bilateral, multiple simple bone cysts reduces the possibility of trauma as a causative factor, as traumatic events rarely occur bilaterally. It has therefore been proposed that the term traumatic bone cyst should be deleted from the nomenclature [2].

First and second branchial arch syndrome is the second most common craniofacial malformation after cleft lip and palate. It is characterized as combined tissue deficiencies and hypoplasia of the face, external ear, middle ear, and maxilla and mandibular arches [7]. Mandibular hypoplasia is a clear skeletal finding associated with this deformity. It sometimes presents with a transverse facial cleft and/or a cleft lip and palate. The syndrome is also categorized by hemifacial microsomia due to branchial arch anomalies, a common cause of unilateral congenital anomalies. We here describe the first case of a simple bone cyst occurring in a patient with first and second branchial arch syndrome.

2. Case report

A 13-year-old girl was referred to the 1st Department of Oral and Maxillofacial Surgery of the Osaka University Dental Hospital for examination of an expansion of the lingual and labial cortex in the middle to left region of the mandible that had been incidentally noticed on CT images. The Department of Orthodontics and Dentofacial Orthopedics had regularly followed up the patient for a jaw deformity and malocclusion since the age of 4 years, which were due to first and second branchial arch syndrome. Left-sided chalinoplasty and accessory ear removal had been performed at another hospital previously. She had not experienced trauma before.

The patient's facial features were asymmetric due to hypoplasia of the jaw (Fig. 1). The patient could open her mouth 2 finger-breadths, with left deflection. Intraorally, there was a painless bone-like bulge at the labial gingiva around the left lower canine. All the mandibular incisal, canine, and pre-molar teeth were vital according to the electric pulp test.

On panoramic radiography, the lesion showed discontinuity with the roots, it was a well-defined, multilocular, scalloped radiolucent area that crossed the midline from the right canine to the left pre-molar region of the mandible (Fig. 2B). In addition, a short ramus and abnormally shaped condyle could be seen at the left side of the mandible, due to the first and second branchial arch syndrome. A CT scan showed minor expansion of the lingual and labial cortex (Fig. 3A). The clinical diagnosis was a tumor or cystic lesion of the mandible.

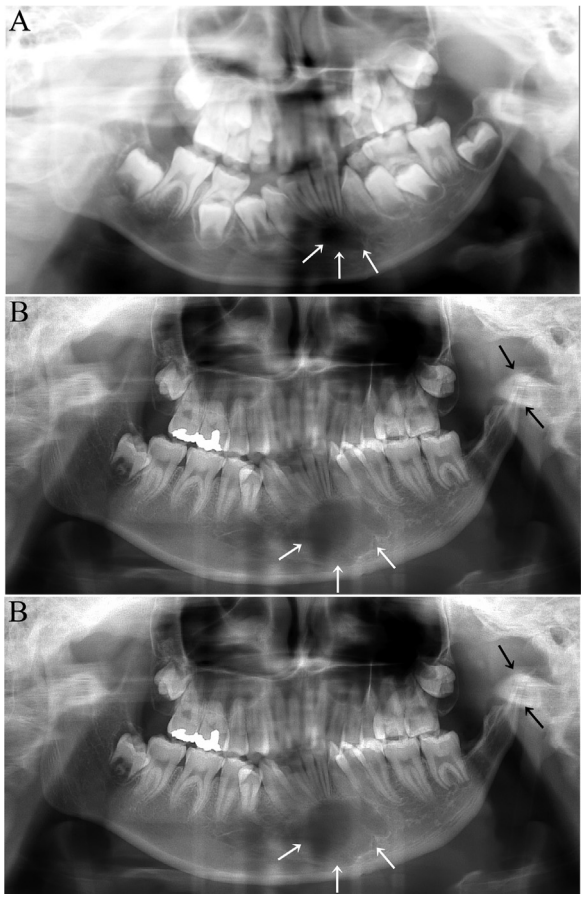

Fig. 2. (A) Panoramic radiographic view at the age of 8 years. A small cyst-like radiolucent area (white arrows) exists. (B) Panoramic radiographic view before the operation. A simple bone cyst (white arrows) crosses the midline from the right canine to the left pre-molar region of the mandible. A short ramus and abnormally shaped condyle can be seen at the left side of the mandible (black arrows). (C) Taken 6 months later after the operation.

We obtained a definite diagnosis of a simple bone cyst by surgical biopsy exploration, approaching from the labial cortex at the right canine. The lesion was without liquid content and was lined with only thin connective tissue layer. There was a possibility that the lesion could grow, causing a pathological fracture, due to the fragile, thin cortex. Moreover, an osteotomy was considered necessary for expansion of the mandible in order to improve the patient's malocclusion. The patient was therefore scheduled for curettage to promote organizing under general anesthesia. A lingual mucoperiosteal flap was made from the right canine to the left pre-molar lesion. Using a round burr, a hole was drilled into the lingual cortex in order to expose the lesion. In the bone cavity, we found a few cyst-wall-like lesions and content. After curetting around the bone wall and promoting bleeding, the flap was replaced and sutured completely. Histological examination of the removed tissue showed fibrous and bone components, without epithelial tissue, and the lesion was therefore diagnosed as a simple bone cyst (Fig. 4).

In the immediate postoperative period, the patient experienced paresthesia of the lower lip, however, it healed completely within 1 month. At 6 months after the operation, the radiolucent area

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