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A case of large developing odontoma, so-called ameloblastic fibro-odontoma, of the mandible

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ABSTRACT

Large developing odontoma is a rare odontogenic tumor showing proliferation of both odontogenic epithelium and ectomesenchyme, with enamel and dentin formation. A case of large developing odontoma affecting the lower molar region to the mandibular ramus of a 14-year-old girl is reported. Panoramic and computed tomography images showed a clearly demarcated, multilocular radiolucency extending from the left lower molar region to the mandibular ramus, containing a tooth-like radiopacity. With the patient under general anesthesia, the mass was removed. The histopathological diagnosis was developing odontoma. There was no sign of recurrence at 1 year after tumor removal.

1. Introduction

Developing odontoma was once called ameloblastic fibro-odontoma (AFO). However, AFO was removed from the latest (4th) edition of the World Health Organization (WHO) Classification of Head and Neck Tumors [1]. Currently AFO has been changed to developing odontoma. Developing odontoma is a rare odontogenic tumor showing proliferation of both odontogenic epithelium and ectomesenchyme, with enamel and dentin formation. In the latest edition of the WHO classification of odontogenic tumors published in 2017, developing odontoma belongs to the group of benign mixed epithelial and mesenchymal odontogenic tumors. Developing odontoma is defined by the WHO as a neoplasm consisting of odontogenic ectomesenchyme resembling the dental papilla, epithelial strands and nests resembling dental lamina, and an enamel organ in conjunction with the presence of dentin and enamel.

Clinically, it presents as a painless swelling of the affected area, usually the posterior portion of the maxilla or mandible. The radiographic findings have been reported to include a well-defined, radiolucent area containing various amounts of radiopaque material of irregular size and form.

A case of a large developing odontoma is presented along with the details of the clinicopathological features of this odontogenic tumor.

2. Case report

A 14-year-old girl was referred to our hospital with swelling in the left cheek and radiological findings of a radiopaque mass in the posterior region of the left mandible.

The patient noticed swelling in her left cheek over a period of months prior to the first visit. She was of medium build with good nutritional status. Extraorally, there was a palpable protuberance in the left cheek, but no hypoesthesia of the chin or trismus was present (Fig. 1). On intraoral examination, there was a palpable bony protuberance on the buccal side of the left lower molar region, with no

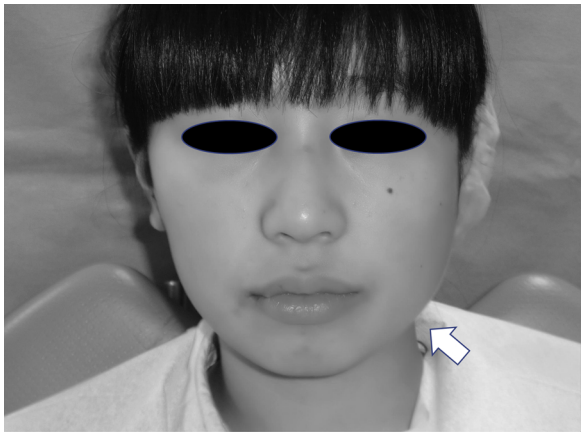

Fig. 1. There is a palpable protuberance in the left cheek.

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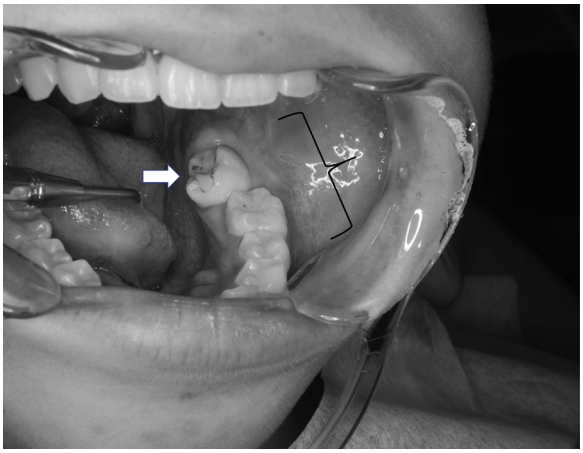

Fig. 2. There is a palpable bony protuberance on the buccal side of the left lower molar region. The left lower second molar is slightly tilted lingually, but it is not mobile.

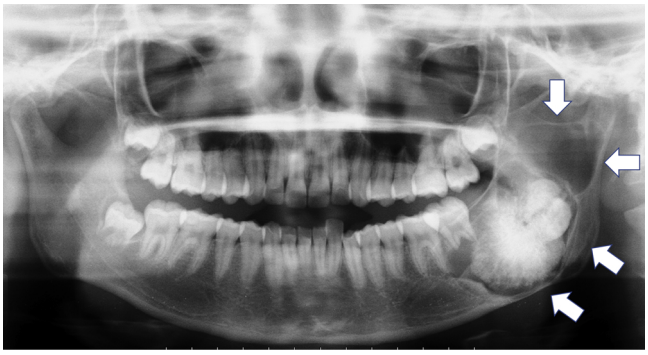

Fig. 3. Panoramic X-ray shows a clearly demarcated, multilocular radiolucency extending from the left lower molar region to the mandibular notch. Root resorption of the left lower second molar is present.

parchment crepitation. The left lower second molar was slightly tilted lingually, but it was not mobile (Fig. 2). Her family and medical histories included nothing of note. A panoramic radiograph showed a clearly demarcated, multilocular radiolucency extending from the left lower molar region to the mandibular notch. Root resorption of the left lower second molar was present (Fig. 3). Computed tomography (CT) images showed a radiolucency extending from the left lower molar region of the mandibular body to the mandibular notch, and there was severe thinning of the cortical bone, although it was not fractured. A radiopacity within the radiolucency showed an appearance suggestive of odontoma (Fig. 4). The clinical diagnosis was a tumor of the left mandible. Under general anesthesia, the patient underwent surgical excision of the lesion, and the left lower second molar was extracted. The wound was left open after tumorectomy. The operative findings showed that the bone from the left lower molar region of the mandibular body to the mandibular ramus had thinned, and the interior of the tumor contained a mixture of capsulated granulomatous tissue and tooth-like hard tissue. To prevent the bone from fracturing during tumorectomy, not much bone was cut, and the hard tissue was removed in pieces (Fig. 5). The tumor was easily detached, enabling complete resection, and it had no liquid content.

Histopathologically, odontogenic epithelium and mesenchyme had proliferated inside the tumor, and enamel and dentin were also present with the appearance of a complex odontoma. The histological presentation was that of an ameloblastic fibroma (Fig. 6A). The tumor contained enamel and dentin formation (Fig. 6B and C). There were no signs of malignancy, and the tumor was diagnosed as a developing odontoma. One year after surgery, there were no signs of recurrence, and CT showed new bone formation within the mandibular body (Fig. 7). Informed consent was obtained from the patient's parents.

3. Discussion

Developing odontoma was once called AFO. However, the true nature of this lesion has long been debated. AFO was removed from the 4th edition of the WHO classification and AFO has been changed to developing odontoma [1].

Developing odontoma is a comparatively rare odontogenic tumor formed by the proliferation of both odontogenic epithelium and ectomesenchyme, with enamel and dentin formation [2]. In Japan, 110 patients with this disorder have been described in the literature to date. It accounts for 1–3% of benign odontogenic tumors, and it commonly occurs in the lower molar region. In the present patient, it occurred in the left lower molar region. It is most frequently seen in young people (age < 20 years), and the present patient was 14 years old. In most cases, the reported long diameter of the tumor is < 20 mm, while it was approximately 75 mm in the present patient, the largest such tumor recorded [2]. In terms of clinical symptoms, it usually manifests as a painless, slow-growing protuberance on the jaw and is frequently associated with abnormal tooth eruption, which in most cases involves impacted teeth. In addition to clinical symptoms, diagnostic imaging such as panoramic X-rays, CT, or MRI is also required for diagnosis. Its imaging characteristics comprise a clearly demarcated unilocular or multilocular radiolucency containing a radiopacity inside, which may include an impacted tooth. The present patient also had a chief complaint of a protuberance in the left cheek, and the left lower second molar was tilted lingually. Imaging showed a clearly demarcated, multilocular radiolucency extending from the left lower molar region to

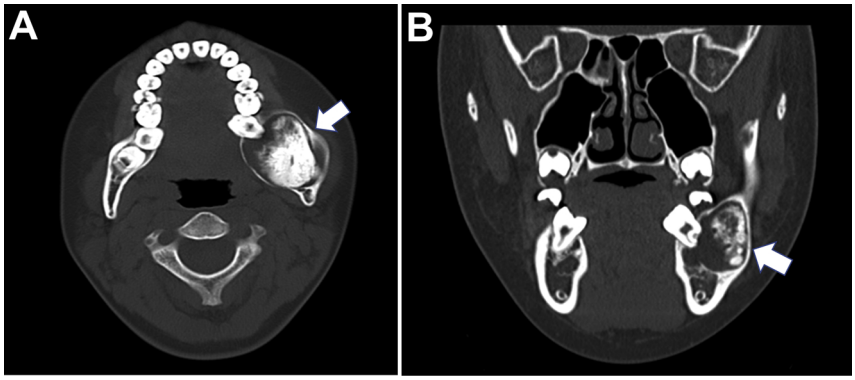

Fig. 4. Computed tomography (CT) images show a radiolucency extending from the left lower molar region to the mandibular notch, with severe thinning of the cortical bone. A radiopacity within the radiolucency shows an appearance suggestive of odontoma.
(A) axial. (B) coronal.

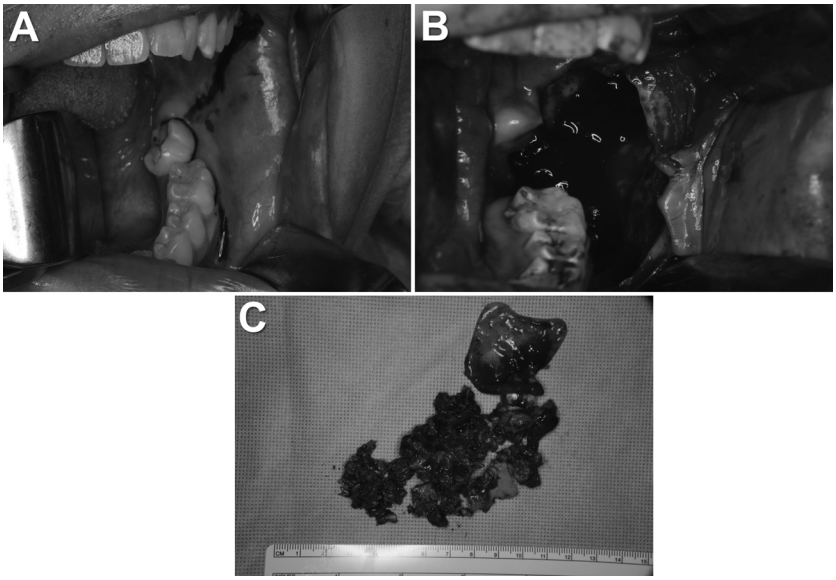

Fig. 5. Photograph during surgery.
(A) Incision line. (B) The tumor of the left mandible is removed, and the left lower second molar is extracted under general anesthesia. (C) Excised specimen.

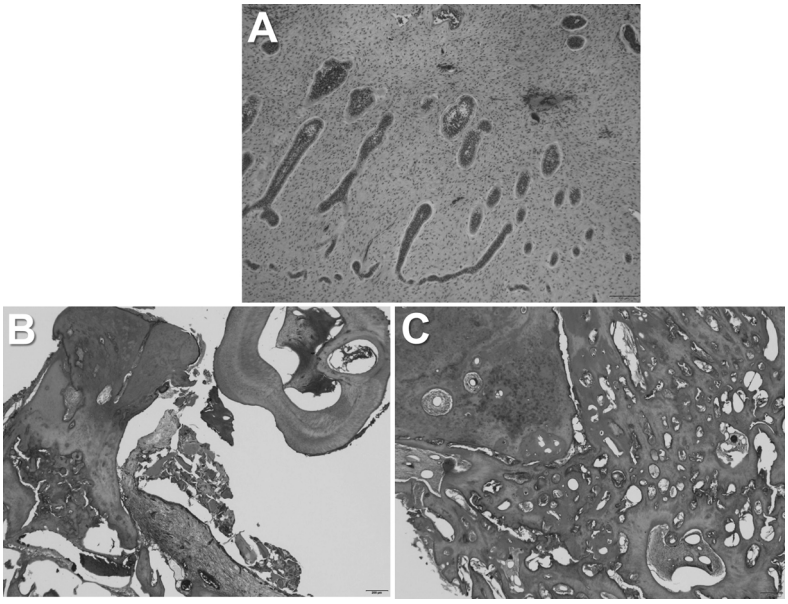

Fig. 6. Histological examination, HE staining.
(A) HE staining shows the histological findings of an ameloblastic fibroma.
(B,C) The tumor contains enamel and dentin formation.

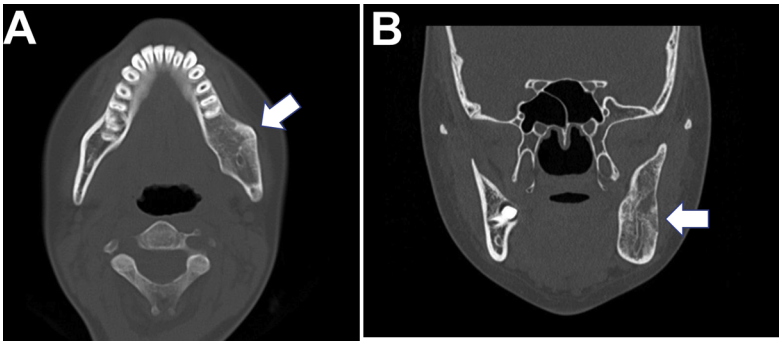

Fig. 7. One year postoperatively, CT shows good bone formation at the tumor removal site, with no sign of recurrence.
(A) axial. (B) coronal.

the mandibular notch, and the left lower third molar was impacted. There is no sex difference or laterality. The treatment is tumorectomy in the vast majority of cases, and there have been some reported cases in which segmentectomy was performed [2]. Since the present patient was young and jaw development was incomplete, tumorectomy was performed. Possible differential diagnoses include ameloblastoma, ameloblastic fibroma, and odontoma. The histopathological characteristics of developing odontoma comprise the proliferation of both odontogenic epithelium and mesenchymal components, with enamel and dentin also present within this tissue. There has been one reported case of recurrence in a patient who underwent fenestration, but no recurrence has yet been reported among patients who underwent tumorectomy in Japan.

However, a case of malignant transformation to ameloblastic fibrosarcoma has been reported outside of Japan [3,4]. The present patient had one of the largest developing odontomas ever reported, and as only one other patient with a tumor of this size has previously been described, this was an extremely rare case. One year postoperatively, the patient's course has been uneventful with no sign of recurrence. Since a case of malignant transformation has been reported, careful observation is important.

In conclusion, the case of a patient with a large developing odontoma that extended from the lower molar region to the mandibular ramus was presented along with a short discussion of the literature. The long diameter of the tumor measured 75 mm, making it only the second developing odontoma of this size ever reported. One year postoperatively, the patient's course has been uneventful with no sign of recurrence. However, since a case of malignant transformation has been reported, careful follow-up is required.

Conflicts of interest

The authors have no conflicts of interest to declare.

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