CASE REPORT

Multiple complex odontomas and subsequent occurrence of an ossifying fibroma at the same site as the removed odontoma

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Abstract We report a rare case of multiple complex odontomas and the subsequent occurrence of an ossifying fibroma at the same site as one of the removed odontomas. A 3-year-old Japanese boy presented three complex odontomas at the unerupted deciduous first and second molars and permanent first molar on the left side of the mandible. The two odontomas at the deciduous teeth were immediately extirpated, and, after 2 years of follow-up, the odontoma at the unerupted permanent first molar was removed. At the age of 7 years, a small odontoma surrounding the deciduous second molar emerged as a recurrence. When the patient was 9 years old, an ossifying fibroma occurred near the apex of the first premolar, i.e., the site where one of the original odontomas existed. Furthermore, a small radiopaque mass over the crown of the unerupted permanent second molar was observed at the age of 10 years; this mass was probably an additional fourth complex odontoma. This is not only a rare case report of ossifying fibroma associated with multiple complex odontomas but also a valuable time course observation of the development of such odontogenic tumors.

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Introduction

Odontomas are lesions composed of developed teeth or tooth-like masses derived from odontogenic epithelial and mesenchymal tissues.\textsuperscript{1-6} These lesions are classified into two types: complex and compound odontomas. The term "complex" refers to the haphazard arrangement of tooth elements such as enamel matrix, enamel, tubular dentin, and pulpal tissue, while the term "compound" refers to the aggregate of recognizable teeth. Both types of odontoma are primarily diagnosed in children, adolescents, and young adults, with no gender predilection. Complex odontomas occur mostly in the posterior part of the mandible and compound odontomas in the anterior maxilla, although both types of odontomas may occur in any tooth-bearing area of the jaws. Odontomas grow slowly without pain and stop growing when they are fully mature. They may cause visible swelling of the jaw. Altered patterns of tooth eruption and/or impaction of teeth may occur. Radiographically, odontomas appear as a radiopaque mass surrounded by a radiolucent zone; recognizable but stunted tooth-like forms may be seen. Although odontomas are one of the most common odontogenic tumors, multiple odontomas rarely occur in humans, unlike in animals.\textsuperscript{7,8} Moreover, because of their limited growth potential, odontomas rarely recur except in the case of incomplete removal at an early, predominantly soft tissue stage.\textsuperscript{1-4,6}

An ossifying fibroma is a well-demarcated lesion consisting of fibrous tissue with varying amounts of mineralized material resembling either bone or cementum or both.\textsuperscript{\textbullet,9} Ossifying fibromas occur in the second to fourth decades of life and are more prevalent in females. These fibromas are mostly observed in the posterior part of the mandible. An ossifying fibroma causes expansion of the involved bone and continues to enlarge until removed. Radiographically, the neoplasm is a well-delineated radiolucent or mixed radiolucent and radiopaque lesion depending on the contributions of soft and hard tissue components.

In this case report, we describe three rare occurrences: multiple complex odontoma, recurrent odontoma, and an ossifying fibroma arising at the same site as the removed odontoma.

Case report

Delayed eruption of the deciduous first and second molars of the left mandible was noted in a 3-year-old Japanese boy during health checkups. He was referred to a local hospital where odontomas were suspected, and surgery recommended. Finally, he was admitted to our hospital for the treatment of these tumors. At presentation, slight swelling on the left side of the mandibular body was felt on palpation, although the patient’s face was almost symmetric with normal-colored overlying skin. Lymph nodes did not show enlargement or sensitivity to pressure, and the family history did not indicate any similar or related abnormalities. Intraorally, the left buccal alveolar bone corresponding to the positions of the unerupted deciduous molars showed slight swelling without spontaneous pain.

Radiographic examination revealed three independent radiopaque masses in the left mandibular body (Fig. 1A).

The first mass was located on the buccal side of the unerupted first deciduous molar (Fig. 1A, arrows). The second mass was located above the crown of the unerupted second deciduous molar, and the third was situated over the tooth germ of the permanent first molar. We diagnosed these masses clinically as odontomas. For descriptive purposes, the three lesions were named Lesions 1, 2, and 3, respectively (Table 1).

Lesions 1 and 2 were enucleated to allow the two deciduous teeth to erupt. Each lesion was surrounded by a dental sac. Bony tissue intervened between the two lesions. Lesion 3 was not treated at this time but was followed up.

The permanent first molar beneath Lesion 3 had not erupted after 2 years of follow-up. A panoramic radiograph at the age of 5 years showed expansion of the radiopaque area of Lesion 3 (Fig. 1B). We therefore excised Lesion 3. In addition, the radiograph showed partial anodontia of the left second premolar in the mandible. Developmental delay and displacement of the permanent second molar were also observed (Fig. 1B, arrow).

When the patient was 7 years old, a small piece of odontoma-like hard tissue (Lesion 4) erupted into the oral cavity, surrounding the distal cervix of the deciduous second molar (Fig. 1C, arrow). After extracting the deciduous first and second molars, we excised Lesion 4. Retrospectively, a radiograph at the age of 3 years revealed a spicular radiopaque structure covering the distal surface of the crown in Lesion 2 before its removal (Fig. 1A, arrowhead). The spicular structure became thicker two years after the excision of Lesion 2 (Fig. 1B, arrowhead).

When the patient was 9 years old, a swelling was noted on the left lower gingiva corresponding to the first premolar with inclination of its cusp. Orthopanoramic radiography showed a well-demarcated, unilocular radiolucency (Lesion 5) near the apex of the first premolar (Fig. 1D). Computed tomography (CT) revealed a low-density area (CT number: 41 Hounsfield units) at the buccal side of the first premolar with focal expansion of the buccal cortical bone (Fig. 2A). Retrospectively, the radiograph taken at five years of age revealed a small cystic lesion just below the first premolar (Fig. 1B, asterisk). The growing cystic lesion could be observed although the digital X-ray image taken at the age of seven years was in poor condition (Fig. 1C, arrowheads). These clinical imaging findings suggested that Lesion 5 was a recurrent tumor of the removed complex odontoma (Lesion 1). The tooth was extracted, and Lesion 5 was totally excised by surgery. Lesion 5 was an opalescent solid mass that did not involve the root of the tooth (Fig. 2B). Histopathologic examination revealed that the tumor was not a recurrent odontoma but an ossifying fibroma.

During 3 years of follow-up after the extirpation of Lesion 5, we detected no signs of further recurrence (Fig. 1E and F). However, at the age of 10, a small, indistinct radiopaque mass was observed in the dental sac of the unerupted, underdeveloped permanent second molar (Fig. 1E, arrows). At age 12, radiopaque mass covering the crown of the second molar was clearly observable (Fig. 1F, arrows). On the basis of the radiographs, we diagnosed this radiopaque mass (Lesion 6) as an odontoma. Lesion 6 was untreated at the time of writing.
Pathologic findings

Histopathologically, Lesions 1 and 2 showed not only a disordered mixture of dental hard tissues characteristic of complex odontoma but also substantial immature odonto- genic soft tissues, which were suggestive of ameloblastic fibro-odontoma (Fig. 3A to C). Based on the sizes and locations of these lesions, complex odontoma at developing stage appeared to be a more appropriate diagnosis than ameloblastic fibro-odontoma. An additional noteworthy finding was the presence of small pieces of highly cellular tissue consisting of spindle cells with small round cementum-like masses in the histological section of Lesion 1 (Fig. 3D); these were not observed in specimens from Lesion 2. These unusual elements in Lesion 1 were similar to those in Lesion 5, an ossifying fibroma, as described later.

Specimens of Lesion 3 showed almost the same histological features as those of Lesions 1 and 2; however,
immature odontogenic soft tissues were considerably less prevalent than hard tissues in Lesion 3 (Fig. 4A). In addition, unlike Lesions 1 and 2, differentiation into cemento-osseous tissue was noted (Fig. 4B).

Lesion 4 was mainly composed of mature dental hard tissue with only a small amount of soft tissues, such as outer enamel epithelium and stellate reticulum of the enamel organ and the periodontal membrane (Fig. 5).

**Table 1** Summary of lesions in the patient’s left mandible.

<table>
<thead>
<tr>
<th>Lesion</th>
<th>Location (figure)</th>
<th>Age (yrs)</th>
<th>Pathology (figure)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Buccal side of the first deciduous molar (Fig. 1A, arrows)</td>
<td>3</td>
<td>CO with a small OF-like area (Fig. 3)</td>
</tr>
<tr>
<td>2</td>
<td>Over the crown of the second deciduous molar (Fig. 1A), [distal side of the same tooth’s crown (Fig. 1A and B, arrowheads)]a</td>
<td>3</td>
<td>CO (not shown)b</td>
</tr>
<tr>
<td>3</td>
<td>Over the crown of the first permanent molar (Fig. 1A and B)</td>
<td>3</td>
<td>CO (Fig. 4)</td>
</tr>
<tr>
<td>4c</td>
<td>Distal cervix of the second deciduous molar (Fig. 1C, arrow) [Fig. 1A and B, arrowheads]a</td>
<td>7</td>
<td>CO (Fig. 5)</td>
</tr>
<tr>
<td>5</td>
<td>Near the apex of the first premolar (Fig. 1D), [Fig. 1B, asterisk; 1C, arrowheads]a</td>
<td>9</td>
<td>OF (Fig. 6)</td>
</tr>
<tr>
<td>6</td>
<td>Over the crown of the second permanent molar (Fig. 1E and F, arrows)</td>
<td>10</td>
<td>Not yet excised</td>
</tr>
</tbody>
</table>

CO = complex odontoma; OF = ossifying fibroma.

a Additional retrospective findings.

b Histological features of Lesion 2 were similar to those of Lesion 1, except for the absence of the ossifying fibroma-like area observed in Lesion 1.

c Lesion 4 was a recurrence of Lesion 2 because of incomplete removal.

Figure 2 Computed tomography (CT) and macroscopic aspect of Lesion 5 at the age of 9 years. (A) Axial CT image shows a soft-tissue-density structure without including high-density structures in the left mandibular body and adjacent cortical bone with lateral expansion defining the boundary of the lesion (arrow); (B) extirpated tumor is an opalescent solid mass. The root of the first premolar is not involved in this mass.
The histological section of Lesion 5 revealed highly cellular fibrous tissue consisting of spindle- to ovoid-shaped fibroblastic cells and dispersed cemento-osseous masses (Fig. 6). Some of the hard tissues had a smooth contour with a radiating fringe of collagen fibers. The trabecular bone and psammoma body-like ossicles were absent. No epithelial elements such as odontogenic epithelial islands were observed in the tumor tissue. The tumor tissue was demarcated by fibrovascular tissue. We diagnosed Lesion 5 as an ossifying fibroma according to the latest (2005) World Health Organization classification of odontogenic tumours.9 Although Lesion 5 occurred at the age of 9 years, there was no clinical or histological feature such as a juvenile trabecular ossifying fibroma or juvenile psammomatoid ossifying fibroma.9 Because Lesion 5 was a well-demarcated mass, fibrous dysplasia was ruled out.6,10 Macroscopic and microscopic findings of Lesion 5 did not match those of focal osseous dysplasia, which is composed of multiple hemorrhagic fragments with cavernous-like vascularity and ginger root-like thick curvilinear trabeculae.9,11–14

**Discussion**

It is widely accepted that the fully developed calcified odontoma should be classified as a hamartoma rather than a neoplasm, although a developing complex odontoma is sometimes difficult to distinguish from other neoplastic mixed odontogenic tumors such as ameloblastic fibroma and fibro-odontoma.1,2,4–6 In our case, the three lesions found at the age of 3 years (Lesions 1–3) showed haphazardly arranged dental hard and soft tissues in histological findings. Because such findings commonly occur both in a developing complex odontoma and an ameloblastic fibro-odontoma, it is difficult to make a differential diagnosis of these lesions from histological findings alone. It seemed appropriate, however, to diagnose the three lesions as complex odontomas at a developing stage, rather than ameloblastic fibro-odontomas, because all the lesions were small and close to the impacted teeth—particularly Lesions 2 and 3, which directly overlaid the crowns of unerupted teeth.1,6

Although odontomas are among the most common odontogenic tumors, multiple odontomas are rare, except in some systemic syndromes such as familial adenomatous polyposis.7,8,15,16 In the present case, the three odontomas were independently located with no continuity between the flanking lesions, indicating multiple odontomas without systemic lesions. Moreover, a small radiopaque mass (Lesion 6) over the crown of the unerupted permanent second molar was observed from the age of 10 years (Fig. 1E and F). Lesion 6 was probably an additional fourth complex odontoma.

Moreover, recurrence of odontoma is rare.2,3,6 The complex odontoma found at the age of 7 years (Lesion 4) probably resulted from incomplete excision of Lesion 2, because the radiographs at the ages of 3 and 5 years revealed the presence of a radiopaque mass covering the distal side of the crown before and after the excision of Lesion 2 (Fig. 1A and B).

On the other hand, Lesion 5 emerged from the region where the complex odontoma (Lesion 1) previously existed as an ossifying fibroma rather than a recurrent odontoma. Ossifying fibromas in the jaw are generally considered to originate from periodontal tissue although there may be
some exceptions. Microscopically, Lesion 1 revealed small pieces of highly cellular fibrous tissues containing small cementum-like masses together with bony fragments (Fig. 3D). The fibrous tissues in Lesion 1 were similar to those in the ossifying fibroma in Lesion 5. Because Lesion 1 was excised from the adjacent buccal side of the deciduous first molar, pieces of developing periodontal tissues of the tooth may have been included in the excised tumor tissue. Histological findings raise the possibility that Lesion 5 may have developed from a microscopic remnant of the periodontal tissues of the deciduous first molar. However, it is also likely that this case of ossifying fibroma occurred de novo in the overlapping area where a complex odontoma previously existed. Furthermore, it is also possible that immature pluripotent cells from the remnants of the complex odontoma (Lesion 1) differentiated in the environment of root formation of the first premolar and developed into an ossifying fibroma immediately below the root.

Interestingly, there is another case report similar to the present case. A case report by Ohtake et al describes three complex odontomas associated with ossifying fibroma of the right mandible in a 10-year-old Japanese boy with no significant related factors except treatment of epilepsy. These authors found a proliferation of highly cellular fibrous tissue containing cemento-osseous hard tissues close to the neighboring complex odontoma and the roots of the unerupted tooth. Considering the case reported by Ohtake et al together with our own, it appears reasonable to surmise that, under certain conditions, an immature element of an odontoma near the root of an existing tooth may develop into an ossifying fibroma.

Although it is difficult to determine the origin of the ossifying fibroma in our case, the fact that multiple complex odontomas and an ossifying fibroma developed close to each other in the fairly limited area of the left side of the mandible suggests the possibility of a local environment permissive for the growth of multiple odontogenic tumors. It would be interesting to investigate the gene expression in this local environment and the genetic background of patients with such lesions.

Finally, we wish to emphasize that this case report provides a valuable time course observation of the development of

Figure 4  Histological features of Lesion 3 (hematoxylin and eosin stain). (A) Compared with Lesion 1 (Figure 3), immature odontogenic soft tissues are considerably less common than hard tissues in Lesion 3; (B) differentiation into cemento-osseous tissue is observed.

Figure 5  Histological features of Lesion 4 (hematoxylin and eosin stain). (A) Lesion 4 is mainly composed of mature dental hard tissue with only a small amount of soft tissue; (B) outer enamel epithelium and stellate reticulum of the enamel organ with enamel and dentin; (C) tubular dentin and cementum; (D) cementum with Sharpey fibers and periodontal membrane.
multiple odontogenic tumors, including complex odontomas, a recurrent odontoma, and an ossifying fibroma.

References