Dentigerous Cyst Associated with a Supernumerary Malformed Tooth: Report of a case and a clinicopathologic review

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A case of a dentigerous cyst associated with a malformed supernumerary tooth is reported, and the clinicopathological characteristics of dentigerous cysts with abnormal dentition are reviewed from the literature and our hospital records. A 7-year-old boy had a painless and radiolucent lesion in his right molar region of the mandible. Radiographic examinations revealed that a well-defined cystic lesion, measuring 20 mm in diameter and containing a tooth-like calcified structure, was located between the first and second molar tooth germs. Histopathologically, the cyst wall was composed of fibrous granulation tissue with a squamous epithelial lining. The calcified structure was a reversely concaved tooth crown which protruded into the cystic lumen with its enamel layer at the inner surface. The cyst lining epithelium was continuous with the reduced enamel epithelium covering the malformed tooth. A review of the literature and a case survey in our hospital showed that about 10% of dentigerous cysts were associated with supernumerary teeth which were mostly located in the median region of the maxilla, but that those in the mandibular molar region were extremely rare.

Key words: dentigerous cyst, mandible, supernumerary teeth, tooth malformation

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Introduction
Dentigerous cysts are the most common development cysts arising in the jaw bones. Most typical examples are those associated with the third molar teeth of the mandible, followed by maxillary canines and premolars of both maxillary and mandibular bones (1).

Tooth impaction is most frequently observed in third molar teeth and in teeth in the premaxilla region of both jaw bones. On the other hand, the majority of supernumerary teeth are impacted, and only a small number of them erupt but are malformed in most instances (2). Although the incidence of supernumerary teeth is not so small among impacted teeth, there have been only a small number of studies on the association of dentigerous cysts with supernumerary teeth (3-8).

In this study, we report a case of a dentigerous cyst involving a supernumerary and malformed tooth, which was located in the retromolar region of the mandible of a 7-year-old boy. In addition, we surveyed the incidences of dentigerous cysts involving supernumerary teeth in our hospital and in the English literature.

Case report
A 7-year-old boy was referred to the Oral and Maxillofacial Surgery Unit of the Niigata University Dental Hospital for evaluation and treatment of a painless and radiolucent lesion in his right molar region of the mandible, which had been noticed for a month by his dentist. The patient had no history of trauma or dental treatment around the lesion. On intraoral examination, a slight bone expansion was palpable from the retromolar region to the anterior ramus area. The overlying oral mucosa was normal in color and texture. There was no paresthesia over the mandibular area. A panoramic radiograph revealed...
a well-defined radiolucent lesion, measuring 20 mm in diameter, involving a tooth-like hard tissue located between the first molar and the second molar tooth germ (Fig. 1a). Computed tomography demonstrated that the radiolucent lesion extended from the right molar region to the anterior part of the ramus and involved an inverted-shaped tooth-like calcified structure (Fig. 1b). Other than this lesion, the patient's general health and development were normal. Under a clinical diagnosis of a dentigerous cyst of the mandible, marsupialization of the cyst was performed and it was histopathologically diagnosed as a dentigerous cyst. Seven months after the initial surgical procedure, the cyst was extirpated with the patient under general anesthesia. His postoperative course was uneventful for 3 years following the surgery.

Pathological findings

Macroscopic findings
The surgical specimen was 25 15 10 mm in size. It was a cystic structure, composed of fibrous connective tissue at the distal/buccal side in part and of hard tissue at its medial/lingual side in part. The hard tissue showed a dome-shaped protrusion with a smooth, white and round-ended outer surface. On the cut surface, the cystic space extended around the hard tissue, which had a tooth-like appearance. The inner surface of the cyst wall was brown in color and coarsely granular (Fig. 2a). The specimen was demineralized and processed for histological examination.

Microscopic findings
The cyst wall was composed of fibrous granulation tissue with a squamous epithelial lining. A dome-shaped hard tissue constituted one third of the cyst wall (Fig. 2b). The inner layer of the granulation tissue was still immature, and the lining epithelium showed reactive hyperplasia due to severe inflammatory backgrounds. The hard tissue was composed of dental components: its main part consisted of dentin with a cementum-like layer at the outer surface and with an apparent predentin layer in the inner side (Fig. 3a). The inner surface of the dentin...
layer was partially attached with conglomerates of dysplastic dentin, which had irregular- and various-shaped holes resulting from demineralization of enamel or needle-shaped clefts due to cholesterol crystals, and indicated a prolonged inflammatory process (Fig. 3b). A larger part of the inner layer of the tooth-like structure was vacant due to demineralization of the enamel and was covered by a thin layer of reduced enamel epithelium, which was partially detached into the cystic lumen. The reduced enamel epithelium was continuous with the cyst lining epithelium at the base of the hard tissue (Fig. 3c). Thus, the tooth-like structure was considered to be a “concavo-convex” tooth crown with inside-out layering of enamel and dentin. These definite spaces and the layering of dentin and enamel suggested that this particular structure was a malformed single tooth, although there were no pulp nor periodontal ligament around the hard tissue. The final diagnosis was a dentigerous cyst associated with a supernumerary malformed molar tooth of the mandible.

Clinicopathologic and literature reviews

Dentigerous cyst cases were collected from the file of the Surgical Pathology Laboratory of the Niigata University Dental Hospital during the past 34 years 9 months, from December 1967 to August 2002. The patient records, including age, sex, teeth associated with cysts and anatomical location were examined, and their tissue sections were histopathologically reviewed. The reported dentigerous cysts associated with supernumerary teeth in the literature were also retrieved (3-8). They are summarized in Table 1.

In our hospital from 1967 to 2002, a total of 252 dentigerous cysts were diagnosed among 249 Japanese patients (3 patients had two independent cysts each), among which 25 cysts (9.9%) were associated with super-

### Table 1: Anatomical distribution of dentigerous cysts involving regular and supernumerary teeth

<table>
<thead>
<tr>
<th>Jaw bones/ sources</th>
<th>Teeth associated with dentigerous cysts</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Regular teeth</td>
<td></td>
</tr>
<tr>
<td></td>
<td>incisor</td>
<td>canine</td>
</tr>
<tr>
<td>Niigata</td>
<td>18</td>
<td>14</td>
</tr>
<tr>
<td>Literature</td>
<td>23</td>
<td>71</td>
</tr>
<tr>
<td>Subtotal</td>
<td>41</td>
<td>85</td>
</tr>
<tr>
<td></td>
<td>Supeneruous teeth</td>
<td></td>
</tr>
<tr>
<td></td>
<td>incisor (mesiodens)</td>
<td>canine</td>
</tr>
<tr>
<td>Niigata</td>
<td>22 ( 5 )</td>
<td>0</td>
</tr>
<tr>
<td>Literature</td>
<td>20 ( 7 )</td>
<td>0</td>
</tr>
<tr>
<td>Subtotal</td>
<td>42 (12 )</td>
<td>0</td>
</tr>
</tbody>
</table>

### Table 1 Legend

- **Maxilla**
- **Mandible**
- Niigata: Data from our hospital's records
- Literature: Data from the literature
- Incisor: Upper incisors
- Canine: Upper canines
- Premolar: Upper premolars
- Molar (third): Upper third molars
- Molar (mesiodens): Upper mesiodens
- Total: Sum of all teeth associated with cysts
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Numerary teeth. The age and sex distribution in these samples is shown in Fig. 4. In the cysts with teeth in regular dentition, there were 138 male and 89 female patients, a ratio of 1.6:1. These cysts were more frequently observed in patients who were in their second and the fifth decades, which together constituted about 35% of the total cases, and the mean age of the patients was 35.1 years. There were 51 cysts in the maxilla and 287 in the mandible, a ratio of 1:3.5. The maxillary dentigerous cysts involved the incisor (35%), the third molar (29%) and the canine (27%) teeth, while the mandibular cysts involved exclusively the third molar teeth (75%), which accounted for 58% of the cases, and the premolar (18%) or the canine (2.0%) (Table 1). In contrast, the majority of the mandibular cysts occurred in the third molar region (196 out of the 287, 68%), followed by the premolar (14%), canine (5.2%) and incisor (2.4%) teeth. In addition, 25 dentigerous cysts associated with supernumerary teeth were documented. Among them, male patients were more frequently observed than female patients at a ratio of 2:1. The age of the patients ranged from 9 to 71 years, peaking in the fifth decade and with a mean of 29.0 years, which was much younger than the patients in our hospital. Most of the cysts with supernumerary teeth were found in the incisor region of the maxilla (20 out of the 25 total cases, 80%), including 7 mesiodens (28%), which was almost the same as the ratio of the patients in our hospital.

Discussion

It was shown from the present review of the literature and a case survey in our hospital that dentigerous cysts most often involve the third molars in the mandible and the incisor and canine teeth in the maxilla. The occurrence of cysts with supernumerary teeth is exclusively limited to the incisor region of the maxilla including mesiodens, but those in the mandibular molars are quite rare. In addition, the supernumerary tooth in the present case was malformed, hence, this is the first case report of a dentigerous cyst associated with a malformed supernumerary tooth.

Supernumerary teeth are more frequently found in the permanent dentition than in the primary one (12-13). It has been reported that there is a sex predilection for the occurrence of supernumerary teeth. Males are affected twice as often as females, and the occurrence in the maxilla is about 8 times as frequent as that in the mandible (14). According to a radiographic study by Stafne (15), 60% of his series of supernumerary teeth were found in the incisor and canine regions, and 38% were found in the fourth molars in the maxilla, while only 2% were found in the fourth molars and no other tooth region in the mandible. They also reported that unerupted supernumerary teeth were found 5 times more frequently than erupted ones based on a radiogram. Thus, it is obvious.

Fig. 4: Age and sex distribution of patients with dentigerous cysts associated with regular teeth vs. supernumerary teeth from the Niigata University Dental Hospital (upper) and the literature (lower). □ male patients with dentigerous cysts involving regular teeth. ■ female with regular teeth. ◯ male with supernumerary teeth, □ female with supernumerary teeth, □ both male and female with regular teeth.

gion of the maxilla (88%), while only 3 (12%) were in the mandible (Table 1).

In the English literature from 1964 to 2002, 694 cases of dentigerous cysts associated with regular teeth were reported (9-11). Among them, there were 371 male and 246 female patients, a ratio of 1.5:1. The patients’ ages ranged from one to nine decades with more frequencies in the third to fourth decades, which together constituted about 40% of the total cases. The mean age of the patients was 34.5 years. There were 139 cysts in the maxilla and 287 in the mandible, a ratio of 1:2.1. Among the 139 maxillary dentigerous cysts, the canine teeth were most frequently involved (51%), followed by the molar (22%), incisor (17%), and premolar (11%) teeth. In contrast, the majority of the mandibular cysts occurred in the third molar region (196 out of the 287, 68%), followed by the premolar (14%), canine (5.2%) and incisor (2.4%) teeth. In addition, 25 dentigerous cysts associated with supernumerary teeth were documented. Among them, male patients were more frequently observed than female patients at a ratio of 2:1. The age of the patients ranged from 9 to 71 years, peaking in the fifth decade and with a mean of 29.0 years, which was much younger than the patients in our hospital. Most of the cysts with supernumerary teeth were found in the incisor region of the maxilla (20 out of the 25 total cases, 80%), including 7 mesiodens (28%), which was almost the same as the ratio of the patients in our hospital.
that some of the dentigerous cysts resulted from supernumerary teeth that remain embedded, and this condition can only be revealed by a radiographic examination in the later stage of patients' lives unless they have no complications.

Mourshed (10) reported in 1964 that 9 (5.2%) out of 172 dentigerous cysts were associated with supernumerary teeth. This ratio is about half of the ratio of our hospital data, which was 25 (9.9%) of the 253 dentigerous cysts. This may be because the prevalence of supernumerary teeth in the Japanese population is larger than that in a Caucasian population or because contemporaries have developed such a tendency after one generation since 1964. The average age of the patients with dentigerous cysts with supernumerary teeth in Niigata (35.7-years-old) was greater than that found in the literature (29.5-years-old). In recent years, the accidental discovery of supernumerary teeth is more likely to occur in older patients, who are more likely to undergo a radiographic examination in this country.

There have been no histological studies on supernumerary tooth variation nor a histological record of teeth involved in dentigerous cysts. The malformation of the tooth in the present case was just a tooth crown with inside-out layering. A similar malformation was experimentally generated by the anti-sense for hepatocyte growth factor (HGF), which blocked the normal histogenesis without a toxic effect to the cytodifferentiation of ameloblasts and odontoblasts (16). This experiment indicates that growth factors, such as HGF, function in this particular step of tooth morphogenesis, which is mediated by epithelial-mesenchymal interactions. Therefore, it is suggested that some disturbed expression of the genes of growth factors, which might have taken place during the development of the dentigerous cyst, was one of the causes of the tooth malformation in the present case. It remains unknown why the supernumerary tooth was induced and malformed in the molar area because there was no contributory history in the patient.

**References**


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