Dentigerous Cyst Involving Two Impacted Molars; Report of a case

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The authors report a rare case of a mandibular dentigerous cyst involving two impacted molars in a 48-year-old Japanese man. Two crowns of the third molar and the fourth supernumerary one were facing each other in the cystic lumen. Histologically, the cyst wall was composed of fibrous granulation tissue with a squamous-epithelial lining. The pathogenesis is discussed, based on a review of the literature of dentigerous cysts and supernumerary teeth.

Key words: dentigerous cyst; impacted teeth; supernumerary tooth

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Introduction

Dentigerous cysts are the most common of odontogenic and developmental cysts arising in the jaw bone. They are frequently found in association with impacted third molars of the mandible or maxilla. This type of cyst usually contains one molar crown in the cystic lumen (1).

In this report, we describe a rare case of a dentigerous cyst involving two crowns of the mandibular molars. Clinical and radiographic findings of the lesion are presented with histopathologic observations of the cyst wall. We also discuss the possible pathogenesis of this rare cyst.

Case report

A 48-year-old Japanese man visited the Oral and Maxillofacial Surgery Department of Niigata University Dental Hospital complaining of swelling and pain in the left retromolar region of his mandible. He had ignored dull pain in the same area for years without receiving any medical treatment. However, one month ago, he visited a dentist for trismus and a severe throbbing pain in the region. After an anti-inflammatory medication, he was referred to our hospital. On examination, a diffuse and tender swelling was found in the left gonial area. The overlying skin showed no surface changes. The left lower molars were missing and the oral mucosa was slightly red in the left retromolar area. Radiographic examination showed two impacted teeth; the posterior one was in an inverted position, and they faced each other crown to crown within a well-defined unilocular radiolucency in region from the left mandibular angle to the ramus (Fig. 1). The oval-shaped radiolucent area sur-

rounded the two impacted tooth crowns (Fig. 2). These two impacted teeth could neither be identified as second and third molars nor as third and fourth supernumerary molars, because the patient had little recollection of his past dental treatments. The clinical diagnosis was dentigerous cyst with two impacted teeth.

The two impacted molars and the dentigerous cyst were removed under local anesthesia. The overlying bone cortex was noticeably thinner at the buccal side. After marsupialization of the cyst from the buccal side, the two impacted teeth were extracted, and then the rest of the cyst wall was enucleated. There was no obvious fluid content in the cystic lumen. The postoperative healing was uneventful.

Gross findings

The extracted teeth had similar appearances. Their cusps were not well developed with corrugation; their roots were conical and had no bifurcation. Macroscopically, we regarded them as a third molar and a supernumerary tooth, namely a fourth molar, because of their immature shapes as mandibular molars. A cut section of the 10%-formalin-fixed specimen revealed that the gray-white cyst wall was 1 to 2 mm in thick. The inner surface was brown in color and coarsely granular. The cyst wall was thicker on the lingual side than the buccal side.

Histological findings

The cyst wall was composed of granulation tissue. The inner layer consisted of a severe infiltration of lymphocytes, plasma cells, and macrophages and a proliferation of small blood vessels. The outer layer was fibrous

Fig.1: Panoramic radiograph showing two impacted molars in an oval radiolucency in the left retromolar region of the mandible.

Fig. 2: Dental radiograph of the left retromolar region showing a dentigerous cyst with two crowns of impacted teeth.

Fig. 3: Histology of cyst wall (Hematoxylin & Eosin, $\times 50$). Cyst wall is composed of granulation tissue, and its inner aspect is lined by stratified squamous epithelium with reactive hyperplasia.

with new bone formation. The inner surface was lined with stratified squamous epithelium with elongated and irregular-shaped rete processes (Fig.3). No keratin formation was obvious. There were no particular findings suggestive of the route of infection.

Discussion

A dentigerous cyst involving two impacted teeth in one follicle is exceedingly rare. As far as we determine from the Medline, there has been no such case reported. However, we have learned that such cysts are reported in the literature with the help of the electric mailing list "Bulletin Board of Oral Pathology (BBOPLIST)." One

was reported by Schofield (2). In his case, the right third molar and fourth supernumerary one were involved in one follicle in the mandible of a 27-year-old female. The radiographic appearance was almost the same as ours. The two molars faced each other in the common follicular space. The author described such a condition as "kissing molars." However, they did not mention any cystic development of the follicle. Similar radiographs were published in textbooks by Bhaskar (3) and Beeching (4). The latter presented two cases with multiple impacted teeth: one dentigerous cyst with two molars in an 8-year-old boy with familial fibrous dysplasia or cherubism and another with four numerary premolars in a 38-

year-old man with cleidocranial dysostosis. It is well known that such systemic disorders as Gardner's, syndrome and cleidocranial dysostosis tend to manifest hyperodontia (5). In the present case, however, there was no such syndromic sign.

Dentigerous cysts usually involve mandibular and maxillary third molars and maxillary canines. Associations with supernumerary teeth are rather rare; the incidence is estimated to be 5-6% of all dentigerous cysts (1,6). According to the review by Lustmann and Bodner (6), most of supernumerary teeth involved in dentigerous cysts are maxillary mesiodens. They found only a few mandibular cases in the literature, one of which, reported by Abrams and Nelson (7), was a dentigerous cyst around a supernumerary fourth molar fused to a normally erupted third molar. Fourth molars not associated with cysts do not seem to be uncommon (8,9).

Since dentigerous cysts seem to develop after the tooth crown is mostly formed, two speculations could explain the pathogenesis of the multiple tooth involvement: one is a fusion between two adjacent dentigerous cyst linings, and the other is a fusion between the lining of one preexisting cyst (possibly of the fourth molar in the present case) and the lining of reduced enamel epithelium surrounding the adjacent tooth (the third molar). These possibilities have been nicely demonstrated by Altini and Cohen (10,11) in their work on the histogenesis of the follicular keratocyst. They suggested that a tooth might erupt into a preexisting keratocyst cavity in exactly the same way that a tooth erupts into the oral mucosa. Anyhow, problems with the last stages of tooth formation would cause such a fusion of two tooth follicles.

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