

Dentigerous cysts associated with impacted supernumerary teeth in the anterior maxilla

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Abstract. Dentigerous cysts are thought to be caused by a developmental abnormality derived from the reduced enamel epithelium of the tooth forming organ. Most typical dentigerous cysts are those associated with the third molar teeth of the mandible, but rarely involve impacted supernumerary teeth in the anterior maxilla. Swelling and/or pain may be the major complaints of the patients. Herein, we review the literature spanning the past 22 years concerning dentigerous cysts associated with supernumerary teeth in the anterior maxilla, and present four additional cases with emphasis given to the clinicopathological characteristics of this type of dentigerous cyst.

Introduction

Dentigerous cyst, also known as follicular cyst, is an odontogenic cyst caused by fluid accumulation between the reduced enamel epithelium and the enamel surface of a formed tooth. It is thought to be a developmental abnormality derived from the reduced enamel epithelium of the tooth forming organ (1).

A dentigerous cyst is most frequently found in individuals in the age group between 20 and 40 years (2). Most typical dentigerous cysts are those associated with the third molar teeth of the mandible, followed by maxillary third molars, maxillary canines and premolars of both the maxillary and mandibular bones (3). They are occasionally associated with supernumerary teeth (3,4). Stafne first described dentigerous cysts associated with supernumerary teeth and found an incidence of 5.5% among 200 supernumerary teeth (8). Most supernumerary teeth are noted in the anterior maxillary region.

The most common supernumerary tooth which appears in the maxillary midline has also been named a mesiodens due to its position in the center of the maxilla (6).

In the present study, we review the literature spanning the past 22 years concerning dentigerous cysts associated with supernumerary teeth in the anterior maxilla, and present four additional cases with emphasis on the clinicopathological characteristics of this type of dentigerous cyst. A search of Medline from 1988 to 2010 was conducted, using the key words 'dentigerous cyst', 'mesiodens' and 'supernumerary tooth'.

Case reports

Case 1. A 55-year-old Chinese female attended our clinic with a chief complaint of painless swelling in the palatine for a duration of 6 months. There was no history of allergic symptoms and systemic illness. On intraoral examination, a soft, fluctuant, painless swelling was palpable in the anterior portion of the hard palatine. There was no history of trauma or no dental treatment around the lesion. The maxillary incisors were sensitive to percussion, but responded positively to vitality tests. The patient's neurological exam was normal. The upper occlusal radiograph of the lesion revealed an impacted supernumerary tooth with a short root apical to the maxillary left central incisor which was associated with a well-defined radiolucent area in the anterior region of the maxilla. It also showed that the pericementum of the maxillary central incisors was continuous and intact, and the apices of the left tooth existed in the cyst. Displacement of the roots of the maxillary central incisors was evident (Fig. 1D). Computed tomography (CT) scan revealed soft tissue density measuring 1.9x1.4 cm in the midline, containing the impacted supernumerary tooth (Fig. 1A-C). A clinical diagnosis of an infected dentigerous cyst associated with a supernumerary tooth (a mesiodens) was made.

After the acute stage subsided, routine blood investigations were normal. With the patient under local anesthesia, a full-thickness mucoperiosteal flap was reflected at the right mucopalatine fold from the maxillary right second premolar tooth to the maxillary left second premolar. A cyst measuring 2 cm in diameter was enucleated. The cyst consisted of a smooth outer surface and a well-formed wall that was 1-mm thick, surrounding the impacted supernumerary tooth

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(Fig. E). During surgery, there was no connection between the supernumerary tooth and the apices of the maxillary left center incisor. Histological examination of the specimen confirmed the initial diagnosis (Fig. 1F). Postoperative antibiotics were prescribed for 5 days, and the stitches were removed after 1 week. One month later, the patient presented no complications after receiving cyst enucleation.

Case 2. A 46-year-old Chinese male attended our clinic with a chief complaint of painless swelling in the palate for a duration of one year. There was no history of trauma or no dental treatment around the lesion. Intraoral examination revealed a solitary and well-defined swelling in the right premaxillary region. There was no history of trauma, and vitality of the associated teeth was intact. The radiological examination included panoramic and occlusal radiographs that revealed one supernumerary tooth involved in a solitary, well-defined radiolucency, extending from the midline to the right maxillary lateral region, apically to the right upper central incisor (Fig. 2A and B). CT revealed a supernumerary tooth surrounded by a soft tissue mass measuring 1.6x2.04 cm in the right premaxillary region, with the palatal cortical expanded. There was no bone destruction (Fig. 2C and D). A tentative diagnosis of dentigerous cyst associated with a supernumerary tooth was made.

Routine blood investigations were normal, and the cyst was enucleated along with the supernumerary tooth under local anesthesia. The macroscopic findings revealed cystic lining attachments to the supernumerary tooth (Fig. 2E). Routine histological examination of the enucleated specimen confirmed the initial diagnosis (Fig. 2F). After surgery, antibiotics were prescribed for 5 days, and the stitches were removed after 1 week. The postoperative period of the patient was uneventful.

Case 3. A 53-year-old Chinese male attended our clinic with a chief complaint of a painless slow-growing mass on the upper lip for a duration of one year. The right upper lateral incisor had been extracted several years earlier. Intraoral examination revealed a soft, fluctuant, labial cyst-like swelling measuring 2.5x1.5 cm on the right upper lip (Fig. 3A). The maxillary right canine and maxillary right central incisor were vital and not sensitive to percussion. A panoramic radiograph revealed a relatively large and well-defined radiolucency between the maxillary right canine and the right central incisor, enveloping an unerupted horizontal supernumerary tooth (Fig. 3B). CT showed a supernumerary tooth surrounded by a soft tissue mass measuring 2.4 cm horizontally, 1.5 cm vertically and 1.3 cm sagittally, with the labial cortical bone expanded and heavily eroded (Fig. 3C-E). A tentative diagnosis of infected dentigerous cyst associated with a supernumerary tooth was made.

The patient's general health and development were normal except for this lesion. Under local anesthesia, the cyst was wholly enucleated together with the unerupted supernumerary tooth. The extracted specimen consisted of a brown cyst measuring 2.5x0.5x1.0 cm with a small monoradicular malformed supernumerary tooth (Fig. 3F). Routine histological examination of the enucleated specimen confirmed the initial diagnosis (Fig. 2F). After surgery, antibiotics were prescribed for 5 days, and the stitches were removed after 1 week. The postoperative period of the patient was uneventful.

Case 4. A 23-year-old Chinese male attended our clinic with a chief complaint of painless swelling in the palate for a duration of one year. There was no history of allergic symptoms and systemic illness. On intraoral examination, a soft, painless swelling was palpable in the anterior portion of the hard palate. There was no history of trauma or no dental treatment around the lesion. Maxillary anterior teeth were vital and not sensitive to percussion. The radiological examination including panoramic and occlusal radiographs revealed an impacted inverted supernumerary tooth with a short root apical to the maxillary right lateral incisor which was associated with a well-defined radiolucent area extending from the maxillary right first molar to the maxillary right central incisor. None of the apices of these teeth existed in the cyst (Fig. 4A and B). CT demonstrated that the radiolucent lesion measuring 1.74x1.5 cm in the right premaxillary region involved an inverted-shaped tooth-like calcified structure (Fig. 4C).

Routine blood investigations were abnormal with a hemoglobin level <68 g/l. Surgery could not be performed because of severe anemia.

Discussion

Supernumerary teeth are commonly located in the anterior maxillary region and can often cause developmental and eruption disturbances of adjacent permanent teeth, leading to crowding, displacement, diastema and, in some cases, radicular resorption and dentigerous cyst formation (6-8). The four cases described the dentigerous cyst formation involving a supernumerary tooth in the anterior maxilla. In addition, displacement of the adjacent central incisors was only observed in case 1 and none of the 4 patients had root resorption.

Dentigerous cysts are the second most common odontogenic cysts of the jaws after periapical or radicular cysts, while a dentigerous cyst associated with a supernumerary tooth is a rare entity. A review of the literature since 1988 disclosed 16 reported cases of dentigerous cysts associated with premaxillary supernumerary teeth (1,3,10-15). Our four cases bring the total number of documented cases to 20 (Table I). The reported prevalence of dentigerous cysts associated with supernumerary teeth ranges between 1 and 9.9% in the literature (6,7,9,11,16). Most reports revealed a peak incidence of dentigerous cysts in the second and third decades of life. The age range for reported cases varies widely, from 9 to 71 years of age. The mean age of the 16 previously reported cases, not including our four cases, was 30.5 years. The mean age of the present four cases was 44.3 years. Among the 20 cases summarized in Table I, the incidence is significantly higher in men (n=14) compared to women (n=6). This finding is similar to previous studies, which found that dentigerous cysts appear to have a distinct predilection for the male gender (4,17).

Dentigerous cysts associated with supernumerary teeth in the premaxilla are easily diagnosed radiographically due to their radiopaque image. Panoramic and upper occlusal radiographs are simple and inexpensive methods, which can be used to determine the location of dentigerous cysts, the structure of the impacted teeth, the influence on adjacent teeth and the resorption of adjacent roots (18). However, radiograph film has two inevitable disadvantages: i) a ghost image which

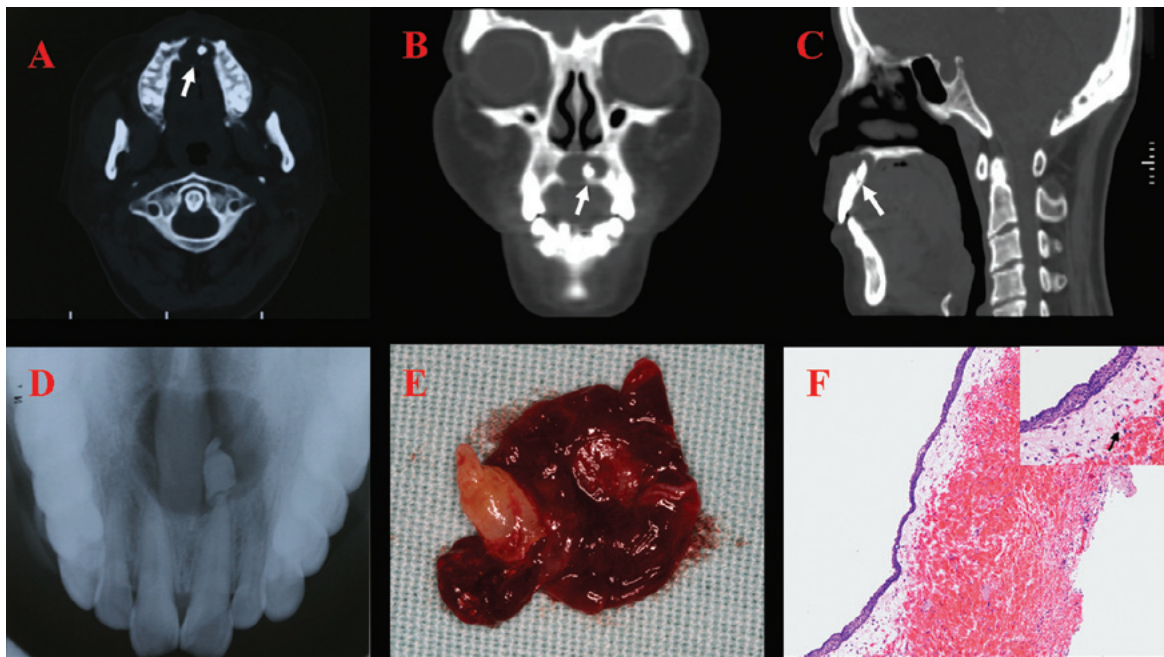


Figure 1. Case 1. (A-C) CT reveals that the cyst and the supernumerary tooth are located in the anterior maxillary region (white arrows). (A) Axial; (B) coronal; (C) sagittal planes. (D) Occlusal radiograph showing a unilocular radiolucent lesion surrounding the crown of an inverted supernumerary tooth. (E) Excised specimens show a cystic soft tissue associated with a supernumerary tooth. (F) Histopathological examination shows that the cyst was lined by non-keratinizing squamous epithelium. The connective tissue walls are cellular and composed of fibrous collagen, while some areas exhibit mononuclear inflammatory infiltration (black arrow). H&E staining; magnification, x10.

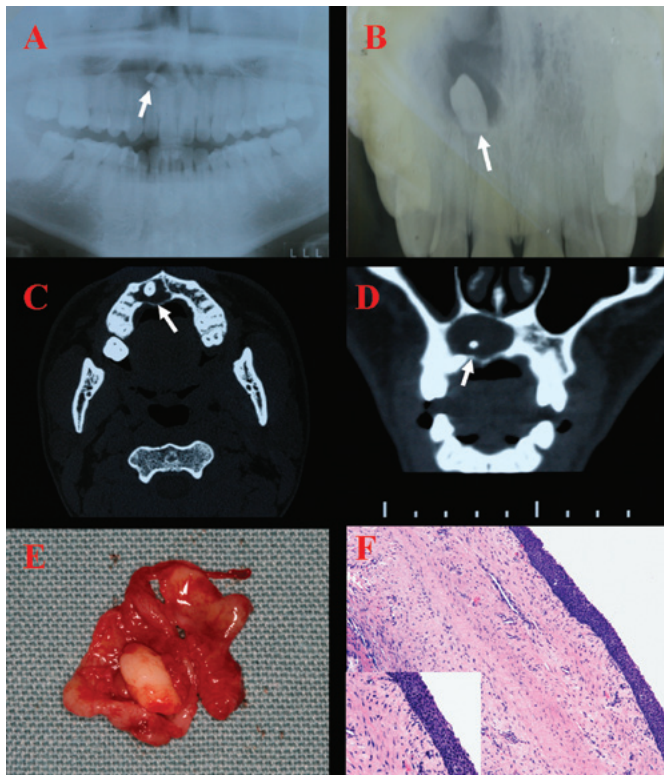


Figure 2. Case 2. (A and B) Radiological examination shows a unilocular radiolucent lesion surrounding the crown of a supernumerary tooth (arrows). (A) Panoramic radiograph; (B) occlusal radiograph. (C and D) CT reveals that the cyst and the supernumerary tooth are located in the right premaxillary region (arrows). (C) Axial; (D) coronal. (E) Excised specimens showing a cystic soft tissue associated with a supernumerary tooth. (F) Histological image indicating a cystic lining with non-keratinizing squamous epithelium. H&E staining; magnification, x10.

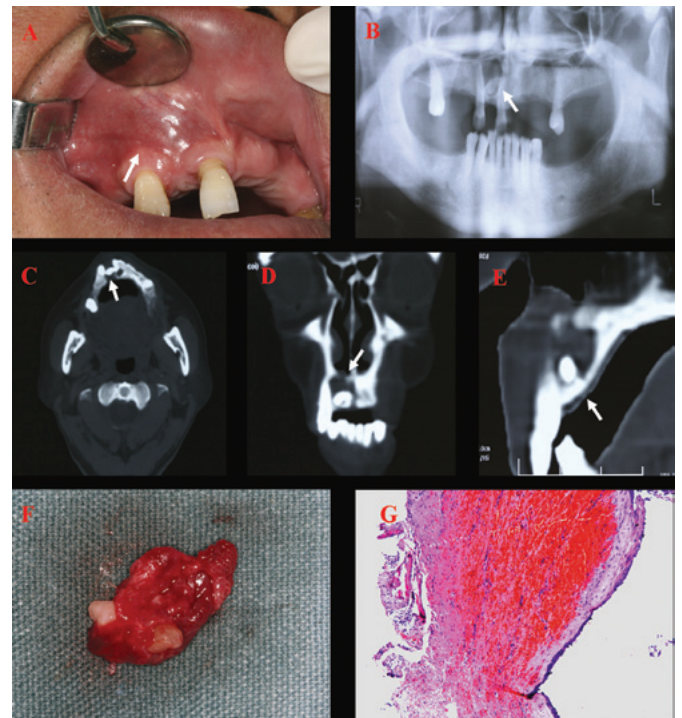


Figure 3. Case 3. (A) Intraoral view showing a labial cyst-like swelling in the anterior maxilla (arrow). (B) Panoramic radiograph shows a unilocular radiolucent lesion surrounding the crown of a supernumerary tooth between the maxillary right canine and the right central incisor (arrow). (C-E) CT reveals that the cyst and the supernumerary tooth are located in the right premaxillary region (white arrows). (A) Axial; (B) coronal; (C) sagittal planes. (F) Excised specimens show a cystic soft tissue associated with an impacted tooth. (G) Histopathological examination indicates that the cyst was lined by non-keratinizing squamous epithelium. The connective tissue walls are cellular and composed of fibrous collagen, and mononuclear inflammatory infiltration and hemorrhage are exhibited. H&E staining; magnification, x10.

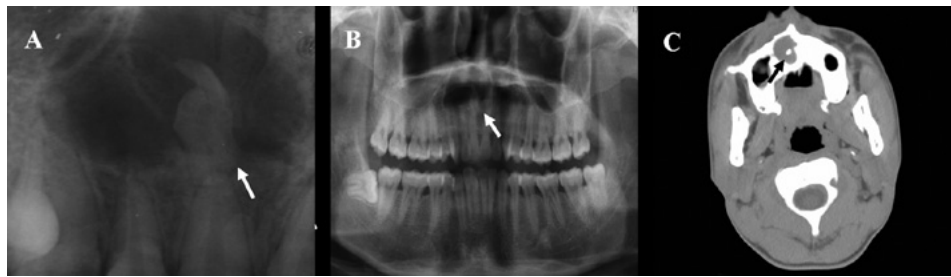


Figure 4. Case 4. (A) Occlusal radiograph shows a unilocular radiolucent lesion surrounding the crown of an inverted supernumerary tooth. (B) Panoramic radiograph shows a unilocular radiolucent lesion surrounding the crown of a supernumerary tooth extending from the maxillary right first molar to the right central incisor. (C) Axial non-contrast CT reveals that the cyst involving an impacted tooth is located in the right premaxillary region.

Table I. Documented cases in the literature of dentigerous cysts associated with supernumerary teeth.

Authors/Reference	Patient age (years)	Gender	Site (premaxilla region)	Symptoms	Treatment
Lustmann and Bodner (3)	9	Female	Right central incisor	-	Enucleation
Lustmann and Bodner (3)	12	Male	Left central and lateral incisor	-	Marsupialization
Lustmann and Bodner (3)	37	Male	Entire premaxilla	Swelling, pain	Enucleation
Lustmann and Bodner (3)	38	Male	Midline to the left, maxillary second premolar	Swelling	Enucleation
Lustmann and Bodner (3)	68	Female	Midline to the left maxillary premolar	Swelling, pain	Enucleation
Lustmann and Bodner (3)	71	Female	Left premaxillary region	Asymptomatic	Enucleation
Awang and Siar (6)	34	Male	Midline to the upper left first premolar	Swelling	Enucleation
Awang and Siar (6)	24	Female	From the upper right canine to the upper left central incisor	Swelling	Enucleation
Scolozzi, <i>et al</i> (21)	42	Male	Premaxilla	Swelling	Enucleation and autogenous cancellous bone graft from the iliac crest
Dinkar, <i>et al</i> (15)	14	Female	Maxillary anterior region	Pain, swelling	Enucleation
Khan, <i>et al</i> (20)	24	Male	Incisor region	Swelling	Enucleation
Gulses, <i>et al</i> (13)	10	Male	Right central incisor	Asymptomatic	Enucleation
Kumar, <i>et al</i> (10)	14	Male	Central incisor region	Swelling	Enucleation
John, <i>et al</i> (7)	22	Male	From the maxillary right central incisor to the right canine	Swelling	Enucleation
John, <i>et al</i> (7)	24	Male	Maxillary anterior region	Swelling	Enucleation
John, <i>et al</i> (7)	46	Male	Maxillary anterior region	Pain, swelling	Enucleation
Present study					
Case 1	55	Female	Central incisors	Swelling	Enucleation
Case 2	46	Male	Midline to the upper right canine	Swelling	Enucleation
Case 3	53	Male	From the maxillary right central incisor to the right canine	Swelling	Enucleation
Case 4	23	Male	From the maxillary right first molar to right central incisor	Swelling	-

cannot reflect the 3-dimensional structure of the lesion owing to a low resolution ratio, and ii) different degrees of distortion or amplification. Therefore, CT is necessary and valuable to obtain more information concerning the lesion. CT can be used, not only to identify the pathology of the dentigerous

cyst and the exact location of the impacted tooth, but also to determine the full extent of the lesion, thus contributing to proper treatment planning as well. Meanwhile, CT can also be used to identify erosion of cortical bone and invasion into adjacent soft tissues. In the present four cases, the

dentigerous cyst with the supernumerary tooth appeared to be within the anterior maxilla in the panoramic radiograph or/and upper occlusal radiograph. In order to determine the exact location of the impacted tooth and the degree of bone destruction, CT images were scrutinized. Therefore, to ensure appropriate treatment decisions and follow-up based on accurate information regarding dentigerous cysts associated with supernumerary teeth, we recommend panoramic radiograph and/or upper occlusal radiograph as a first-line diagnostic tools, and further evaluation of the lesion by CT examination.

Enucleation is the standard treatment for a dentigerous cyst along with extraction of the associated supernumerary tooth (9,10). Among the documented cases in the literature, the second case (Table I) was treated by marsupialization because of the intimate relation of the lesion to the apices of the incisor teeth. Marsupialization is recommended for a large cyst when a single draining may not be effective and complete removal of the surrounding structure is not desirable (19). For a large cyst, Scolozzi *et al* recommended enucleation followed by an immediate bone grafting procedure (21). In the present four cases, surgical removal of the impacted supernumerary tooth and enucleation without using bone grafting of the associated cyst were performed.

A broad range of conditions may lead to a clinical presentation of painless swelling along the lingual surface of the palate or on the upper lip. Differential diagnosis of a median palatine cyst, nasopalatine duct cyst, radicular cyst, odontogenic keratocyst (OKC) or adenomatoid odontogenic tumor (AOT) was considered in our cases. Median palatine cysts and nasopalatine duct cysts are not associated with non-vital teeth as non-odontogenic cysts of the hard palate (20,21). Most radicular cysts appear as round or pear-shaped, unilocular, lucent lesions in the periapical region, and the associated tooth usually has a deep restoration or large carious lesion radiographically (21). Approximately 40% of OKCs contain an impacted tooth, and the lumen of the cyst often contains 'cheesy' material and has a parakeratinized epithelium lining. They are more likely to show aggressive growth than other odontogenic cysts and may have undulating borders and a multilocular appearance upon radiography (22). Approximately 75% of cases are associated with an unerupted tooth, and the most common location is in the anterior maxilla. AOTs are more common in young people, affect females more than males and, most importantly, the radiolucency in cases of AOTs extends apically beyond the cemento-enamel junction (18).

In conclusion, dentigerous cysts arising from impacted supernumerary teeth in the anterior maxilla should be considered in the differential diagnosis for painless swelling along the lingual surface of the palate or on the upper lip. To prevent the development of a dentigerous cyst and to avoid unwanted effects on adjacent teeth, early detection consisting of a thorough clinical and radiographical examination is necessary for accurate diagnosis and proper treatment planning.

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