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Upper lip swelling caused by a large dentigerous cyst

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Abstract Swelling of the upper lip can result from various diseases such as salivary tumors, infectious and inflammatory diseases and cysts. Among the latter, dentigerous cysts, typically involving unerupted teeth, are sometimes associated with supernumerary teeth in the maxillary anterior incisors region called the mesiodens. We report an unusual case of a large dentigerous cyst associated with an impacted mesiodens in a 42-year-old male who presented with a slow-growing swelling in the upper lip.

Keywords Upper lip · Dentigerous cyst · Mesiodens

Introduction

Dentigerous cysts, also known as follicular cysts, are developmental cysts formed by fluid accumulation between the reduced enamel epithelium and the enamel surface. They surround the crown of an unerupted tooth, most often the mandibular third molars and maxillary canines. They are occasionally associated with supernumerary teeth. First named by Bolk in 1917, mesiodentes are the most common supernumerary teeth [4]. They are typically found in the midline of the anterior maxilla, in

the palate area between the apical and cervical regions of the permanent incisors, and most of them remain impacted. The incidence of mesiodentes as reported in the literature ranges from 0.15 to 4 percent [16]. Although the permanent dentition is most often affected, deciduous supernumerary teeth are occasionally reported.

We report a case of a relatively large dentigerous cyst associated with an unerupted mesiodens in a 42-year-old male who presented with a slow-growing mass in the upper lip. The cyst was treated by enucleation and immediate autogenous cancellous bone graft.

Case report

In October 2001, a 42-year-old Ethiopian male was referred to our Maxillofacial Unit by the ENT Department for investigation of a slow-growing mass on the upper lip. The mass had been present for several years and resulted in malpositioning of the central maxillary incisors. Intraoral examination revealed a soft, labial cyst-like swelling measuring 3.5×2.0 cm (Fig. 1). Maxillary anterior teeth were vital and not sensitive to percussion, except for the right central incisor. A panoramic radiograph revealed a relatively large and well-defined radiolucency extending from the right canine to the left lateral maxillary incisor, enveloping an unerupted mesiodens. Computerized tomography showed a cystic lesion measuring 3.4 cm horizontally, 2.5 cm vertically and 2.3 cm sagittally, with the labial and palatal cortical bone expanded and eroded (Fig. 2). The right maxillary canine, the right lateral and central incisors and the left central and lateral incisors were treated endodontically the day before the operation because of their intimate relationship with the lesion. Under general anesthesia, the cyst was wholly enucleated together with the unerupted mesiodens (Fig. 3). The bony defect was filled with autogenous cancellous iliac bone graft. The post-operative course was uneventful.

The gross specimen consisted of a brown cyst measuring 2.7×0.4×2.0 cm with a small monoradicular

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Fig. 1 Intraoral view showing a labial cyst-like swelling

malformed and dwarfed supernumerary tooth. The cyst was inserted on the cervical zone of the tooth. Microscopic examination showed a cyst lined by an epithelium with cuboidal and columnar cells resting on a flat basal lamina. In focal areas, non-keratinizing stratified squamous and transitional epithelium was also observed (Fig. 4). The connective tissue wall consisted of fibrous tissue without inflammatory infiltrate. The final diagnosis was that of a dentigerous cyst associated with mesiodens.

Discussion

Swelling of the upper lip can result from different diseases ranging from infectious diseases (abscesses or cellulites), allergic diseases (angioedema), neoplasms,

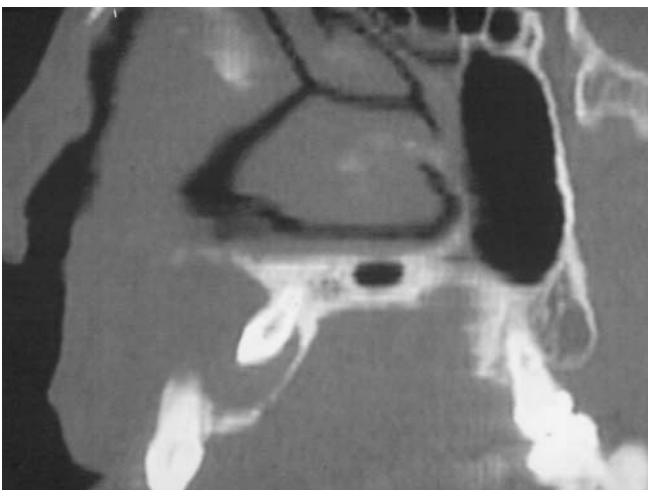


Fig. 2 Sagittal CT image showing the mesiodens impacted within the palatal bone. The labial cortical bone is completely eroded by the cyst



Fig. 3 Intraoperative view showing the cyst bulging from the anterior maxilla

especially of salivary origin (canalicular adenoma, pleomorphic adenoma), granulomatous diseases (isolated Miescher's granulomatous cheilitis or as part of Melkersson-Rosenthal syndrome, Crohn's disease and sarcoidosis), inflammatory diseases (cheilitis glandularis) and cysts (epidermoid or epidermal inclusion cyst, nasolabial cyst, aggressive odontogenic cyst such as keratocysts extending through the bone cortex and dentigerous cysts).

Dentigerous cysts—typically associated with impacted teeth—are sometimes associated with supernumerary teeth in the maxillary anterior incisors region called the mesiodens. The most common pathological findings reported to be associated with mesiodens are retention of adjacent incisors, malposition and diastema formation [16, 2]. Contrarily, development of a dentigerous cyst on an impacted mesiodens is relatively rare

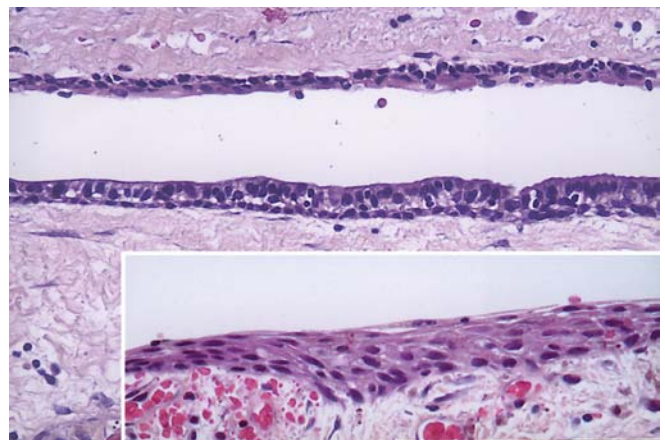


Fig. 4 Microscopic examination showing a portion of the cyst lined by an epithelium with cuboidal and columnar cells resting on a flat basal lamina. *Inset* shows an area of the cyst lined by nonkeratinizing stratified squamous epithelium. (hematoxylin-eosin, original magnification $\times 20$)

[16]. Stafne [15] first described the association of dentigerous cyst with the supernumerary maxillary teeth and found an incidence of 5.5% among 180 patients with 200 supernumerary teeth occurring in the region of the upper central incisors. Bodin reviewed 350 supernumerary teeth in 290 patients and found a widened pericoronal space in 12% of cases, a rather high frequency that probably indicates that the risk of cyst formation with supernumerary teeth should not be disregarded [2]. Primosch reported an enlarged dental sac in 30% of cases, but a histological diagnosis of dentigerous cyst in only 4 to 9% of cases [13]. Recently, von Arx clinically and radiologically reviewed 113 mesiodentes in 90 patients and found a follicular cyst (pericoronal space > 5 mm on the conventional radiograph) in only three patients [16]. Lustmann reviewed the literature from 1900–1988 and found 42 cases of dentigerous cyst associated with supernumerary teeth [9].

In our review of the literature, we have found only eight cases of large dentigerous cysts (> 3 cm) associated with a mesiodens (Table 1), and none was treated by

bone graft. While some authors propose enucleation without using bone grafting for treating large mandibular cysts [5], enucleation followed by immediate bone grafting procedure is the recommended treatment for large jaw cysts with supernumerary or wisdom teeth [3, 14]. Enucleation guarantees the complete removal of the lesion and provides the entire cyst for the pathological examination. Different grafting materials have been used to fill the residual cavity, including alloplastic bone, allogenic bone or xenogenic bone or a combination of these materials, but autogenous cancellous grafts produce the most favorable and predictable results, both experimentally and clinically [3, 14]. They also possess excellent space maintaining properties, a high concentration of osteoinductive cells and act as an osteoconductive scaffold during new bone formation. The iliac crest provides large amounts of cancellous bone and therefore is the site of choice to harvest graft, even though this has the disadvantage of involving a second surgical site procedure with increased morbidity [3, 14].

Table 1 Large dentigerous cysts (> 3 cm) associated with impacted mesiodens

Authors	Gender	Age (years)	Localization	Duration	Symptoms	Size (cm)	Therapy
Stafne (1931) [15]	–	–	Palate	–	–	Entire palate	–
Haag, Frank and Nicolas (1966) [7]	F	13	Between the right central incisor and the left canine	15 days	Left hemifacial pain and swelling of the left paranasal region	3×3.5 cm	Enucleation
Ciola and Catena (1972) [6]	M	46	Apical to the maxillary anterior teeth between the two canines	–	“Front teeth sensitive to cold air.”	Large	Enucleation
Archer (1975) [1]	1) F	12	Between from left central incisor and the first right molar.	–	–	Large	Marsupialization
	2) –	–	Between the two canines	–	–	Large	Marsupialization
Papadopoulos (1981) [12]	F	40	From the upper right central incisor to the upper left second premolar	10 years	Repeated swelling over the upper left central and lateral incisor region (buccal and palatal)	3×2×2 cm	Enucleation
Most and Roy (1982) [10]	M	30	Entire right antrum and one half of the left antrum	–	Pain and swelling of the right maxillary mucogingival fold area	9×1.5×0.8 cm	Enucleation
Lustmann and Bodner (1988) [9]	1) M	37	Premaxilla	6 days	Acute painful swelling of the premaxillary region. Asymptomatic	Entire premaxilla	Enucleation
Noor Awang and Huat Siar (1989) [11]	2) F	71	Premaxilla	–	–	Large	Enucleation
	1) M	34	From the maxillary midline to the upper left first premolar	1 month	Swelling in the left premaxilla.	3×2 cm labially	Enucleation
	2) F	24	From upper right canine to the upper left central incisor	–	Swelling obliterating the upper labial sulcus	2×2 cm (palate) 3×3cm	Enucleation
Present case	M	42	Premaxilla	Several years	Malposition of the central incisors and swelling of the upper lip	3.4×2.5×2.3 cm	Enucleation and autogenous cancellous bone graft from iliac crest

In our opinion, marsupialization should be limited to those particular cases in which teeth have to be conserved and brought into the dental arch or in an unhealthy or debilitated patient.

In conclusion, large dentigerous cysts arising from unerupted mesiodens should be kept in mind in the differential diagnosis of upper lip swelling, particularly if associated with dental anomalies of the maxillary incisors such as malposition and diastema. To prevent the development of a dentigerous cyst and to avoid unwanted effects on neighboring teeth, supernumerary anterior teeth should be removed surgically or observed with regular radiographic controls [8].

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