



Dentigerous Cyst Associated with an Impacted Mesiodens: A Rare Case Report with Review of Literature

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Abstract:

Dentigerous cyst develops around an unerupted tooth by accumulation of fluid between reduced enamel epithelium (REE) and the enamel, or between layers of REE. These cysts are second most common odontogenic cysts after radicular cyst. Dentigerous cysts are commonly associated with the 3rd molar teeth of the mandible. Dentigerous cysts around the supernumerary teeth account for 5% of all dentigerous cysts, most developing around a mesiodens in the anterior maxilla. 'Mesiodens' are the most common supernumerary teeth situated between the maxillary central incisors. Herein, we review the literature spanning the past 30 years concerning dentigerous cysts associated with supernumerary teeth in the anterior maxilla, and present a case of a dentigerous cyst in association with an impacted mesiodens in a 23-year-old male patient with emphasis given to the clinicopathological characteristics of this type of dentigerous cyst.

Keywords: Anterior Maxilla, Dentigerous Cyst, Enucleation, Mesiodens, Supernumerary Tooth

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1. Introduction

Dentigerous cyst is a common developmental odontogenic cyst caused by fluid accumulation between the reduced enamel epithelium and the enamel surface of a formed tooth¹. This cyst most frequently occurs in patients between 10 and 30 years of ages and there is a greater incidence in males with a 1.6:1 ratio. Most of the dentigerous cysts are associated with mandibular third molars, followed by maxillary third molars, maxillary canines, premolars and rarely supernumerary teeth². Dentigerous cysts around supernumerary teeth account for 5% of all dentigerous

cysts, most developing around a mesiodens in the anterior maxilla^{3,4}.

A mesiodens, a kind of supernumerary tooth, is located in the maxillary central incisor region. A mesiodens has an overall prevalence of 0.15-1.9%. A mesiodens can occur individually or in multiples and often does not erupt. Mesiodens sometimes bring about median diastema, delayed eruption of the permanent central incisors, alteration of the position of the permanent incisors, and dentigerous cyst formation^{4,5}.

In the present study, we review the literature spanning the past 30 years concerning dentigerous cysts associated with supernumerary teeth in the anterior maxilla,



and present an additional case with emphasis on the clinicopathological characteristics of this type of dentigerous cyst. Pubmed search from 1988 to 2017 was conducted, using the key words 'dentigerous cyst', 'supernumerary tooth', 'mesiodens' and 'anterior maxilla'.

2. Case Report

A 23-year-old male patient presented to the Outpatient Department of the Narayana dental college, nellore, India, with a swelling in the maxillary anterior region, which was gradually increasing in size over the past 6 months. On general examination, the patient was apparently healthy. There was no significant past medical history. Clinical examination of the oral cavity revealed a painless, hard swelling, which had extended onto the maxillary alveolar process (Figure 1). The routine laboratory tests were within normal limits. A diagnostic orthopantomograph (OPG) revealed a radiolucent lesion in the alveolar process of the left anterior maxilla (Figure 2).

A radio opaque area resembling to supernumerary tooth (mesiodens) was observed within the lesion just above the roots of the right maxillary lateral incisor. There was an evidence of divergent roots of central incisors but no resorption of the roots of the associated permanent teeth. The lesion was well-defined, extending from the mesial aspect of left second premolar tooth region to right incisor tooth region.

The buccal cortical plate showed slight expansion and there was no sign of any acute periodontal condition or carious lesions. After a clinical and radiological examination, a provisional diagnosis of dentigerous cyst was made, and also, large periapical cyst, odontogenic keratocyst, central giantcell granuloma, adenomatoid odontogenic tumor, and ameloblastic

fibroma were considered in the differential diagnoses.

Under local anesthesia, the cyst was wholly enucleated together with radio opaque mass. The extracted specimen consisted of a brown cyst measuring 2.5x0.5x1.0 cm with a small mono radicular malformed supernumerary tooth (Figure 3).

Routine histological examination of the enucleated specimen revealed a thin fibrous cystic wall lined by two to three layers of thick non-keratinized, stratified, squamous epithelium, with islands of odontogenic epithelium. The connective tissue showed a slight inflammatory cell infiltrate, which confirmed the diagnosis of a dentigerous cyst, associated with the mesiodens (Figure 4). The patient was on regular follow-up and no complications developed within the six-month follow-up.

3. Discussion

Dentigerous cysts account for approximately 16.6% of all jaw cysts. About 95% of these cysts involve permanent dentition and only 5% are associated with supernumerary teeth^{3, 4}. The exact etiology of supernumerary teeth is still unknown but it can be a result of local, independent or conditioned hyperactivity of dental lamina. Mesiodens, first named by Bolk in 1917, is the most frequent type of supernumerary tooth and is situated in the maxillary anterior incisors region. It is a rare entity with a prevalence of 0.15–1.9% in the general population and a slight male predilection^{4, 5}.

A review of the literature since 1988 disclosed 24 reported cases of dentigerous cysts associated with anterior maxillary supernumerary teeth (mesiodens)¹⁻¹⁴. Our case brings the total number of documented cases to 25 [Table 1]. 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



8 to 71 years of age. The mean age of the 24 previously reported cases, not including our case, were 33 years. Among the 24 cases summarized in Table I, the incidence is significantly higher in men (n=18) compared to women (n=7). This finding is similar to previous studies, which found that dentigerous cysts appear to have a distinct predilection for the male gender^{1, 5}.

A wide range of conditions may lead to a clinical presentation of painless swelling in the anterior maxilla. Differential diagnosis of a median palatine cyst, nasopalatine duct cyst, radicular cyst, odontogenic keratocyst (OKC) or adenomatoid odontogenic tumor (AOT) was considered in our case. Median palatine cysts and nasopalatine duct cysts are not associated with non-vital teeth as non-odontogenic cysts of the hard palate^{6, 7}. 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 radiographically⁶. 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 radiography¹. 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 junction¹.

Radiologically, the dentigerous cyst appears as a well-circumscribed, unilocular radiolucency around the crown of an impacted tooth. The radiolucency usually arises in the cemento-enamel junction of the tooth^{2, 8}. The internal aspect of the cyst is

completely radiolucent except for the crown of the involved tooth. One of the most difficult conditions to distinguish in the differential diagnosis is hyperplastic follicle. Other conditions that must be excluded in the diagnosis are odontogenic keratocyst, ameloblastic fibroma, and cystic ameloblastoma³.

Water's, panoramic, and skull radiography are simple and inexpensive methods that can be used in daily practice. Although the structure of a tooth can be clearly detected on panoramic radiographs, they are inadequate for localizing maxillary ectopic teeth due to their inherently less sharp image and ghost image. CT scan provides superior bony detail, allowing for the visualization of the size and extent of the lesion with determination of orbital or nasal invasion or involvement⁴.

The standard treatment for a dentigerous cyst is enucleation and extraction of the cyst-associated impacted or unerupted tooth⁴. 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 desirable⁵. For a large cyst, Scolozzi *et al* recommended enucleation followed by an immediate bone grafting procedure⁶. In the present case, surgical removal of the impacted supernumerary tooth and enucleation without using bone grafting of the associated cyst was performed.

4. Conclusion

In summary, dentigerous cysts arising from impacted supernumerary teeth should be considered in the differential diagnosis for painless swelling in the



anterior maxilla. To prevent the development of a dentigerous cyst and to avoid unwanted effects on adjacent teeth, early detection consisting of a thorough clinical and radiological examination is necessary for accurate diagnosis and proper treatment planning.

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Table 1: Reported cases in the literature of dentigerous cysts associated with supernumerary teeth.

Sr. No.	Authors	Age of occurrence(years)	Gender
1	Lustmann and Bodner(1988) ¹³	09	Female
2	Lustmann and Bodner	12	Male
3	Lustmann and Bodner	37	Male
4	Lustmann and Bodner	38	Male
5	Lustmann and Bodner	68	Female
6	Lustmann and Bodner	71	Female
7	Awang and Siar(1989) ¹⁴	34	Male
8	Awang and Siar	24	Female
9	Scolozzi, et al(2005) ⁶	42	Male
10	Dinkar, et al(2007) ³	14	Female
11	Khan, et al(2008) ⁷	24	Male
12	Shun Y (2008) ¹⁰	08	Male
13	Jiang, Q, et al(2011) ¹	55	Female
14	Jiang, Q, et al	46	Male
15	Jiang, Q, et al	53	Male
16	Jiang, Q, et al	23	Male
17	Hosseini, et al (2011) ²	18	Female
18	Neha, et al (2012) ¹	55	Male
19	Neha, et al	46	Male
20	Neeraj K Agrawal (2012) ⁹	11	Male
21	Shah, et al (2013) ⁵	18	Male
22	Patel K, et al (2013) ¹¹	30	Male
23	Kim KS & Mun SK (2013) ¹²	35	Male
24	Hasan, et al (2014) ⁸	32	Male
25	Present case (2018)	23	Male

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Figures & Legends:



Figure 1: Photomicrograph showing diffuse intra oral swelling obliterating labial vestibule



Figure 2: OPG showing a large unilocular radiolucency in the anterior maxilla with mesiodens



Figure 3: Excised specimen showing a cystic soft tissue associated with mesiodens.

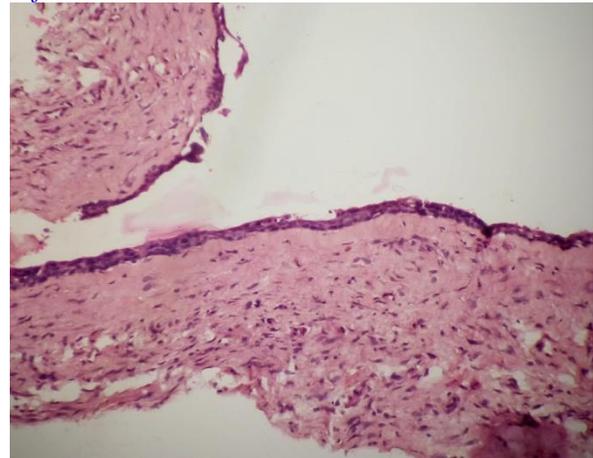


Figure 4: Photomicrograph showing cystic wall with non-keratinizing squamous epithelium (H&E staining, x20)