Case Report

Dentigerous Cyst of Maxilla Involving Multiple Impacted Teeth: A Rare Case Report
Praveen Ganesh, Venkatesh Anehosur, Abhijit Joshi, Gopalkrishnan K

Abstract
Dentigerous cyst is a developmental epithelial odontogenic cyst which arises from remnants of tooth forming organ. It is the second most common type of odontogenic cysts occurring during the second and third decade of life with involvement of an impacted tooth. The site predilection is usually the molar ramus area. This case report describes a rare incidence of involvement of multiple impacted teeth in the maxillary anterior region.

Keywords: Dentigerous Cyst; Bone Cysts; Jaw Cysts; Odontogenic Cysts; Maxilla; Impacted; Tooth; Unerupted; Enucleation.

Introduction
Odontogenic cysts comprise a sizeable percent of jaw swellings. The cyst cavity is lined by epithelial cells derived from the reduced enamel epithelium of the tooth forming organ. Dentigerous cyst are the second most common type of odontogenic cysts accounting for 49% of all cystic lesions. They are the most frequent developmental odontogenic cyst affecting the permanent teeth. Dentigerous cysts arise from degenerative changes in the reduced enamel epithelium with an accumulation of fluid between the crown of the tooth and the lining epithelium.

Dentigerous cysts are more common in male subjects, occurring most often in the second and third decades. About 70% of these cysts occur in mandible and 30% in maxilla. It is more commonly associated with impacted mandibular third molars. The most common site of occurrence is molar-ramus area. In maxilla the incidence is rare. It arises in relation to maxillary canine as it is the second most common impacted tooth.

Dentigerous cyst commonly present as asymptomatic swelling unless there is acute inflammatory exacerbation, thus explaining the fact that these lesions are often detected only during routine radiographic examination. They are generally discovered when radiographs are taken to investigate a failure of tooth eruption, missing tooth, or malalignment of teeth. The radiographic pattern is characterized by a symmetric; well defined, unilocular radiolucent lesion surrounding the crown of unerupted tooth.

Surgical treatment of cystic lesions is indicated to prevent local extension from compromising adjacent osseous tissue and later impinging on anatomical structures.

This type of cyst usually encircles a single molar crown in cystic lumen. In this report we describe a rare case of Dentigerous cyst of anterior maxilla which was an incidental radiographic finding in a 14 year old patient which had certain unusual features. The cyst was found involving more than one impacted anterior teeth in the maxilla.

Case report
A 14 year old boy presented with a chief complaint of decayed maxillary anterior teeth. The past medical history was non-contributory. Examination revealed retained primary maxillary right central incisor and canine, primary left maxillary central, lateral and canine, partially erupted right permanent lateral incisor (Figure 1a). Routine topographic occlusal radiograph and an orthopantomograph was done which revealed a well-defined radiolucent lesion involving the crown of impacted right maxillary permanent central incisor and canine in the alveolar process of anterior maxilla.

Radiologically the lesion was measuring about 3 X 2 cm in largest dimension, extending from periapical region of right permanent maxillary premolar till the left permanent maxillary premolar region running along the impacted left maxillary central, lateral and canine on the opposite side, superiorly towards nasal spine and...
floor of the nose (Figure 1b). There was no evidence of resorption of the roots of associated permanent teeth. An aspiration biopsy of the swelling done using a 20 gauge needle revealed the presence of clear straw colored fluid. Based on these findings, a provisional diagnosis of dentigerous cyst of anterior maxilla was considered. Possibility of retrieving the impacted teeth in arch was ruled out after consultation with orthodontist.

The planned surgical procedure was done under local anesthesia. Crevicular incision was placed extending from right maxillary premolar to left maxillary premolar. Two vertical releasing incisions were placed on either side and mucoperiosteal flap was raised. A bony window was created on the labial aspect of maxilla. Surgical exposure of the lesion was done which revealed involvement of all impacted teeth associated with the lesion (Figure 1c). Enucleation of the lesion along with impacted right maxillary central, canine and left maxillary central, lateral and canine along with partially erupted maxillary lateral incisor was done (Figure 1d). Thorough curettage and irrigation with copious saline and betadine irrigation was carried out. The wound was packed with betadine soaked gauge and closed using 4-0 vicryl sutures. The procedure was well tolerated by the patient and he was discharged on the same day with prescription for five day course of oral antibiotics and analgesics. The pack was removed on the second post-op day through the releasing incision which was re-sutured under local anesthesia. The surgical specimen was sent for histopathological examination.

Gross examination showed that the soft tissue specimen had cystic quality and was found attached to the crown of maxillary central incisor and canine. The cystic mass was measuring about two centimeter in length and two centimeter in breath. It was creamy white in colour and had a fluctuating consistency with irregular surface and well defined borders. Histopathological examination revealed stratified squamous epithelium having three to four layers of cell thickness and surrounded by fibre-vascular connective tissue capsule. There was evidence of epithelial proliferation towards the connective tissue. In general, it was seen that the infiltrations were mostly composed of macrophages and lymphocytes (Figure 1e).

Regular clinical and radiographic evaluation revealed normal healing with no evidence of recurrence. A prosthodontic consultation was taken for the replacement of his missing teeth. As the patient had malalignment of his lower teeth, he underwent orthodontic treatment for six months for the correction so as to facilitate denture placement. As evident in the panoramic radiograph his mandibular teeth were brought into occlusion (Figure 1f). Patient was rehabilitated with a removable partial denture.

Discussion
Dentigerous cyst is a common developmental odontogenic cyst which encloses the crown of unerupted tooth and is attached at the cemento-enamel junction. It is also called follicular cyst as it arises between the enamel epithelium and the tooth or from remnants of odontogenic epithelium. Dentigerous cysts have site predilection for molar ramus area as the mandibular third molars are the most commonly impacted teeth. The second most commonly involved tooth being maxillary canine. The present case report highlights the fact that apart from involvement of maxillary canine, the cyst was also found to involve the central incisors, which is uncommon. They commonly present as solitary lesions. The exact histogenesis remains unknown, but most authors favor a development origin. It is usually found involving the crown of a single tooth, most commonly impacted mandibular third molar or maxillary canine with the crown of the involved tooth present within the cystic lumen. They progress slowly and may exist for several years without being noticed. Because the histopathological appearance of the lining epithelium is not specific, the diagnosis relies on the radiographic and surgical observation of the attachment of the cyst to the cement-enamel junction.

Studies state that 1.44% of impacted teeth may undergo dentigerous cyst transformation. The involvement of central incisor is unusual with studies reporting an incidence of 0.1 to 0.6% and 1.5% by Daley, Wysocki and Shear respectively. Since dentigerous cysts develop after the tooth crown is formed, two speculations could explain the pathogenesis of the multiple tooth involvement; one is a fusion between two adjacent cyst linings. The second theory being fusion between the linings of preexisting cyst with the lining of reduced
enamel epithelium surrounding the adjacent tooth. The other possibility is of the cyst arising as a result of periapical inflammation from any source but usually from a non-vital deciduous tooth which spread to involve the follicles of the unerupted permanent successors, the inflammatory exudates causing separation of the reduced enamel epithelium from the enamel with resultant cyst formation. This proposes the existence of two types of dentigerous cysts, one developmental and the other inflammatory in nature. Considering the decayed, retained deciduous teeth in our case we take an inflammatory origin of the cyst formation.

Figure 1: The preoperative photograph showing mixed dentition (a) and the orthopantomograph showing multiple impacted teeth (b). The intraoperative photograph (c) and enucleated cyst with associated teeth (d). The histopathological photomicrograph showing a non-keratinized stratified squamous epithelium (e). The postoperative orthopantomograph after six months following orthodontic therapy showing well aligned lower anterior (f).

A Dentigerous cyst involving two or more impacted teeth is very rare. Detailed review of literature revealed only very few cases of the cyst involving multiple teeth (Table 1). In a unique case report, cyst was found to be enclosing three permanent maxillary teeth; the central incisor, lateral incisor and canine. Majority of the patients showed the common site of involvement as mandible. None of the case reports had involvement of more than three teeth and association of single cystic lining with multiple teeth has not been substantiated.

In the present case, a dentigerous cyst involving the crowns of right maxillary canine and maxillary central incisor with the possible involvement of the other side maxillary central incisor was considered preoperatively. The involvement of other impacted teeth, the right maxillary lateral incisor, left maxillary lateral incisor and canine was evident clinically. All the maxillary anterior teeth were involved which is a rare case scenario. However the exact tooth of origin could not be well made out. Surgical excision and pathologic analysis of the lesion is essential for the definitive
diagnosis. Pathological report confirmed that the cystic lining was associated with maxillary central incisors and maxillary canine.

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Table 1: Detailed review of literature of few cases of the cyst involving multiple teeth.

To conclude, this case report highlights a rare variety of dentigerous cyst with multiple teeth involvement and of which the exact tooth of origin could not be made out. As such, this remains a rare case report.

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References

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