Case Report
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Dentigerous Cyst Associated with Compound Odontome in the Anterior Region of Maxilla – A Case Report

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Abstract

Dental professionals encountered tumor and cystic lesions in their daily practice. However, a concurrent lesion are quite rare and poses diagnostic challenge. Odontoma are the most common odontogenic tumor of the jaw. Although transformation from cyst to tumor is quite common (orgin of ameloblastoma from a dentigerous cyst) but tumor to cyst is very rare[1]. Here, we report a case of a dentigerous cyst associated with odontome and an unerupted tooth in the anterior region of the maxilla.

Keywords: Compound Odontome, Dentigerous Cyst, Odontogenic Tumor

1. Introduction

World Health Organization (WHO) classification (2017) of odontogenic tumors, classified odontoma as benign mixed odontogenic tumor. It is one among the commonest benign mixed odontogenic tumor arising from an alteration of differentiated epithelial and mesenchymal odontogenic cells and it accounts for about 35-76% of odontogenic tumors[1,2]. In common there are 2 types of odontomas-complex and compound odontomas. One of the developmental jaw cyst that is associated with the crown of an unerupted/impacted tooth is dentigerous cyst and is most often associated with mandibular third molars, maxillary canines, mandibular second premolars, and maxillary third molars. Less frequently, they can be found in relation to supernumerary teeth, odontome and unerupted deciduous teeth. Simultaneous occurrence of dentigerous cyst and odontome are rare entities and they cause significant jaw destruction and represent a diagnostic challenge.[3] In this article, we had discussed a report of case with dentigerous cyst associated with compound odontome in the anterior region of the maxilla.

2. Case Report

A female patient of 55 years old reported to our institution with the chief complaint of loosening of prosthesis and swelling for more than 6 months in left upper front tooth region. The patient had difficulty in mastication and primarily which affects her esthetics. There was no contributing medical and surgical history. During her 3rd visit she undergone extraction and prosthetic rehabilitation. The patient had no deleterious oral habits.

On extra oral examination, the face appeared symmetrical, with no deviation of the temporomandibular joint while opening and closing the mouth without any tenderness and clicking. No lymphadenopathy in submandibular and submental lymph nodes.

On intraoral examination, there were 2 bony spicules like projections seen in relation to 21, 22, 23 edentulous regions measuring about 0.5×0.5 cm in dimensions. The color was normal and similar to that of normal mucosa. The cortical expansion was seen in the buccal and palatal aspects of the 21, 22, 23 regions. On palpation, all the inspectory findings were confirmed and the projections were sharp

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and non-tender. The cortical expansion was noted along the buccal and palatal aspect of 21, 22, 23 regions and hard in consistency (Figure 1(a) and Figure 1(b)).

Occlusal-Maxillary radiograph showed multiple radio-opaque tooth-like structures in the alveolar crest region of anterior maxilla suggestive of compound odontome (Figure 1(c)). OPG (Orthopantomagraph) showed multiple radio-opaque tooth-like structures with a narrow radiolucent rim in the region of 21, 22 and 23 and also above that region, there are 2 radiopaque tooth-like structures suggestive of impacted teeth 21, 22. Hence, the provisional diagnosis was given as compound odontome with impacted teeth 21, 22 (Figure 1(d)).

Under local anesthesia excision of the lesion was done and the specimen was sent for histopathological examination. On gross examination, the specimen showed multiple hard tissues (Tooth 21, 22 and along with several bits of hard tissue mass) and soft tissue (Figure 2(a)). Soft tissue was grayish-white in color, firm in consistency which measures about 1.4 x 1.2 cm in size. Microscopic examination of soft tissue showed discontinuous cystic epithelial lining supported by connective tissue capsule. The lining was stratified squamous non keratinized type of 3-5 layers thickness in some areas made up of flat to cuboidal cells and hyperplastic in other areas. The connective tissue capsule showed parallelly arranged dense collagen fibers, with numerous endothelial lined blood vessels, areas of hemorrhage and mild diffuse chronic inflammatory cell infiltrate. The odontogenic epithelial island was also evident. In few areas, numerous cholesterol clefts associated with giant cells were also noticed suggestive of dentigerous cyst (Figure 2(b)). Microscopic examination of the ground sectioned hard tissue mass showed enamel, dentin and cementum of normal architecture arranged in an orderly pattern suggestive of compound odontome (Figure 2(c)).

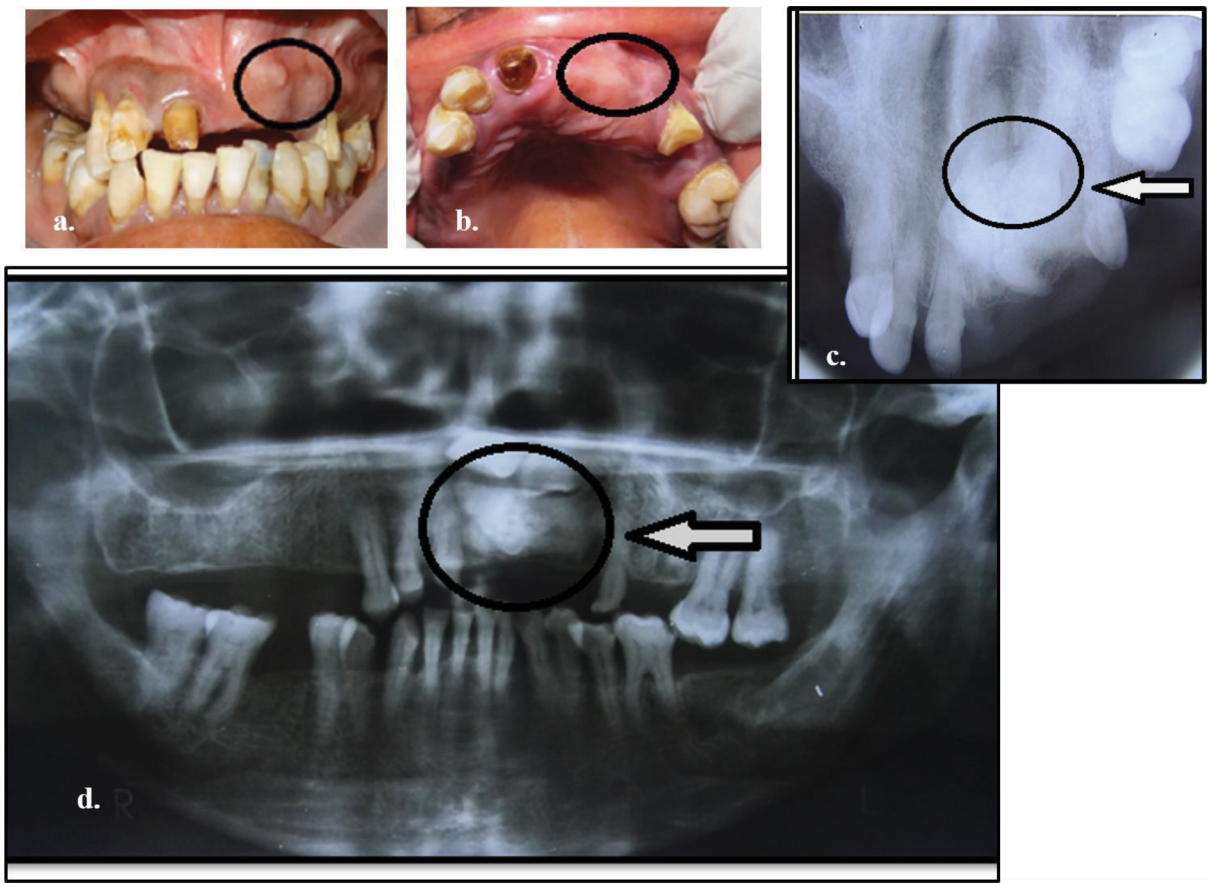


Figure 1. Intra-oral presentation and radiographic picture of Dentigerous cyst in the case. **(a)** Bony spicule like projections seen in 21, 22, 23 edentulous region. **(b)** Cortical expansion was seen in the palatal aspect of 21, 22, 23 region. **(c)** Occlusal radiograph showing radiopacities in 22, 23 region. **(d)** OPG showing radiopacities in 22, 23 regions.

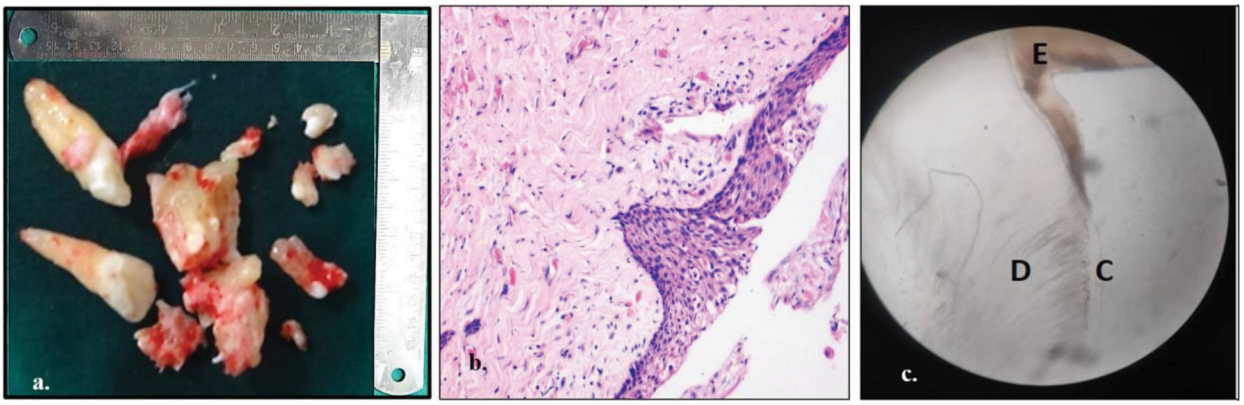


Figure 2. **(a)** Gross specimen showing hard and soft tissue mass. **(b)** H&E Stained section shows thin non-keratinised stratified squamous epithelium with underlying connective tissue capsule. (Arrow mark shows hyperplastic epithelium). **(c)** Ground section showing Enamel (E), Dentin (D) & Cementum (C) of normal architecture arranged in orderly pattern.

Hence on correlating clinical, radiographic, and histopathological findings a final diagnosis of dentigerous cyst associated with compound odontome along with impacted 21, 22 were made.

3. Discussion

Odontomas being most common in the first two decades of life and there is no sex predilection with compound odontoma having increased frequency compared to that ofthe complex[4]. Anterior region of the maxilla being the frequent location for compound and posterior regions of the mandible for complex odontoma. In most of the cases, there are no symptoms, but cortical bone thickening is noticed many number of times[5].

The dentigerous cyst arises from the crown of an impacted tooth having well-defined radioluceny with a sclerotic border and attached to neck of the unerupted tooth in the radiograph[6].

The case presented here shows the synchronous occurrence of dentigerous cyst associated with odontoma in the left anteriorbuccal and palatal region of the maxilla. It was asymptomatic and identified during routine radiographic examination. Simultaneous occurrence of odontomas and dentigerous cysts are rare and scarce. To the best of our knowledge this is the first case reported in the elderly patient in anterior region of maxilla involving both buccal and palatal region (search engine: pubmed). Indecesive appaerence in clinical and radiographic examinations makes the diagnosis difficult, hence histo-pathological examination gives definitive diagnosis (Table 1).

Table 1. Reported cases of compound odontome associated with the dentigerous cyst

Reported By	Gender	Age	Location
Bansal *et al.*, 2014[7]	F	13	Maxilla (right)
Jayam*et al.*, 2014[8]	F	11	Maxilla (anterior)
Morae*set al.*, 2016[9]	M	12	Mandible (right)
Kim *et al.*, 2019[10]	M	56	Mid portion of palate
Pilati*et al.*,2019[11]	M	16	Mandible (right)
Current case	F	55	Maxilla (left anterior buccal – palatal region)

When odontome is associated with dentigerous cyst it may result in malignant transformation includes squamous cell carcinoma, mucoepidermoid carcinoma and ameloblastoma[6]. Hence immediate management is needed to prevent the carcinomatous transformation and to sustain the coherence of the masticatory system. It has a favorable prognosis and a low recurrence rate.

Primary factor in treatment plan is the age of patient and the treatment is different with these cases. Exact treatment plan with regarding to age is not yet reported and it's to be discussed. In young children occurrence of odontomas along with dentigerous cysts are non aggressive than those in adults and treatment plan of marsupialization is preferred, in contrast coming to older age groupits aggressive and there is significant jaw destruction and requires enucleation[10]. In our present case, the patient was treated with complete removal of the compound odontome along with enucleation of the cyst as she was 55 years old.

4. Conclusion

Association of dentigerous cyst along with compound odontome that too in the elderly patient is extremely rare. If odontome is associated with dentigerous cyst it may result in malignant transformation. Clinical and radiographic examination aids in the diagnosis but histopathological examination will lead to a definitive diagnosis. When an elderly patient is considered, enucleation of the lesion along with replacing the defect with the autogenous bone graft helps in rapid healing.

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