

Case Report

Dentigerous cyst accompanied by a calcifying epithelial odontogenic tumor: a case report

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(Received: 21 September 2021, accepted: 17 December 2021)

Keywords:

Calcifying epithelial odontogenic tumor (CEOT) / dentigerous cyst / adenomatoid odontogenic tumor (AOT)

Abstract – Introduction: Calcifying epithelial odontogenic tumor (CEOT) is a rare benign, but locally infiltrating neoplasm which accounts for less than 1% of all other odontogenic tumors. The tumor is often associated with impacted teeth and is twice as prevalent in the mandible. Dentigerous cysts are developmental odontogenic cysts, commonly manifesting in the second and third decades of life. A greater incidence in men than in women with a ratio of 1.6:1 and 61.4% associated with impacted or unerupted teeth. **Observation:** A 17-year-old Indian male reported to the hospital with an asymptomatic lesion associated to an impacted lower first molar with the presence of radiopaque entity inside. The radiological differential diagnosis was calcifying odontogenic tumor, adenomatoid odontogenic tumor and dentigerous cyst. The lesion was completely enucleated under general anesthesia and was sent for histopathologic examination. **Discussion:** Calcifying epithelial odontogenic tumor (CEOT) is a rare benign odontogenic neoplasm derived from epithelial tissue, forming 0.4–3.0% of all intraosseous odontogenic tumors. A dentigerous (follicular) cyst is a developmental odontogenic cyst usually attached to the crown of an unerupted tooth lined by reduced enamel epithelium. **Conclusion:** The patient is still under follow-up and has not shown any signs of recurrence in past 8 months after surgery.

Introduction

Calcifying epithelial odontogenic tumor (CEOT) is a benign locally invasive slow growing neoplasm representing less than 1% of all odontogenic tumors. Pindborg (1958) first described four cases of this unusual lesion; subsequently Shafer *et al.* coined the term Pindborg tumor [1]. This lesion arises from epithelial tissue and is seen commonly in 4th–5th–6th decade of life with no gender predilection. Dentigerous cysts (DC) are the second most common type of odontogenic cyst of developmental origin. DCs form at a frequency of 1.44/100 unerupted teeth, representing ~17.1% of all true jaw cysts [2]. As for CEOT and DC, certain studies have observed recurrent cases with subsequent malignant transformation [3]. The present study describes the case of a 17-year-old boy who exhibited CEOT and DC within the same cavity, an occurrence that, to the best of our knowledge, has been previously identified once in the literature [4,5].

Observation

A 17-year-old Indian male reported to the hospital referring painless swelling in the right side of the mandible. He had no other comorbidities with no history of any trauma. On extraoral examination, a slight swelling on the right side of the mandible was recognized in comparison with the opposite side. On palpation, there was neither local rise in temperature nor was the swelling tender. Intraoral examination revealed painless bony swelling extending bilaterally to the buccal and lingual vestibule from right mandibular premolar to right mandibular second molar (Fig. 1). The tooth associated with the swelling was right second premolar and second molar which gave a positive response to vitality tests. The right first molar was clinically missing. Conventional radiographs showed a well-defined, unilocular mixed radiopaque and radiolucent lesion in the mandible extending from the right canine to the right second molar. The large lesion was associated with horizontally impacted and displaced right first molar with radiopaque flecks within the lesion located in relation to mesial root of second molar (Fig. 2A). Thinning of inferior cortex of mandible was also noticed. The patient was further subjected to a cone beam computed tomography (CBCT) examination (Fig. 2B), which

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Fig. 1. Intra-oral examination demonstrating obliteration of buccal and lingual vestibule from right mandibular premolar to right mandibular second molar.

demonstrated a well-defined mixed radio-opaque and radio-lucent lesion in relation to right posterior mandible. The dimensions of the lesion measured 30.4 mm X 49.9 mm X 27.0 mm (Fig. 2C,D). The lesion was associated with the horizontally impacted and displaced right first molar with presence of granular radiopaque entity within the lesion (Fig. 2D). Biopsy was performed under local anesthesia. Histopathologically, cystic lining epithelium with fibrous connective tissue and mixed inflammatory cell infiltrate was present (Fig. 3). The lining epithelium was non-keratinized stratified squamous epithelium with arcading pattern. Thus, the lesion was suspected to be an inflammatory dentigerous cyst. Under general anesthesia, the lesion was completely enucleated along with extraction of the impacted right first molar and the right second premolar. The mandible on the right side was exposed after a crestal incision on the alveolar ridge along with release incisions medially near the left lateral incisor and distally near the right first molar. The buccal cortex was intact. After extraction of the right second premolar, a reddish, bulky layer of granular-like tissue became evident inside the bony cavity. The cavity was filled with solid tumor including the tooth. The tumor was consequently enucleated from the cavity. The tooth was attached loosely to the tumor and was removed easily and Carnoy's solution was applied along the entire bed of the lesion. Inferior alveolar nerve was preserved by applying vaseline jelly. Macroscopically, the mass was well encapsulated with cystic areas along with an embedded mandibular first premolar in the tumor mass (Fig. 4A). Patient was discharged on the 2nd postoperative day without any IMF (inter-maxillary fixation) and was advised regular follow-up. At the 1st follow up visit, which was at the

1st week interval, the patient's occlusion was dearranged for which orthodontic brackets were placed and the occlusion was guided with interarch elastics (Fig. 5A). Histopathological examination after excisional biopsy revealed epithelium arranged in the form of sheets and connective tissue.

Epithelial cells are polyhedral in shape with centrally placed nucleus. Areas of calcifications seen. The connective tissue is fibrous with few inflammatory cells (Fig. 4B). Based on these findings, the lesion was histopathologically diagnosed as calcifying epithelial odontogenic tumor. Healing of the wound was uneventful postoperatively. The patient is still under follow-up and has not shown any signs of recurrence in the past 8 months after surgery (Fig. 5B).

Discussion

The current case presents two important clinical points, namely that CEOT and DC may occur simultaneously and adjacently in a single cavity of the same jaw. It reports a case of the simultaneous occurrence of CEOT and DC in the mandible of a patient. To the best of our knowledge, the synchronous occurrence of CEOT and DC as distinct lesions has once been previously identified [4,5]. The presence of a single mandibular radiolucent lesion led to the suspected diagnosis of a dentigerous cyst with internal calcifications or odontome. However, the definitive diagnosis of the two pathologically distinct entities of CEOT and DC was made by pathologists based on the excisional biopsies. CEOT may occasionally be an aggressive and recurrent tumor therefore close post-surgical follow-up is preferable [6]. Based only on histopathological data, odontogenic lesions containing calcifications in general are particularly difficult to diagnose.

Simultaneous occurrence of odontogenic lesions as hybrid or distinct lesions has been described in previous literatures [7,8]. Among these, Chindasombatjaroen *et al.* [8] described the case of a patient with CEOT associated with an odontoma, a supernumerary tooth, and DC that simultaneously occurred at varying locations in maxilla. From a radiological viewpoint, the focus was directed towards the internal structure of the lesion which was a mixed radiopaque-radiolucent structure seen lying distal to that of lesion (Fig. 4). The CBCT images demonstrated that the calcification inside the lesion had granular appearance and irregular borders with radiodensity similar to enamel and radiolucency was thought to be similar to pulp making the entity to appear as an odontome. Histopathologically, CEOT is characterized by the presence of homogenous eosinophilic amyloid-like material, and calcifications with epithelial cells arranged in nests and sheets and are polygonal, with clear to eosinophilic cytoplasm and vesicular nuclei having prominent nucleoli [9]. Radiographically it is usually radiolucent with scattered areas of calcification emanating from the central radio dense lesion described as a "driven snow" appearance [10], although this can vary from completely radiolucent (seen early on) to more uniformly dense.

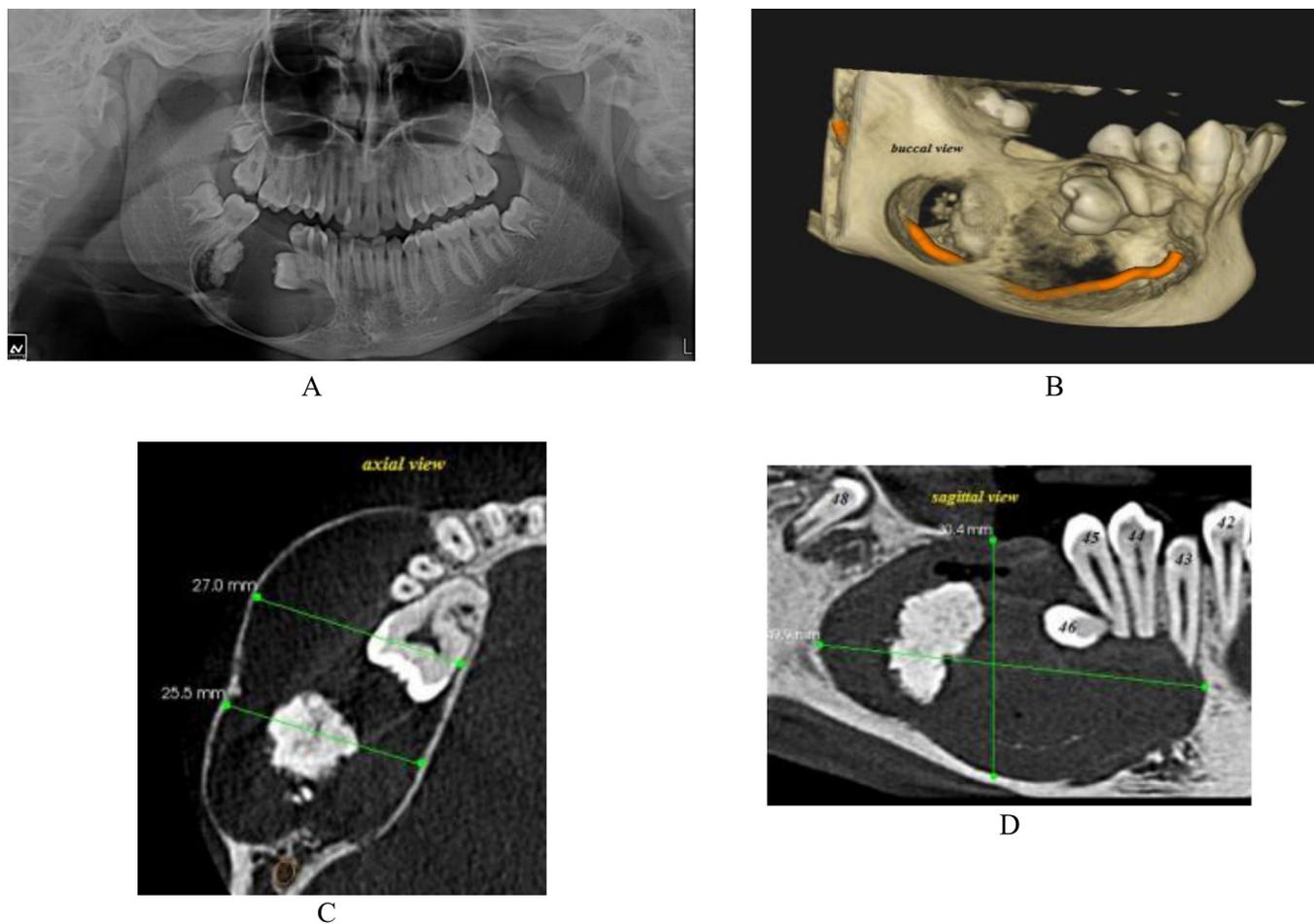


Fig. 2. (A) Initial panoramic radiograph. (B) CBCT images demonstrating the buccal view of the lesion. (C) Axial view and (D) sagittal view demonstrating an impacted lower right first molar with mixed radiopaque –radiolucent structure seen lying at distal aspect of lesion.

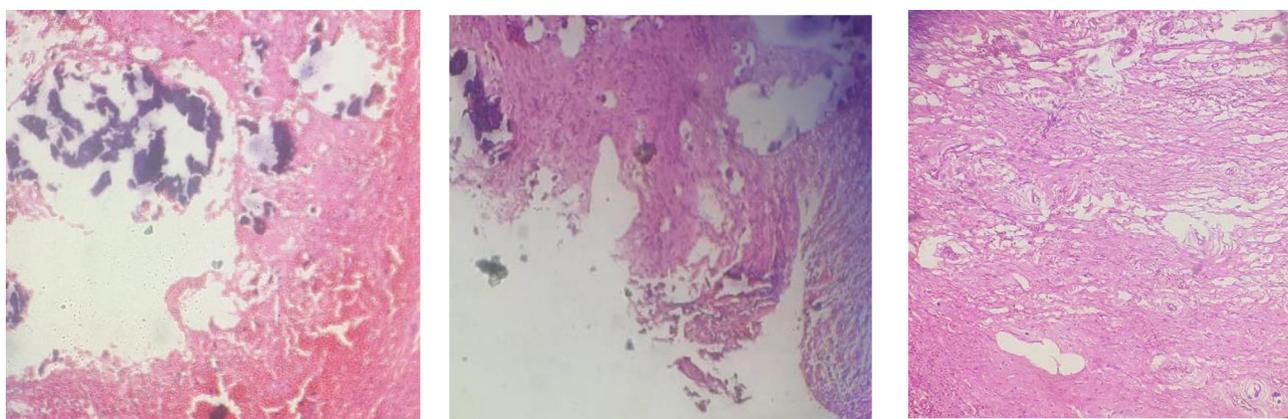
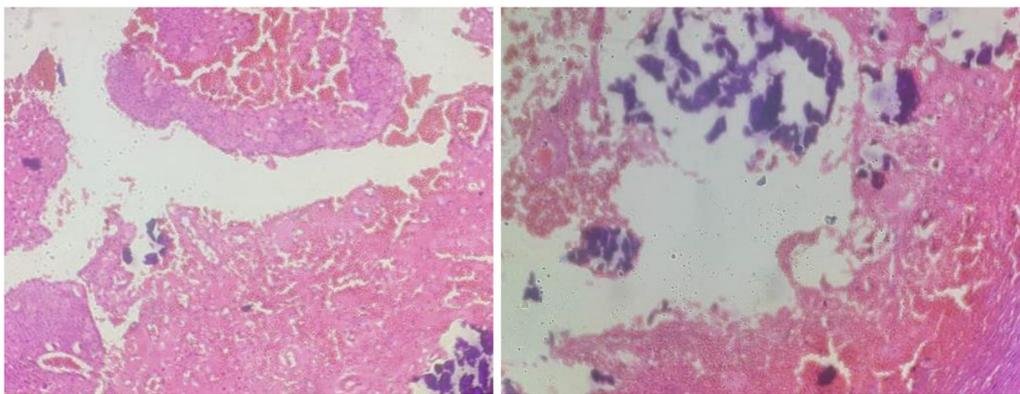


Fig. 3. Histopathological data: The given Hand E section shows cystic lining epithelium with connective tissue. The lining epithelium is nonkeratinized stratified squamous epithelium with arcading pattern. Areas of calcification are also noted. The underlying connective tissue is fibrous with mixed inflammatory cell infiltrate.



A



B

Fig. 4. (A) Macroscopically, the mass was well encapsulated with cystic areas along with an embedded mandibular first premolar in the tumor mass. (B) The given H and E section shows epithelium and connective tissue. Epithelium is arranged in the form of sheets. Epithelial cells are polyhedral in shape with centrally placed nucleus. Areas of calcifications seen. The connective tissue is fibrous with few inflammatory cells.

On conventional radiograph, well-defined unilocular form and regular margins were observed. The CBCT images demonstrate slightly scalloped outline. The septum-like structure present only on the panoramic radiograph was a key result in the interpretation of the case, as it was present in a single lesion. Considering the nature of tumors and cysts, the growth of the CEOT was potentially quicker compared with that of the DC, which could result in pressure from the CEOT on the side of the DC.

DC, ameloblastic fibro-odontoma, adenomatoid odontogenic tumors are all included in the differential diagnosis of a CEOT [11]. By definition, a DC encloses the crown of an unerupted tooth as a result follicular expansion, and it is attached to the cemento-enamel tooth junction. The peak incidence for DCs is within the second and third decades of life, with the mandibular third molars being the most frequently involved teeth [5]. Radiologically, a well-circumscribed osteolytic lesion that contains the crown of the tooth is observed. As the cyst grows, it pulls the unerupted tooth with it.

In the present case, achieving a diagnosis based on radiology was challenging for the following reasons: CEOT and DC are occasionally associated with impacted or unerupted teeth and no

septum-like structure was observed on the CBCT images. It is possible that resorption of the septum occurred due to the skeletal growth of the patient. However, the amount of calcification was markedly different around the second molar.

The present case may serve as a valuable warning that CEOT, which may recur and transform into malignancy if improperly treated, may be present in such lesions.

Conclusion

In conclusion, the present case demonstrated that CEOT and DC may be present simultaneously in a single cavity. The presence of embedded teeth in association with CEOT makes it possible that a dentigerous cyst might develop in the area and vice-versa. Hence, it sounds acceptable to suggest that DC and CEOT may develop in the same site in association with an embedded tooth, since both lesions could arise from reduced enamel epithelium. When the cyst is present in the site of the tumor, it should be considered as one of its components. Careful gross and histological examination of the surgical specimens must be pursued to find out more cases of this

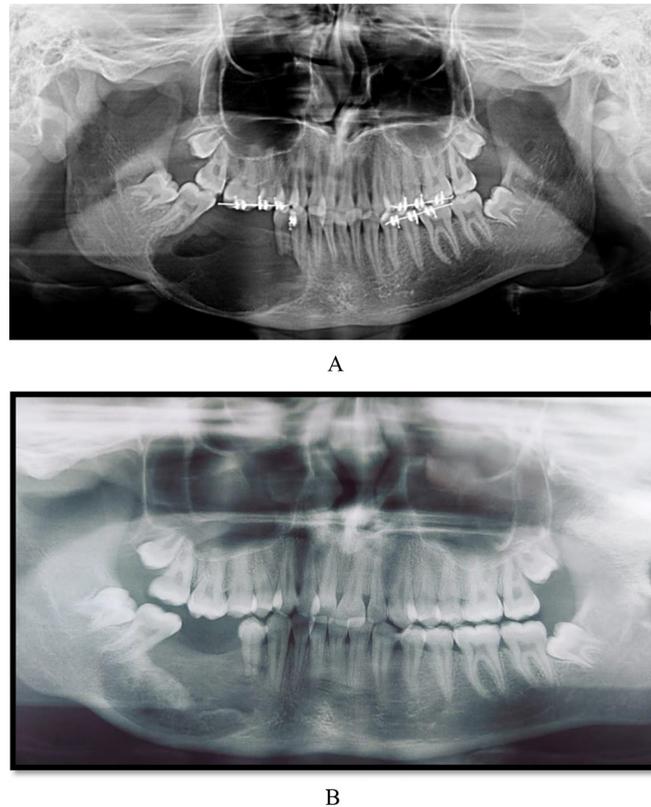


Fig. 5. (A) 1st follow up visit after one week of surgery, the patient's occlusion was dearranged for which orthodontic brackets were placed and the occlusion was guided with interarch elastics. (B) Follow up after 8 months of surgery.

type. Once this entity is recognized, proper follow-up is required to observe their behavior in comparison with other CEOT with – out cyst and/or embedded teeth. Additionally, CBCT also played an important role in careful evaluation of the lesion thereby playing a pivotal role in radiological diagnosis.

Authors contributions:

Rashmi.J.Kurup & Sherman Gomes: Writing original draft.
Amandeep Sodhi: Supervision. Amandeep Sodhi , Swaroop.R. Telkar & Pritham.N.Shetty: Reviewing and Editing.

Informed consent:

Informed consent was obtained while recording case history.

Ethical committee approval:

The authors declares that Ethical approval not required.

Source of funding:

This research did not receive any specific funding.

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