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CALCIFYING ODONTOGENIC CYST WITH AN IMPACTED CANINE MIMICKING AN ODONTOME – A CASE REPORT

[1]*Dr. Sneha R. Sharma, [2]Dr. Freny R. Karjodkar, [3]Dr. Kaustubh P. Sansare, [4]Dr. Ruchika Kapoor, [5]Dr. Isha D. Mishra and [6]Dr. Mohd Saalim

[1]Postgraduate Student, [2]Prof and HOD, [3]Associate Professor, [4]Postgraduate Student, [6]Postgraduate Student, [1,2,3,4,6]Department of Oral Medicine and Radiology, Nair Hospital Dental College, Mumbai Central, Mumbai, Maharashtra, India.
[5]Assistant Professor, Department of Oral Medicine and Radiology, Government Dental College and Hospital, CST, Mumbai, Maharashtra, India.

***Corresponding Author: Dr. Sneha R. Sharma**
Postgraduate Student, Department of Oral Medicine and Radiology, Nair Hospital Dental College, Mumbai Central, Mumbai, Maharashtra, India.

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ABSTRACT

The calcifying odontogenic cyst (COC) is a rare benign odontogenic lesion, first described as a completely different entity in 1962 by Gorlin et al, who named it as the calcifying odontogenic cyst. COCs comprise of approximately 0.3–0.8% of all odontogenic cysts. The COC is categorized as either a cystic variant or a solid (neoplasm) variant. The COC is known to be a type of odontogenic cyst of developmental origin, which can be present in either intraosseous or extraosseous forms, with the intraosseous form being the predominating type. It presents as a benign, slowly growing, asymptomatic swelling, causing an obvious expansion of the jaw. Radiographically, the lesion presents as a well-defined unilocular or multilocular radiolucency with well-defined borders and may also show evidence of irregular calcifications of different sizes, the presence of an odontome or an unerupted tooth. Here we report a case of a 24 year old male patient presenting with a Calcifying Odontogenic Cyst of the mandible in association with an impacted canine, and mimicking the presence of an Odontome.

KEYWORDS: Calcifying Odontogenic Cyst, impacted canine, Odontome.

INTRODUCTION

The calcifying odontogenic cyst is a rare benign odontogenic lesion which was first described in 1932 by Rywkind who regarded it as a different type of cholesteatoma.[1] Later Maitland in 1947 called it as a variant of ameloblastoma.[2] It was Gorlin et al who first described it as a completely different entity in 1962 and named it as the calcifying odontogenic cyst and hence it is also called as the Gorlin Cyst.[3]

It usually presents as a slow growing benign asymptomatic swelling with equal occurrence in the maxilla and the mandible.[4] It has also been reported to occur in association with other odontogenic lesions like the odontomas.[5] Its association with the odontoma is a result of the differentiation and degradation of the cells of the odontogenic epithelium.

Radiographically, the lesion presents as a well-defined unilocular or multilocular radiolucency with well-defined borders and may also show evidence of irregular calcifications of different sizes, the presence of an odontome or an unerupted tooth.[6]

Here we present a case of a 24 year old male patient presenting with a Calcifying Odontogenic Cyst of the mandible in association with an impacted canine.

CASE REPORT

A 24 year old male patient reported to the OPD with a chief complaint of a swelling on the right side of the jaw since 4-5 months. The patient was apparently alright when he noticed a small swelling on the right side of the jaw, which had gradually increased in size for 5 months.

The patient was asymptomatic and there was no pain or paresthesia associated with the swelling. There was no significant past medical or dental history.

On extra-oral examination, the face appeared asymmetrical with an extra-oral swelling in the right parasymphyseal region of the mandible approximately 3x3 cm in size (Figure 1). On palpation, the swelling was well localized, hard and non-tender, along with Egg shell crackling on the superior aspect of the swelling, just lateral to the right side commissure of the mouth. The temperature and texture of the skin overlying the swelling was similar to the adjacent normal skin.

On intra oral examination, a well-defined intra oral swelling along with buccal expansion was noted in the right side of the mandible extending from mandibular right first premolar up to mandibular right second molar with obliteration of the buccal vestibule (figure 2). The mucosa appeared stretched and vascular on the right buccal vestibule. The mandibular right canine was found to be missing and the patient gave no history of extraction with the same. On further examination it was found that there was no mobility or tenderness to palpation, no sign of caries, pulp pathosis or periodontitis with the involved teeth. Bases on the clinical features a provisional diagnosis of a cystic lesion along with a missing tooth was reached.

A Digital OPG was done as a screening radiographic investigation, followed by a small FOV CBCT for the right side body of the mandible.

The Digital OPG (Figure 3) showed a well-defined radiolucent lesion extending antero-posteriorly from the periapical region of mandibular right lateral incisor to the periapical region of mandibular right first molar. Supero-inferiorly the lesion extended from the apical third of roots of mandibular right first premolar, second premolar and first molar till the inferior border of the mandible. The periphery of the lesion was well defined and irregular in shape with partially sclerotic borders. The internal structure was radiolucent except at the anterior aspect where multiple well defined radio opaque structures were present in a clustered form. The mandibular right canine was seen horizontally impacted, displaced inferiorly just above the inferior border of the mandible.

The findings of the CBCT (Figure 4) showed that the overall dimensions of the lesion were 4.5 cm x 2.5 cm x 2.3cm (anteroposterior, buccopalatal and superoinferior respectively).

At 2.5 mm and 13.7 mm axial oblique section a radio opaque tooth like structure with a radiolucent opening in its centre was noted which suggested the presence of an odontome.

At its inferior aspect the lesion showed a presence of a horizontally impacted canine. The tooth bud follicle of the impacted canine was seen to be contiguous with the lesion at the CEJ at 7.4mm in the sagittal oblique section.

Blunting of the root apices was noted with mandibular right first and second premolars and mandibular right first molar suggestive of external root resorption. There was inferior displacement of the inferior alveolar nerve canal at mandibular right first molar region. There was marked expansion and thinning of the buccal cortical plate.

Based on the radiographic features, a provisional diagnosis of a Dentigerous cyst with an impacted canine and an Odontome was reached.

The differential diagnosis consisted of Benign Odontogenic Tumor, most likely Calcifying Epithelial Odontogenic Tumor or Unicystic Ameloblastoma.

Treatment comprised of root canal treatment for the involved teeth (mandibular right first premolar, second premolar, first molar and second molar), followed by enucleation and curettage and extraction of the impacted right mandibular canine under general anaesthesia. The excised tissue (Figure 5) along with the impacted tooth was sent for histopathological examination.

On discussion with the oral pathologist it was found that there was presence of ghost cells in the stroma and a calcification within the epithelial lining. The overall histopathological features were suggestive of a Calcifying Odontogenic cyst associated with an impacted mandibular right canine.(Figure 6) The patient has been kept under regular follow up and a digital OPG (4 months post-operative) showed no signs of recurrence. (Figure 7).

FIGURES

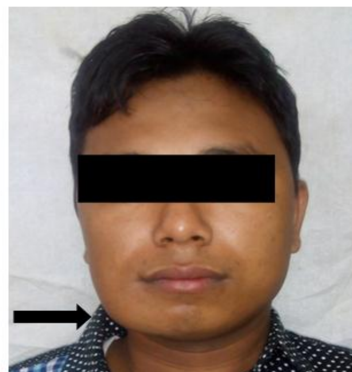


Figure 1: Extra oral Clinical Photograph showing a well-defined swelling on the right parasymphyseal region of the mandible (black arrow).

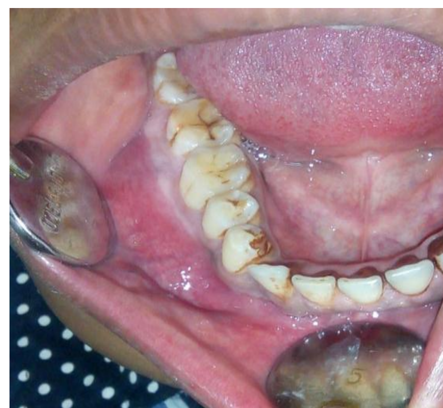


Figure 2: Intraoral Examination showing an intraoral swelling obliterating the lower right buccal vestibule.

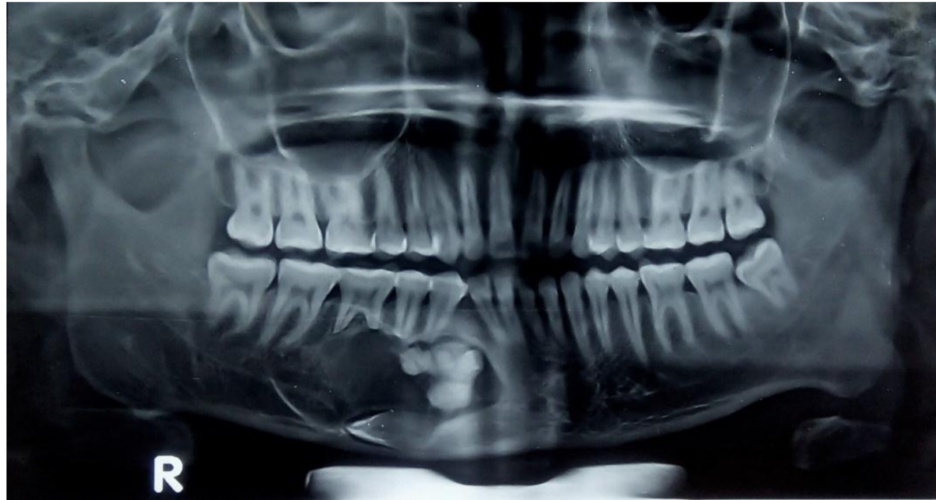


Figure 3: Digital OPG – showing a well defned Radiolucency along with a horizontally impacted mandibular right canine.

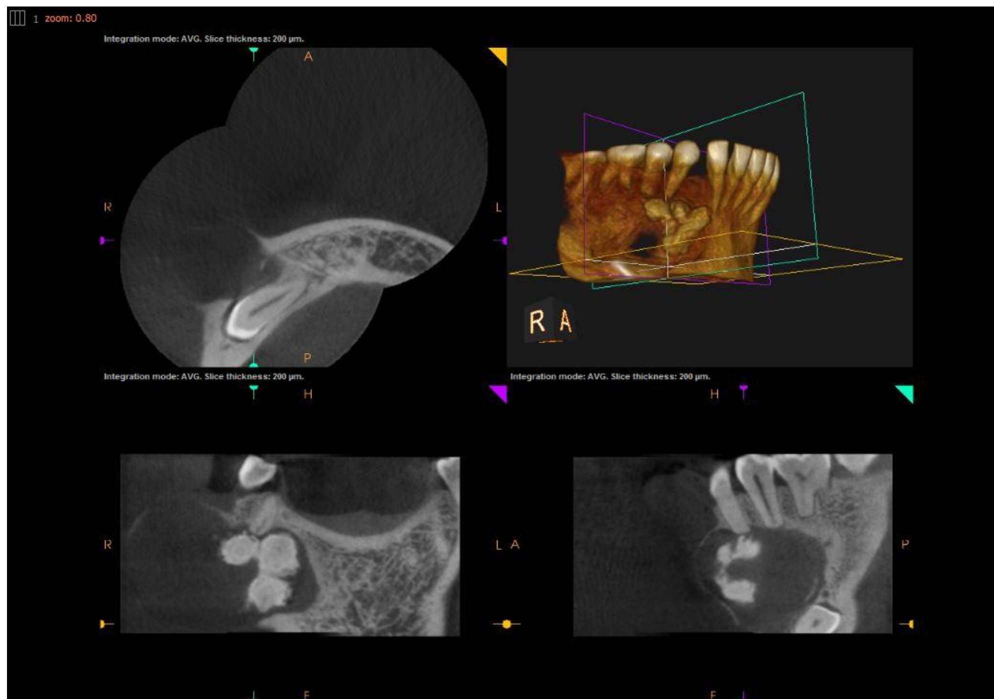


Figure 4: CBCT Images.

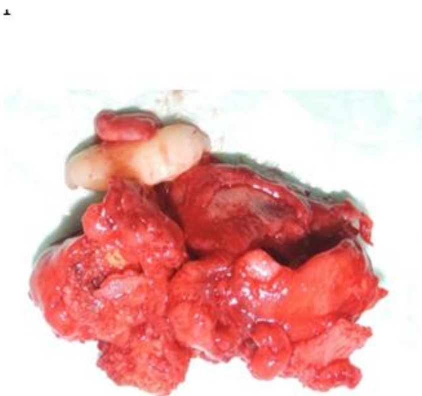
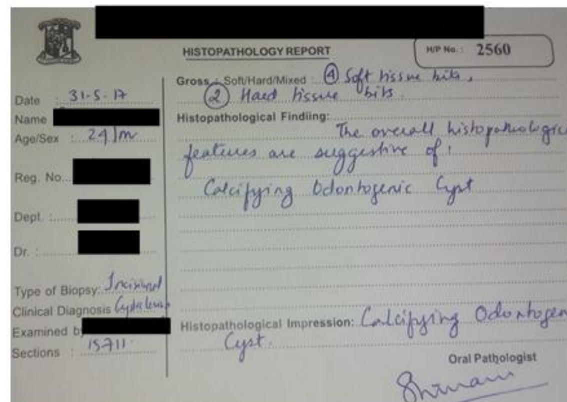


Figure 5 – Excised lesion along with the Impacted tooth and calcified mass



HISTOPATHOLOGY REPORT

H/P No.: 2560

Date: 31-5-17
Name: [redacted]
Age/Sex: 24/m
Reg. No.: [redacted]
Dept.: [redacted]
Dr.: [redacted]
Type of Biopsy: [illegible]
Clinical Diagnosis: [illegible]
Examined by: [redacted]
Sections: 15711

Gross: Soft/Hard/Mixed: (4) Soft tissue bits, (2) Hard tissue bits.

Histopathological Finding: The overall histopathological features are suggestive of: Calcifying Odontogenic Cyst

Histopathological Impression: Calcifying Odontogenic Cyst.

Oral Pathologist

Figure 6 – Histopathology report

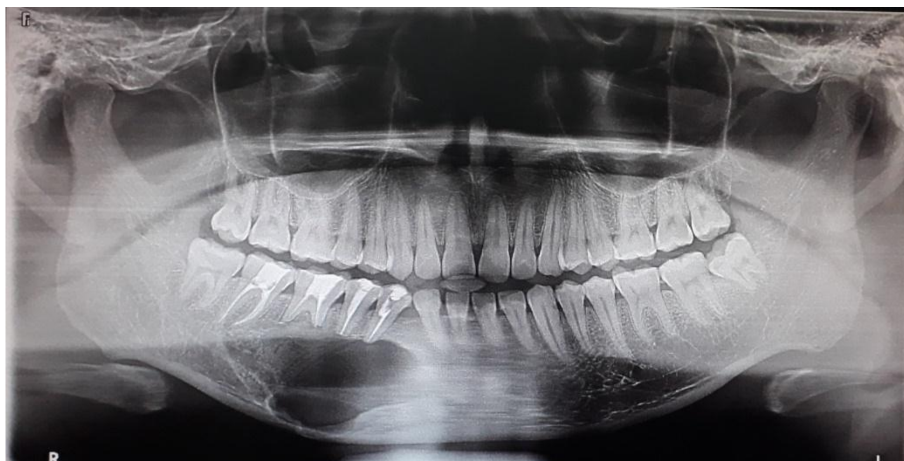


Figure 7: Post-operative Digital OPG after 4 months.

DISCUSSION

COCs comprise of approximately 0.3–0.8% of all odontogenic cysts.[7, 8] Praetorius et al. suggested that the origin of the COC was from the dental follicle, gingival tissue or bone, or from remnants of odontogenic epithelium or reduced enamel epithelium.[8] There has been considerable debate regarding the classification and nomenclature of this entity, but COC is the preferred term.[8] The COC is categorized as either a cystic variant or a solid (neoplasm) variant. The cystic variant has three forms - simple unicystic type, unicystic odontoma-associated type, and unicystic ameloblastomatous proliferating type. The solid variant is named as the dentinogenic ghost cell tumor, which may present with recurrence and aggressive behaviour.[6]

The COC is known to be a type of odontogenic cyst of developmental origin, which can be present in either intraosseous or extraosseous forms,[8] with the intraosseous form being the predominating type.

It presents as a benign, slowly growing, asymptomatic swelling, causing an obvious expansion of the jaw. They develop in equal frequency in males and females, at any age from the second to the eighth decade of life either.[8] The maxilla and the mandible are equally affected, though some authors report a mandibular predominance with a ratio of 2: 1 or 3:1.[10,11,12] The lesion is usually located anterior to the first molar region.

Only 2% of the cases have been found to be in the posterior molar region, similar to the present case, involving the anterior as well as the posterior portion of the right side of the mandible, where it was justified to have a CBCT as the radiographic examination to avoid the overlapping of other structures and view the complete extent of the lesion.

These cysts have been frequently associated with odontomas in 20-24% of the cases.[13,14,15] Radiographically, they generally appear as a solitary unilocular radiolucency with a well-delineated margin[13,15] and containing specks of calcifications. 5% of the cases may present with a multilocular radiolucency.[13] The presence of calcifications is found in almost 50% of the cases, which is an important diagnostic feature in radiographic interpretation. The calcifications can range from small flecks which can go unnoticed radiographically to large masses of calcifications, as seen in the present case.

The differential diagnosis based on the presence of calcified material includes adenomatoid odontogenic tumor, calcifying epithelial odontogenic tumor, ossifying fibroma, and odontoma.

Approximately half of cases are found to be in association with an unerupted tooth.[8] Some other features which are found are root resorption, root divergence, expansion, thinning and perforation of the cortical plates.

The chances of root resorption are higher when the lesion is associated in close proximity to the root apices of the teeth.

Histopathological features of this cystic lesion show a well-defined basal layer in the epithelial lining, along with a layer of stellate reticulum- like cells above it. Masses of ghost epithelial cells are dispersed in the epithelial cyst lining or in the fibrous stroma. The ghost cells are groups of enucleated broad eosinophilic cells, containing an amorphous material with with a propensity to calcify. The presence of ghost cells in COC is not pathognomonic for COC and it is also found in ameloblastoma, ameloblastic fibroma, ameloblastic fibroodontoma and odontoma.[16] The recommended line of treatment for the calcifying odontogenic cysts of the cystic type is enucleation with curettage, along with removal of the surrounding bone of about 1-2 mm to prevent any chances of recurrence.[6]

Generally the cyst presents with good prognosis and low rates of recurrence, but recurrence is more prone to occur in cases of the solid neoplastic variant (dentinogenic ghost cell tumor). Even though the COCs present with

good prognosis, a rigorous follow up is recommended to monitor any signs of recurrence.

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