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Cover Page Footnote

Dr.P.T.Ravikumar for his reference work of CBCT images.

CALCIFYING ODONTOGENIC CYST– A THREE-DIMENSIONAL RADIOLOGICAL EVALUATION BY CONE BEAM COMPUTED TOMOGRAPHY.

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Abstract : Calcifying odontogenic cysts is an odontogenic cyst that gives rise to painless swelling which expand and occasionally perforate bone. Most of them are between 2 to 4 cm in diameter. Approximately 75% occur in anterior canine-incisor region. The highest incidence of occurrence is between 10 and 29 years. Radiographically they appear as well-circumscribed, unilocular cyst-like areas with scattered radiopacities which range from mere flecks to large masses. The related tooth usually gets displaced. Radiopaque flecks occur in radiographic features of some lesions which undergo osteolysis during their development. They occur as a result of dystrophic calcifications in long standing chronic mature lesions. The location of such entities whether pericoronal or periapical is also crucial in narrowing the diagnosis. Such flecks in radiographs occur near the pericoronal region in calcifying epithelial odontogenic tumour, ameloblastic fibro-odontoma, whereas in calcifying odontogenic cyst, they appears small, discrete, pebble-like with a smooth outline in periapical region. Such pebbles of radiopaque specks were incidentally seen in cone beam computed tomography in a 20-year-old male evaluated radiographically in orthogonal planes in coronal, axial, sagittal slices and three-dimensional reconstructed view and treated by surgical enucleation was described here.

Keywords: Calcifying odontogenic cyst, cone beam computed tomography, surgical enucleation.

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Conflicts of interest:

The authors declare no conflicts of interest.

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KYSTE ODONTOGÈNE CALCIFIANT - UNE ÉVALUATION RADIOLOGIQUE TRIDIMENSIONNELLE PAR TOMODENSITOMÉTRIE.

Résumé : Le kyste odontogène calcifiant est un kyste odontogène qui donne lieu à un gonflement indolore qui se dilate et parfois perce l'os. La plupart d'entre eux mesurent entre 2 et 4 cm de diamètre. Environ 75 % surviennent dans la région canine-incisive antérieure. L'incidence la plus élevée se situe entre 10 et 29 ans. Radiographiquement, ils apparaissent comme des zones bien circonscrites, uniloculaires, semblables à des kystes, avec des radio-opacités dispersées allant de simples taches à de grandes masses. La dent associée est généralement déplacée. Des taches radio-opaques apparaissent dans les caractéristiques radiographiques de certaines lésions qui subissent une ostéolyse au cours de leur développement. Ils surviennent à la suite de calcifications dystrophiques dans des lésions matures chroniques de longue date. La localisation de ces entités, qu'elles soient péri-coronaires ou péri-apicales, est également cruciale pour affiner le diagnostic. De telles taches sur les radiographies se produisent près de la région péri-coronaire dans la tumeur épithéliale odontogène calcifiante, le fibro-odontome améloblastique, alors que dans le kyste odontogène calcifiant, elles apparaissent petites, discrètes, ressemblant à des cailloux avec un contour lisse dans la région péri-apicale. De tels cailloux de taches radio-opaques, sont décrits dans cet article, et ont été observés par hasard, chez un homme de 20 ans, en tomodensitométrie à faisceau conique dans des plans orthogonaux en coupes coronales, axiales, sagittales et une vue reconstruite en trois dimensions et traités par énucléation chirurgicale.

Mots clés: kyste odontogène calcifiant, cone beam computed tomography, énucléation chirurgicale.

Introduction

Calcifying odontogenic cyst or Gorlin's cyst is a rare, uncommon, slow-growing and benign lesion varying from a cyst to an odontogenic tumor [1]. The cyst can sometimes reach a solid neoplasm due to epithelial proliferation [1]. The World Health Organization (WHO) now classifies it as a tumor [1]. The lesion also has a taller columnar epithelial cell, that gives an appearance resembling to an ameloblastoma, although it does not have its clinical behavior [1]. Occasionally, this lesion produces calcified tissue associated with dysplastic dentin and an odontoma [1]. The calcifying odontogenic cyst demonstrates variable clinical behavior and considerable histopathological diversity [1]. This lesion may be associated with other odontogenic tumors such as ameloblastomas, odontomas and adenomatoid odontogenic tumors since ghost cell occurs in all of these lesions [1]. Most calcifying odontogenic cysts are between 2 to 4 cm in greatest diameter but can grow as large as 12cm [1]. Divergence of the root usually occurs [1]. Extraosseous calcifying odontogenic cysts occur as sessile, gingival masses resembling gingival fibromas and gingival cysts [1]. The most characteristic histopathological feature is the presence of "Ghost cells", which are altered epithelial cells characterized by presence of only cell outline with loss of nuclei [1]. Some consider it an aberrant form of keratinization of the odontogenic epithelium; others believe it is the accumulation of enamel protein by coagulative necrosis [1]. Ghost cells fuse to form large sheets of acellular, amorphous material. Calcifications occur within ghost cells [1]. The rare intraosseous variant consists of islands of odontogenic epithelium and ameloblastic strands embedded in a stroma of fibrous connective tissue [1].

Case Presentation

A 20-year-old male reported a chief complaint of facial asymmetry due to swelling involving the an-

terior maxilla in the cuspid region. History reveals it started as small swelling and has progressed to the present size in one year. History revealed no other similar swellings on his body. The patient had no harmful oral habits like smoking, chewing tobacco, or alcohol. Intraoral examination revealed a swelling obliteration of the maxillary labial vestibule with the 22,23 region. On Palpation, the swelling was non-tender, fluctuant, and firm in consistency. The radiological evaluation was done by Cone Beam Computed Tomography, which revealed multiple discrete pebble-like radiopaque flecks with smooth outlines. CBCT Panoramic

image showed pathological migration of 22 and 23 with discrete radiopaque dots between them. CBCT Axial section revealed an expansile lesion measuring approximately 17.2 mm x 13.2 mm with pathological migration of 22 and 23 with an expansion of the lingual cortical plate and destruction of the labial cortical plate. It also revealed mucosal thickening within the left maxillary sinus and destruction of the anterior portion of the hard palate CBCT -Sagittal section revealed destruction of the floor of the nasal cavity in the 21 regions and posteriorly extending to the nasopalatine nerve canal. [Figure 1 A, B, C, D, E, F].

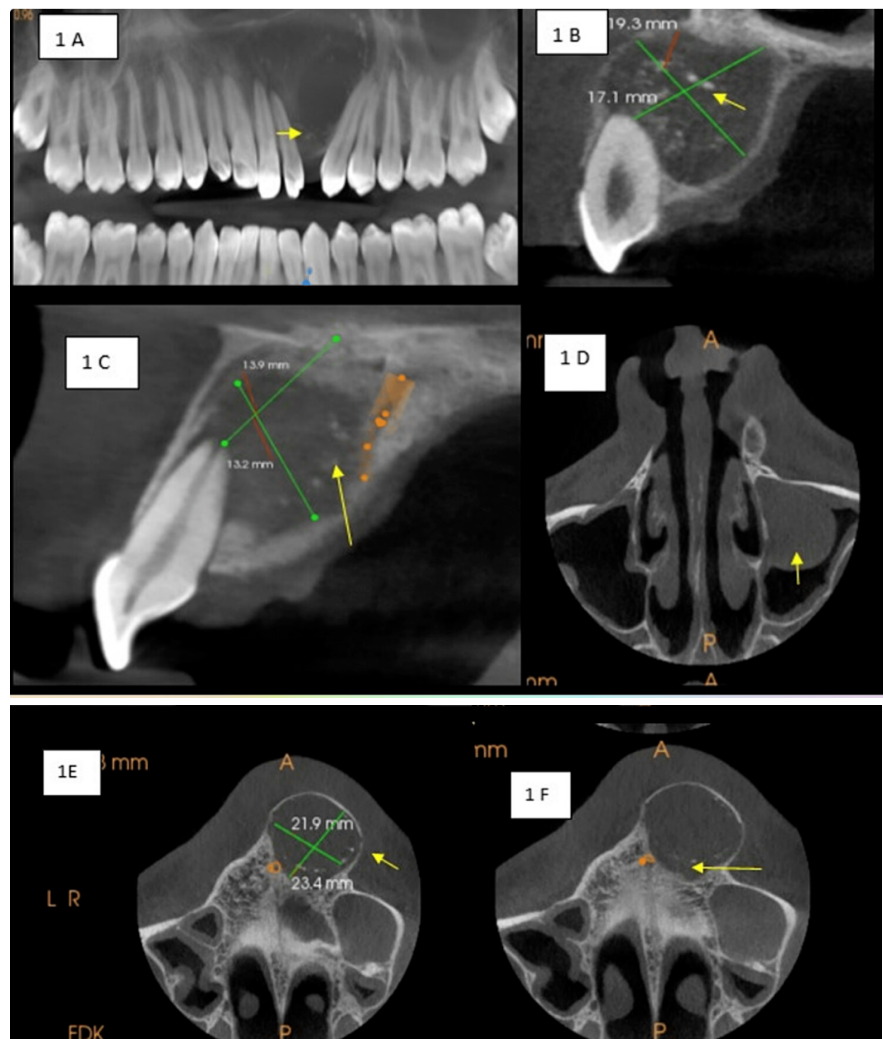


Figure 1. A. CBCT Panoramic image shows radiopaque flecks and pathological migration between 22 and 23. B. CBCT sagittal section shows expansion of lingual cortical plate and destruction of labial cortical plate. C. CBCT Sagittal section shows destruction of floor of nasal cavity and nasopalatine canal. D. CBCT Axial section shows mucosal thickening within left maxillary sinus E. F. shows destruction of anterior region of hard palate

The three-dimensional reconstructed CBCT images [Figure 2 A, B].

Postoperative analgesics Tablet.Ketorolac tromethamine 10 mg twice daily and antibiotic clindamycin 300 mg BD given for seven days. The patient was followed up after three months and the swelling did not recur.

Discussion

Definition

The calcifying odontogenic cysts represent a heterogeneous group of lesions that exhibit various clinicopathologic and behavioral features [1]. Calcifying odontogenic cyst is a benign cystic entity lined by odontogenic epithelium and simulates the clinical behavior of ameloblastoma, such as locally aggressive, anatomically benign and clinically persistent [1]. In 1962 Gorlin and his colleagues first described the "calcifying odontogenic cyst" as a distinct clinicopathological entity [2].

Terminologies

In 1963, Gold M chose the term "Keratinizing and calcifying odontogenic cyst (KCOC)" [2]. In 1965, Bhaskar M suggested the term "keratinizing Ameloblastoma (KA)" [2]. In 1972, the term "calcifying Ghost cell odontogenic tumor" was suggested by Fejerskov and Krogh. Freedman and his associates suggested a "cystic calcifying odontogenic tumor" [2]. In 1981 Praetorius et al. suggested the term "dentino-genic ghost cell tumour". In 1986, Ellis and Smoother termed it an "Epithelial odontogenic ghost cell tumour". In 1990, Colmenero et al. suggested the term "odontogenic ghost cell tumor". In 1994, Shear M stated it as "odontogenic ghost cell ameloblastoma". Hirschberg et al. in 1994 stated it as an "odontocalcifying odontogenic tumor". In 2005, the World Health Organization "calcifying cystic odontogenic tumor" [2].

Theories

Monistic theory: The term "Monistic" refers to the "concept of oneness" or the beginning or "source".

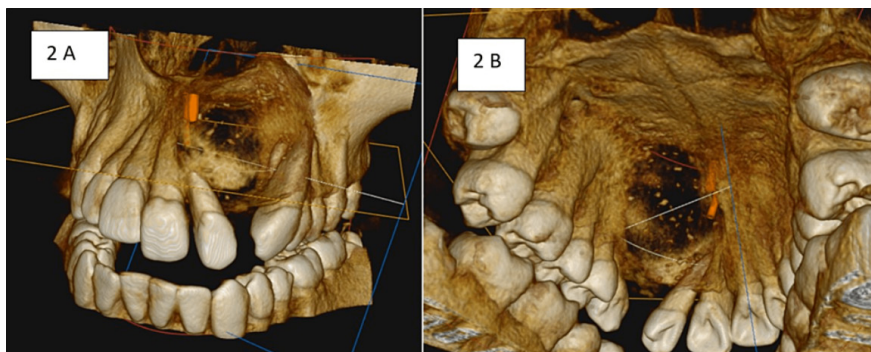


Figure 2. A. Three-dimensional reconstructed CBCT image revealed pathological migration of 22 and 23 B. Destruction of Hard palate



Figure 3. Excised specimen.

The monistic theory considers calcifying odontogenic cysts as neoplastic even though they are cystic and benign [2]. Dualistic theory: calcifying odontogenic cyst appears in two different clinical behaviors such as neoplastic or cystic forms [2].

Etiopathogenesis

Gorlin and his associates suggested that the cyst might be an oral analog of the calcifying epithelioma of Malherbe (Pilomatricoma) of the skin [3]. In addition, the Gorlin cyst shares histological features of a rare intracranial neoplasm, "craniopharyngioma," due to immunologic similarities [4]. The increased expression of claudin occurs in calcifying odontogenic cysts [5]. Calci-

ifying odontogenic cysts result from a mutation in the CTNNB1 gene that causes abnormal ghost cell proliferation [6]. The ghost cell keratinization of odontogenic epithelium is a striking histological feature of the entity [6].

Types

Calcifying odontogenic cyst is divided into the following types a) cystic-turning into unicystic ameloblastoma, b) cystic associated with other hamartomas such as odontomas, c) solid-turning into peripheral ameloblastoma, d) malignant counterpart-odontogenic ghost cell carcinoma [6].

Epidemiology

The prevalence of Gorlin cysts is about 1.5% among the south Indian population [4]. 13 to 30 % occur in extraosseous locations such as on gingiva. 38% occur in maxillary incisor and canine region, 27% in mandibular incisor and cuspid regions, 12 % in maxillary premolar regions, 11 % in mandibular molar regions, 10 % in mandibular premolar regions, 2 % in maxillary molar regions. They are also called "Hybrid lesions" since they occur along with other ghost cell-containing lesions such as ameloblastic fibro-odontoma [7].

Clinical features

Gorlin's cysts usually occur in age groups from 10 to 19 years. The mean age of occurrence of calcifying odontogenic cysts is 39 years. Clinically, the lesion usually appears asymptomatic and painless, with jaw swelling resulting in facial asymmetry with a slow-growing nature. Occasionally when the lesion expands and destroys the cortical plate and extend into the soft tissue, the cystic mass become palpable. 75 % of calcifying cystic odontogenic tumors occur anterior to the maxillary first molar, especially in the region of incisors and canine. The periphery can vary from well-defined corticated border with a curved, cyst-like shape to ill-defined and irregular outline. The internal aspect of the lesion varies from discrete foci of calcified material, which are small that appear as white spots or pebbles with a smooth outline. Displacement of teeth usually occurs, but root resorption does not occur. Cortical expansion and destruction occur with enlarging lesions [7]. The lesions can be confused with desmoplastic ameloblastoma, but they occur more in the anterior mandible around multiple impacted teeth [8,9].

Table 1. Differential diagnosis for Radiopaque flecks on radiograph

Lesion	Mean Age/ Gender	Radiographic description
Adenomatoid odontogenic tumour [10]	16 / Female	Usually associated with an impacted tooth
Calcifying epithelial odontogenic tumour [11]	22/ Male	snow storm radiopaque flecks in a pericoronal location. The radiopacities appear close to the crown of embedded tooth
Compound odontome [12]	18/ No gender predilection	Numerous small fused tooth-like radiopaque structures above impacted teeth
Ameloblastic fibro-odontoma [13]	19 / Male	Small, discrete, round shaped calcifications around impacted tooth
Chronic osteomyelitis [14]	50 / Male	Early phases are characterised by osteolysis. chronic stages are characterised by sequestrum or involucrum (new bone) formation.
Desmoplastic ameloblastoma [15]	42/ Male	Mixed radiolucent-radiopaque flecks resembling honey-comb in anterior mandible

Differential diagnosis

The various differential diagnosis for radiopaque flecks in radiograph described [Table 1]

Adenomatoid odontogenic tumour most commonly seen among females in anterior maxilla. Radiographically they contain faint radiopaque foci associated with an unerupted tooth. Calcifying epithelial odontogenic tumour radiographically appear as diffuse radiopacities of variable size, within the lesion. These may be so extensive as to make the lesion relatively radiopaque and occasionally so small as cannot be seen in conventional radiographs. Radiopacities are uniformly distributed to give the lesion a driven-snow appearance. compound odontomas radiographically shows multiple small tooth-like radiopaque denticles above impacted teeth. Ameloblastic fibro-odontomas radiographically appear as opaque odontomatous tissues in a circumscribed but otherwise radiolucent area. Chronic osteomyelitis occurs more commonly in mandible

than maxilla. Bony changes do not appear until after at least 10 days. The sharp trabecular pattern of the bone is lost where it has been resorbed and areas of radiolucency indicate bone destruction. These areas have ill-defined margins and have a fluffy or moth-eaten appearance. Areas of dead bone appear as relatively dense areas which become more sharply defined as they are progressively separated as sequestra. Later subperiosteal bone formation occurs as a thin strip of new bone below the lower border of the bone resembling onion-peel appearance. Desmoplastic ameloblastoma frequently involve anterior mandible and radiographically appear as irregular lucent areas containing fine calcifications and with indistinct borders or as mixed radiolucent and radiopaque areas.

Histopathological features

Calcifying odontogenic cyst consists of a fibrous cyst wall that encloses an epithelial lining, which is irregular in thickness and structure.

In some areas they contain well-defined basal cell layer resembling ameloblast-like cells with reversed nuclear polarity. Superficial to this layer, there is frequently a zone of loose edematous cells bearing some resemblance to stellate reticulum. The most characteristic histological finding is the presence of ghost cells which are swollen, keratinized, anucleate cells. Such ghost cells form small foci within the epithelial lining or large masses extending into or even filling the cystic lumen. They may also extend into the underlying connective tissue and provoke a foreign body reaction. The ghost cells may become mineralized to form calcified masses of dentin-like material of variable size.

Treatment

The treatment of calcifying odontogenic cysts greater than 1.5 cm in diameter includes surgical marsupialization and curettage followed by enucleation. Further Large lesions above 4 cm in diameter can be treated by dredging, which includes decompression, followed by scraping

of the lesion repeatedly and providing a patent tube made of acrylic to allow drainage of the cystic fluid and minimizing damage to vital adjacent structures [16,17].

Prognosis

The Prognosis for calcifying odontogenic cysts is good or bad, according to the occurrence of calcifying odontogenic cysts in conjunction with other hamartomas like odontomas or ameloblastic carcinoma. Those calcifying odontogenic cysts in the circumstances with odontomas have a good Prognosis, whereas those associated with ameloblastic carcinomas have a poor Prognosis. The recurrence is minimal after simple surgical enucleation. Recurrences are common only in malignant odontogenic ghost cell carcinomas, as they have unpredictable behavior.

Conclusions

Cone beam computed tomography has revolutionised the field of imaging and is a boon for dentists

in today's digital era. The exact location and pattern of distribution of radiopaque flecks plays an important role in the diagnosis of maxillofacial pathological lesions such as calcifying odontogenic cyst (Gorlin cyst), calcifying odontogenic tumour, adenomatoid odontogenic tumour, odontoma, desmoplastic fibroma. A careful history taking is also essential in identifying such odontogenic lesions which show radiopaque flecks on radiographs, which may not be much discernible on conventional radiographs. There is no doubt that CBCT helps in identifying such oral lesions with varied clinical behavior and heterogeneity. Furthermore, CBCT helps to identify and study the internal architecture of such heterogenous lesions involving the maxilla or mandible, which causes diagnostic dilemmas to the dentist, thereby aiding dentists in narrowing their diagnosis, not only depending on the histopathology.

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