ODONTOGENIC KERATOCYST: AN UNPREDICTED HAVOC IN MANDIBULAR BODY- A CASE REPORT WITH REVIEW OF LITERATURE

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ABSTRACT

The Odontogenic keratocyst (OKC) is a developmental, non -inflammatory chronic cystic lesion that occurs most commonly in the third and fifth decade. There is a slight male predilection. Mandibular occurrences are more as compared to the maxilla, with a marked tendency to involve the posterior body and ascending mandibular ramus. OKC tends to grow in an anteroposterior direction within the medullary cavity without causing much buccolingual bone expansion. It is a cyst of odontogenic origin, usually asymptomatic with an aggressive clinical behavior including a high recurrence rate and tendency to invade bone and adjacent soft tissues. Radiographically OKC may be unilocular or multilocular. Diagnosis is based on the clinical history, clinical appearance radiographs and histology. Here, we present a case of a 25-year-old male patient who reported with mobile deciduous teeth and an incidental diagnosis of massive OKC along with an impacted tooth which was made on routine radiographic investigations and CBCT findings were emphasized.

Keywords:WHO Classification, Odontogenic Keratocyst, Orthokeratinized Variant, CBCT, Recurrence.

INTRODUCTION

Odontogenic keratocysts (OKCs) are developmental cysts that arise from remnants of the dental lamina.[1] They may appear either as a single entity or as multiple cysts associated with syndromes like Gorlin–Goltz syndrome.[2,3] Odontogenic keratocyst is a distinctive form of developmental odontogenic cyst that deserves special consideration because of its specific clinical behavior and histopathologic features.[4]

The term odontogenic keratocyst was first given by Philipsen in 1956.[5] OKC's most commonly occur in the second and third decades of life and show a slight predilection for males (males to female ratio 1.3:1).[5] The recent WHO classification categorizes OKC as a developmental non-inflammatory odontogenic cyst.[6,7]

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The majority of the OKC's occur within the mandible, most commonly it occurs in the angle-ascending ramus region. Clinically, OKC presents itself asymptomatically, usually with a medullary growth pattern, which causes minimal expansion of the cortical bone, but sometimes it may be related to pain and aggressive growth.[8]

Radiographically, OKC appears as a well-defined radiolucent area with marginal delimitation. OKC can be unilocular or multilocular. The multilocular form resembles ameloblastoma, especially when it affects the mandible. Also, it may or may not be associated with an included tooth, which can sometimes be confused with the dentigerous cyst.[8,9] This article presents an incidentally diagnosed OKC in the mandibular body with an impacted tooth making it a diagnostic diploma.

CASE REPORT

A 25 years old male patient reported to the Out Patient Department of Oral Medicine and Radiology of Rajas Dental College and Hospital with the chief complaint of mobile milk teeth in the upper front tooth jaw region for the past 1 month. On eliciting the history, the patient was apparently normal before 1 month and later he gives a history of mobile front teeth and difficulty in chewing. He also gives a history of self-fall at the age of 10. No history of paresthesia or pain. The patient gives a history of increased bilirubin level before 6 months and he was not under medication. There was no relevant dental, drug and familial history. On general examination, the patient was conscious, cooperative well oriented to date, time and place. All his vital signs were normal. On extraoral examination facial asymmetry was present due to a mild diffuse swelling in the lower right body of the mandible (FIGURE 1), lips were competent and mouth opening was within normal limits, no tenderness on palpation of joints and no clicking and popping sounds. On examination of the Lymph node, a single right submandibular lymph node was palpable of size approximately 0.5 cm, oval in shape, firm in consistency, mobile and non-tender. On intraoral examination mild vestibular obliteration was seen in the lower labial vestibule extending from 41 to 43 region and on palpation it was non-tender, hard in consistency and non-pulsatile. On hard tissue examination- retained deciduous teeth 53 62 73 and Dental caries – 28 36 37 46 47 was present. To avoid multiple exposures of IOPA, a Panoramic Radiograph was advised to the patient. Panoramic view reveals impacted teeth 13 and 23. An incidental finding of extensive unilocular radiolucency was noticed in the lower body of mandible extending from 33

to 47 region with well-defined sclerotic border and compression of the mandibular nerve was noted. Resorption of the distal root of 46 with thinning of the inferior border of the mandible was seen (FIGURE 3). CBCT was taken for further examination in which axial, coronal, sagittal view (FIGURE 4: A-axial, B-coronal, C-sagittal view) reveals well defined, unilocular, hypodense lesion present in the lower body of mandible with scalloped margins. The buccal cortical bone expansion was noticed with thinning of the inferior border of the mandible. FIGURE 4D reveals a 3D reconstruction view of CBCT that illustrates an extensive lesion in the lower body of the mandible with an impacted tooth. So, a radiographic provisional diagnosis of benign odontogenic cyst in the right lower body of the mandible was given and a differential diagnosis of Odontogenic Keratocyst, Dentigerous cyst and Unicystic Ameloblastoma was given. Further routine blood investigations were made which was non-contributory. Serum Bilirubin: Total-1.22mg/dl, COVID 19- negative and non-reactive to HIV and HBS antigen. Incisional biopsy was taken and a bit of tissue was sent to histopathological investigation. Material received shows sheets of keratin and small bits of bone with hemorrhage (FIGURE 5). Enucleation with Peripheral ostectomy using chemical cauterization (Carnoy's solution) was performed under general anesthesia and the patient was on regular follow-up (FIGURE 6).

DISCUSSION

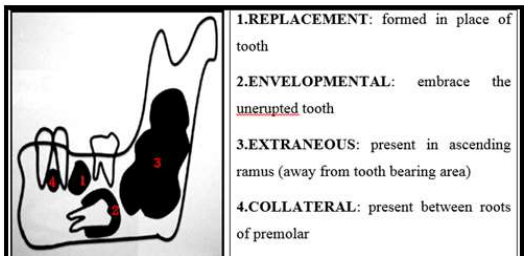
A cyst is a pathological cavity containing fluid, semifluid or gaseous contents and which is not created by the accumulation of pus and is frequently but not always lined by epithelium which was given by Kramer.[8] First described by Philipsen in 1956,[5] later designated by the WHO as a Keratocystic Odontogenic Tumor in 2005 and is defined as 'a benign uni-or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behavior'.[7] It is because of the high recurrence rate, aggressive clinical behavior, association with nevoid basal cell carcinoma syndrome and mutations in the patched (PTCH) tumor-suppressor gene. The 2017 classification reverted to the original and well-accepted terminology of OKC because many papers showed that the PTCH gene mutation could be found in non-neoplastic lesions, including dentigerous cysts, and many researchers suggested that resolution of the cyst after marsupialization was not compatible with a neoplastic process.[9] Pindborg and Hansen (1963) suggested the histological

criteria of OKC, which were confirmed by Browne in 1970 and 1971.[7,10] Toller (1967) suggested that the OKC should best be regarded as a benign cystic neoplasm, rather than an odontogenic cyst.[9]

Most of the time OKC is discovered through routine radiological examination [10], due to its absence of symptoms as it was described in this case report. Here the patient was diagnosed in a panoramic radiograph. Nonetheless, in some cases, OKC can be symptomatic and may cause tooth displacement.[11] Syndromes associated with multiple OKC are Nevoid Basal cell carcinoma syndrome (NBCCS), Gorlin Goltz syndrome, Marfans syndrome, Ehlers danlos syndrome, Noonans syndrome, Orofacial digital syndrome, Simpsongolabi-behmel syndrome.[12]

Radiographically, it may be Unilocular (most commonly): It shows unilocular radiolucency with a well-defined peripheral rim and well-scalloped border. Sometimes it can be related to the crown of an unerupted tooth (pericoronal radiolucency), Minimal or no cortical expansion seen on occlusal radiograph which was in correlation with our present case. Multilocular radiolucent may be due to multiple curved internal septa.

Types of KOT (BASED ON INVOLVEMENT) [12]



Radiographical differential diagnosis are- Dentigerous cyst (40%), Residual cysts, Radicular cyst, Lateral periodontal cysts (25%), Primordial cyst (25%), Globulomaxillary cyst (10%), Unicystic ameloblastoma, A-V malformation, Fibroosseous lesion at initial stages. [12,13]

On aspiration, Dirty, creamy white material with Keratin flakes is seen in the aspiration fluid. A protein content of less than 4 g/100 mL will be present.[15,16]

Histological features given by Pindborg, Phillipsen and Henriksen (Pindborg et al., 1962) suggested series of histological features for the diagnosis of OKC which includes: [5,8]

1. Thin Stratified squamous epithelium lining with ribbon–like appearance typically 8-10 uniform layers thick.
2. Lacks of rete ridges/pegs.
3. Well defined basal cell layer having cuboidal or columnar cells arranged in the palisaded fashion described as "picket fence or tombstone appearance".

4.A thin spinous cell layer which often shows a direct transition from basal cell layer (artefactual separation of epithelium from basement membrane) and spinous cell layer intracellular edema.

5.Surface keratinization which is corrugated and rippled and mostly parakeratosis (keratinized cells with nuclei).

6.Cystic wall composed of fibrous connective tissue which is thin and usually uninflamed.

7.Other findings are satellite cysts, daughter cysts (7- 30%), solid epithelial proliferation, odontogenic rests basal layer budding may be seen. The Fibrous connective tissue wall may get mineralized and may include cholesterol crystals and Rushton bodies.

Recurrence of OKC/KCOT: It has a high recurrence rate ranging from 25-60%.[12] Recurrence rates are reduced when more meticulous surgical treatment is done. The majority of cases reported recurrence within 5 years post-treatment. The mean time of recurrence for males was found to be 4 years and for females, it came out to be 7 years. There have been few cases where recurrence was reported even after 10 years also. So long-term follow-up is necessary.[12]

The causes and factors responsible for KCOT recurrence are 1. incomplete removal of cystic lining 2. Thin and friable nature of epithelial lining, 3. Higher level of cell proliferative activity in the epithelium. 4. Budding in the basal layer of the epithelium 5. Bony perforation. 6. Adherence to adjacent soft tissue. 7. Supraepithelial and Subepithelial split of the epithelial lining. 8. Parakeratinization of the surface layer 9. Remnants of dental lamina epithelium are not associated with the original OKC and development of new OKC in the adjacent area. 10. Growth of new OKC from satellite cyst /daughter cyst/remnants/cell rests. The recurrence rate of the parakeratinized variant is higher than the orthokeratinized variant.[12,17]

The most commonly used therapeutic forms to treat OKC are enucleation and curettage. [11,15] Because it has a thin and friable capsule, it is difficult to remove completely by enucleation, so an aggressive curettage was performed aiming to decrease the recurrence probability. Sousa, et al. [16] and Boyne, et al. [17] in their study affirmed that in some cases, necessary auxiliary therapeutic techniques aiming to decrease those indices assigning chemical and physical methods such as cryotherapy and the Carnoy solution that promote cellular and chemical necrosis respectively. This may significantly decrease this recurrence.

Chemical cauterization [18] is a Composition of 1gm ferric chloride – 24ml absolute alcohol, 12ml of chloroform and

4ml of glacial acetic acid. The Modified Carnoy's solution mixture is of 1gm ferric chloride – 6ml of 100% ethanol and 1ml of glacial acetic acid. A 5-minute application penetrates bone to a depth of 1.54 mm, nerve to a depth of 0.15 mm, and mucosa to a depth of 0.51 mm.

The recurrence rate for various surgical procedures are as below [18,19]:

- Resection – 0%
- Marsupialization – 40%
- Enucleation alone – 26.09%
- Enucleation with Carnoy's solution – 50%
- Enucleation with peripheral ostectomy – 18.18%
- Enucleation with peripheral ostectomy and Carnoy's solution- 0 %

CONCLUSION:

Most of the time OKC is discovered through routine radiological examination, due to its absence of symptoms. In our present case, an incidental finding of OKC in the mandibular body with an impacted tooth was found making it a diagnostic diploma. Hence a thorough clinical, radiographic and histopathological correlations are essential for proper patient treatment and follow-up. This will avoid further complications since KOTs are highly aggressive and have a high recurrence rate.

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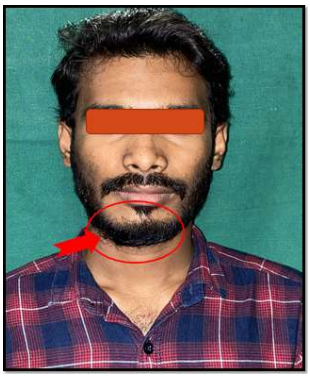

FIGURE 1: PROFILE VIEW OF PATIENT SHOWING FACIAL ASYMMETRY

FIGURE 2: INTRAORAL EXAMINATION

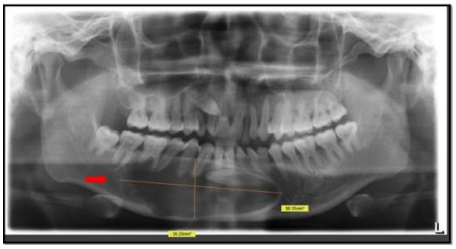

FIGURE 3: PANORAMIC RADIOGRAPH SHOWS UNILOCULAR RADIOLUCENCY IN LOWER BODY OF MANDIBLE

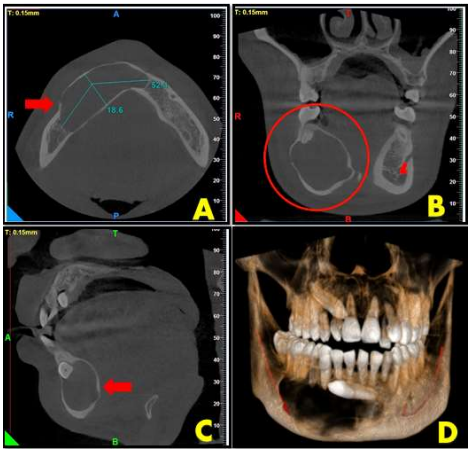

FIGURE 4: A- AXIAL, B- CORONAL, C- SAGITTAL, D- 3D RECONSTRUCTION VIEW OF CBCT

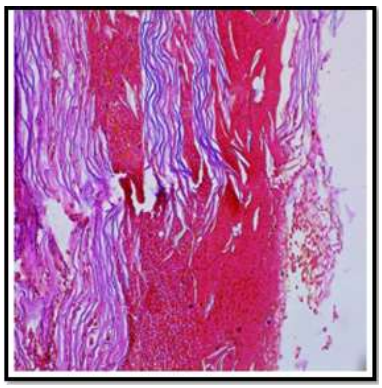

FIGURE 5: MICROSCOPIC EXAMINATION

FIGURE 6: OPERATIVE SURGICAL IMAGE OF THE PATIENT