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Collateral Odontogenic Keratocyst: A Diagnostic Dilemma

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Abstract: Benign lesions of the jaws are quite uncommon (17%) of which odontogenic keratocyst holds the most uniqueness due to its specific clinical characteristics, histological features and aggressive biological behaviour. Presenting the readers with a distinctive case report of a rare case of odontogenic keratocyst-collateral type, which initially appeared to be a lateral periodontal cyst. Furthermore profound radiological examinations lead to a diagnosis of collateral odontogenic keratocyst.

Keywords: Cyst, OKC, Collateral Odontogenic Keratocyst, Aggressive behavior, Sclerotic borde.

1. INTRODUCTION

The term odontogenic keratocyst [OKC] was first described by Philipsen in 1956. As the name depicts, it is a cystic lesion of benign nature commonly located in the maxilla and mandible with a significant potential for growth, expansion and local invasion that accounts 12-14% of all odontogenic cysts of the jaws, with posterior region of the mandible having the maximum predilection.(1)OKCs takes its origin from the dental lamina and contains a cystic space containing desquamated keratin with a uniform lining of parakeratinised squamous epithelium. OKCs report widely shows male predominance with a peak of incidence in the third decade of life. They are characterised by an aggressive behaviour with a relatively high recurrence rate, particularly when OKCs are associated with syndromes(2)). Radiographically, OKC may reveal a small unilocular radiolucency to a large multilocular radiolucency and therefore it may resemble ameloblastoma, dentigerous cyst, lateral periodontal cyst, and radicular cyst making its diagnosis a challenge (4). The most customary clinical presentations are swelling, discharge, and pain, but in some cases OKC can be asymptomatic and thus the frequency of a casual diagnosis ranged between 5.5% and 42.5%.(3)

2. CASE REPORT

A 22 year old male patient reported to the out patient department with a complaint of extra oral swelling for the past 3 months.

On extra oral examination a diffuse swelling was evident on the left side middle third of the face obliterating the nasolabial fold. Patient gives a history of gradually increase in the size of swelling within 3 months(Figure 1). Intraoral examination revealed a discrete swelling in the interdental region of 23 and 24 measuring of size 2x 2 cms obliterating the buccal vestibule. A diffuse swelling approximately of size 3x 2.5 cms was present on the hard palate extending anteroposteriorly from the 23 to 26 and mediolaterally from the attached gingival of 23,24,26 region till the midpalatine raphae . Palpation revealed a swelling which was soft to firm, tender and non compressible . (Figure 2) .

Intraoral periapical , lateral maxillary occlusal projection disclosed a homogenous radiolucency with well defined sclerotic border along with knife edged resorption of the roots of 24, 25, 26. (Figure 3) .Panoramic radiograph revealed a well defined homogenous radiolucency with sclerotic corrugated border involving 24,25,26,27 and left maxillary sinus. (Figure 4) .Computed tomography was taken that disclosed an increased attenuation in the cystic cavity in both coronal and axial views with hypodense area involving the floor of the orbit ,nasal cavity and breeching the hard palate.(Figure 5) . All these positive findings lead to a provisional diagnosis of OKC.

Patient was then recalled after blood investigations for biopsy. Excisional biopsy was performed under Local anesthesia.Tissue retrieved under aseptic conditions and sent for histopathological examination. Histopathological features revealed a epithelial layer with a corrugated para- or orthokeratinized luminal layer and a prominent basal cell layer and without rete ridges (Figure 6). Correlating with histopathological feature a final diagnosis of Odontogenic Ketatocyst – Collateral type was arrived. For further management patient was referred to Department of surgery.

3. DISCUSSION

Continual research advanced to reclassify odontogenic keratocyst a benign tumour into a cystic lesion in the year 2017 because of which odontogenic keratocyst is still a topic for discussion and research (5). The WHO working group recommended that OKC should be replaced by keratocystic odontogenic tumor (KCOT) for the following reasons: 1. Aggressive behavior; 2. Occurrence of a solid variant; 3. Possibility of recurrence; and 4. Mutations of the PTCH gene (6). OKCs represent 2-11% of all odontogenic cysts, affect all ages with a peak from the 2nd–4th decade and have a male to female ratio of 2:1 (7) .Genetic influence can be due to defects in long arm of 9q22 3q31 especially when associated with syndromes (8).The growth of cysts as described by Scharfetter in the year 1989 can be an active growth or passive growth or due to bone Resorption (8).The aggressive behaviour of the cyst has always been a puzzle as its expansion is based on the osmotic tension exerted on the adjacent tissues along with the inherent forces from within the epithelium or due to the enzymatic activity from the fibrous wall (5).In the case presented here, similar aggressive expansion is noted in a small duration of three months, thus attaining the basic characteristic of an OKC.

Inorder to explain the aggressive clinical behavior of keratocysts several hypotheses have been proposed, including interluminal hyperosmolality and epithelial proliferation. This epithelial expansion manifests itself by an eccentric and multilocular growth pattern possibly explained by an increase in mitotic activity within the cyst lining. Donoff et al demonstrated

the presence of collagenase and its enzymatic degradation within the cyst wall may contribute to expansion(9).It is also possible for a OKC to have malignant potential . The neoplastic concept of OKC is aided by molecular studies that verified loss of heterozygosity. Studies instituted evidence of allelomorphic loss mainly in the p16, p53, PTCH, MCC, TSLC1, LTAS2, and FHIT genes(10) and indistinct borders in radiographs (6) . In case of malignant transformation into squamous cell carcinoma PET/CT will typically reveal intense FDG hypermetabolism similar to other head and neck squamous cell carcinomas(11) .OKCs infrequently cause root resorption of adjacent teeth unlike other odontogenic lesions having similar aggressive behaviour such as ameloblastomas (2). Radiographically, OKCs presents as a well-defined unilocular or multilocular radiolucency bounded by corticated margins . Unilocular lesions are more predominant compared to the multilocular variant and is observed in approximately 30% of cases, most commonly in the mandible(2). In the presented case we could appreciate an unilocular radiolucency rather than a multilocular one . Odontogenic keratocyst can be classified into four types based on its radiological position, they are: 1. Replacement type : Cyst which forms in the place of normal teeth. 2. Envelopmental type: Cyst which embraces an adjacent unerupted tooth. 3. Extraneous type: Cyst which occur in asending ramus away from the teeth. 4. Collateral type : Cyst which occurs adjacent to the root of teeth which are indistinguishable radiologically from lateral periodontal cyst.(12)Our case owns the radiological features of collateral OKC which presents in between the roots and has an extensive expansion leading to the resorption of the adjacent roots thus differentiating OKC from lateral periodontal cysts , dentigerous cysts, radicylar cysts and ameloblastoma is complex . Lateral periodontal cysts are developmental odontogenic cysts that most commonly occur in the mandibular premolar area and are incidental. Radiographically they appear as well-circumscribed lytic lesions associated with the tooth root in the premolar mandibular region whereas Odontogenic keratocyst typically occur in the body and ramus of the mandible in association with an impacted tooth ,over again allowing the dilemma between OKC and dentigerous cyst .Histopathology is always the gold standard to diagnose OKC. MRI shows a, minimally peripherally enhancing thin-walled cyst with heterogeneous intensity fluid contents (intermediate T1 signal and inter-mediate to high T2 signal) because of variable proteinaceous content . MRI can help distinguish OKC from ameloblastoma, the latter of which has a mixed pattern of solid and cystic components, irregular thick walls, and avid enhancement of solid components. (11)F. Giovacchini et al describes that clinical or radiological characteristics or the treatments may influence their recurrence , of which OKC has a particular tendency to recur after surgical treatment to a higher extent.(7) The increased recurrence of OKC in syndromic patients over nonsyndromic is noticable. (13)Nick Blanas et.al (2008) stated, the treatment most likely to prevent recurrence is resection with a 0% recurrence rate. When the presence of an OKC (KCOT) is confirmed by examination of a biopsy specimen, 3 types of treatment can be done. 1) For a routine OKC (KCOT) in a person who is likely to return for follow-up treatment, Carnoy's solution appears to be the least invasive procedure with the lowest recurrence rate. 2) If the cyst is very large decompression of the cyst followed by enucleation will also have a reduced recurrence rate. Use of Carnoy's solution at the enucleation stage should be considered. 3) If the patient is unlikely to return for follow- up, lesion should be resected (14). Accoring to Daroit et al the recurrence of OKC is affected by 1) enucleation with or without rupture of the cystic capsule; 2) the surgical approach used (conservative approaches such as enucleation and marsupialization vs. aggressive approaches such as total resection); 3) the use of adjunct therapy (e.g., Carnoy's solution, cryotherapy, peripheral ostectomy) (15). Within 2 days of explanting the OKCs, growth of epithelial cells and

fibroblast like cells, which showed moderate-to-high activity of NADH-diaphorase and acid phosphatise has been reported. (10).It is suggested that, residues of OKC lining which are left behind at surgery are more likely to recur due to the apparently high metabolic activity of the epithelium which is demonstrated by the high activity of oxidative enzymes representing glycolytic, citric acid, and pentose phosphate shunt mechanisms and are likely to be factors of importance to consider high incidence of OKC recurrences.

FIGURES:

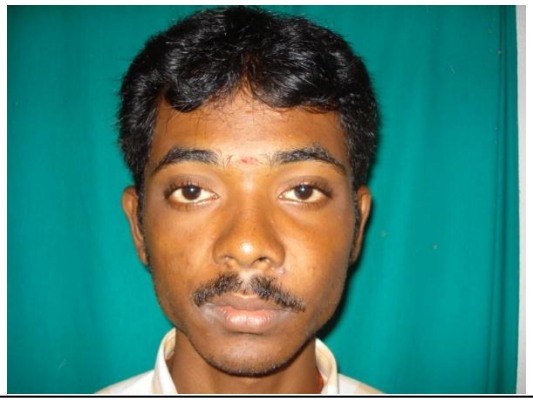

Figure 1: Extraoral picture revealing diffuse swelling on the right middle third of face

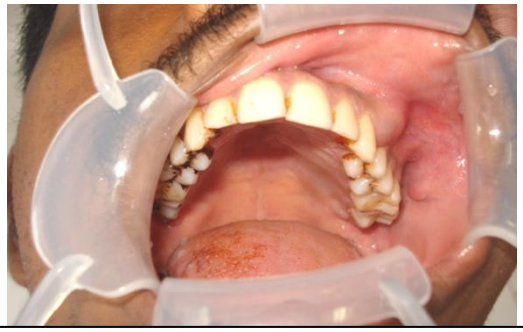

Figure 2: Intraoral picture revealing discrete swelling in the interdental region of 23 and 24

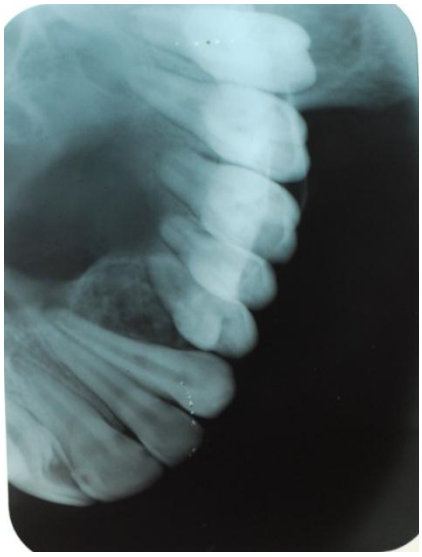

Figure 3 : Lateral maxillary occlusal projection

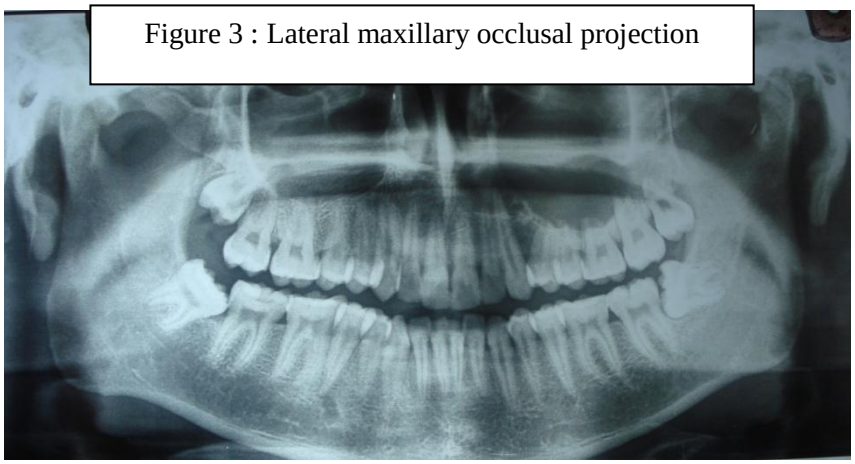

Figure 4: Panoramic radiograph revealing a well defined homogenous radiolucency with sclerotic corrugated border.

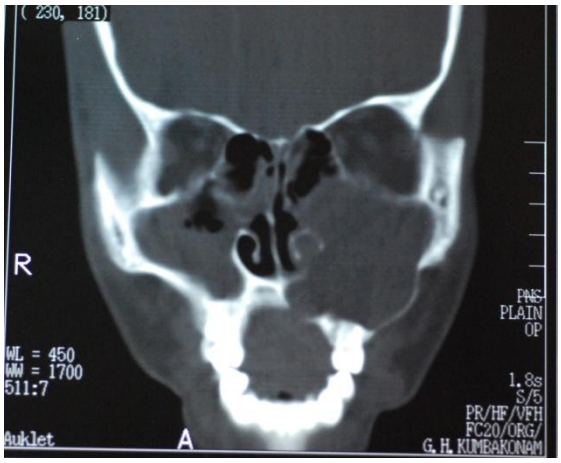

Figure 5: Computed tomography coronal view showing increased attenuation in the cystic cavity

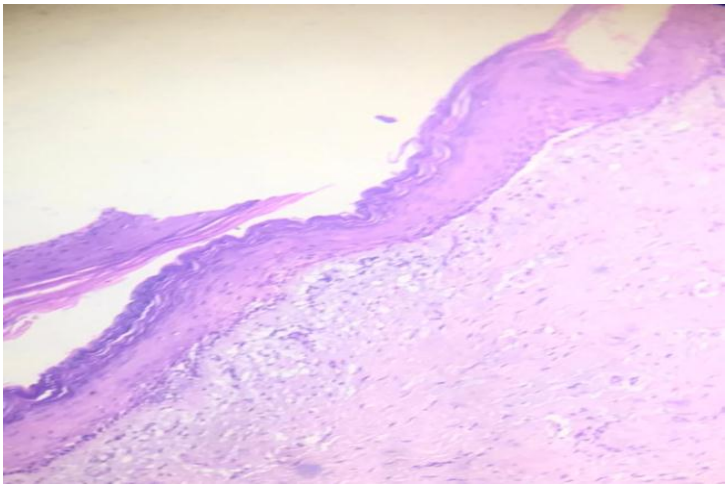

Figure 6: Histopathology showing corrugated surface epithelium with polarised basal layer and keratin HE x10.

4. CONCLUSION

Collateral odontogenic keratocyst pertaining to its presentation and position can be easily misdiagnosed as other cystic lesions which has nearly similar appearance. Moreover okc does not have any pathognomonic characteristics making its diagnosis even more challenging. Treatment of OKC requires arduous efforts with recurrence as a matter of concern. However long-term follow-ups with periodic radiographic examinations are essential to avoid recurrence.

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