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Non-syndromic Bilateral Odontogenic Keratocyst of the Mandible with Diverse Histopathological Features - An Unusual Case Report

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ABSTRACT

Odontogenic Keratocyst (OKC) is a cyst originating from the teeth & it's associated structure, exhibiting aggressive clinical behavior. From the clinician's point of view, its heterogeneous presentation and high frequency of recurrence make it unique. It is aggressive with a comparatively high frequency of recurrence. Most often discovered by chance during routine radiographic examination, usually do not significantly enlarge the jaw bone; instead, expand antero-posteriorly. OKCs generally occur in the second to fourth decades of life and uncommon in younger patients. Even more rare is its presentation bilaterally, & we present one such case of a young male patient,16-years-old.

Keywords: Odontogenic Keratocyst, Mandible, Unerupted Molar, Odontogenic cyst.

INTRODUCTION

Odontogenic Keratocyst (OKC) is a developmental odontogenic cystic lesion that arises from odontogenic epithelium and has an aggressive growth potential and high recurrence rate [1]. Prior to 2005, OKC was designated as keratotic odontogenic tumor (KCOT) by the World Health Organization. But in 2017, it was renamed odontogenic Keratocyst once more [2]. Clinical and radiological examinations of odontogenic cysts reveal similar features. The envelopment type of Odontogenic Keratocyst (OKC) is frequently misdiagnosed as a dentigerous cyst because it has neighboring or with relation to unerupted teeth and radiographs may show a radiolucent cystic lesion surrounding the crown of the teeth. Therefore, the gold standard diagnosis is histological examination [3]

CASE REPORT:

A 16-year-old boy who had been experiencing pain in the area of his lower left back teeth for a month came to the Department of Oral Medicine and Radiology. Pain was sudden in onset, intermittent, localized, mild to moderate in intensity and throbbing in nature. Not associated with any other symptoms. Bilateral submandibular lymph nodes were palpable, enlarged, tender, firm in consistency and mobile in nature. On extra-oral examinions, no gross facial asymmetry was observed. (Figure- 1).

Fig- 1, Facial Profile

Intra oral examination, revealed mild vestibular obliteration w.r.t 35, 36, 37 in the left mandibular region, which was tender on palpation with cortical plate expansion buccally and lingually. No fluctuation, mobility, compressibility, translucency, and no palpable pulsation was noted. (Figure-2a, 2b)

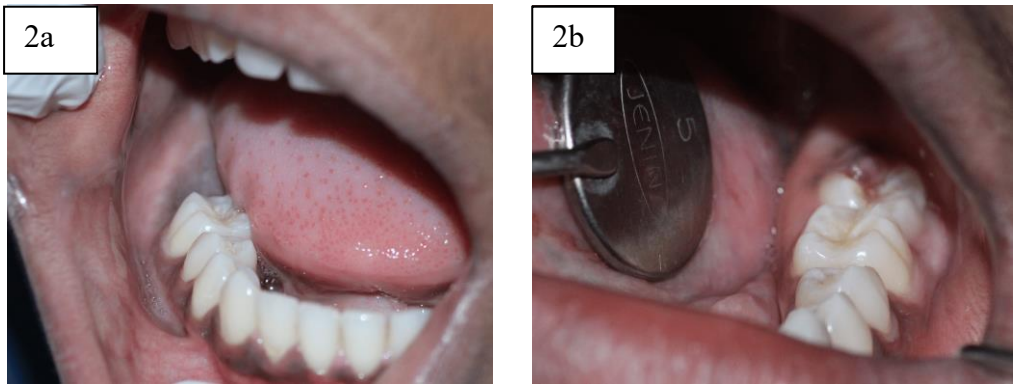


Fig-2a & 2b, Intraoral right and left mandibular posterior teeth region.

Provisionally diagnosis of benign odontogenic lesion on the left mandibular posterior teeth region was considered. Differential Diagnosis of dentigerous cyst, was given due to young adult age group, no symptoms of acute or chronic inflammation and clinically missing left mandibular 3rd molar, and bucco-lingual cortical plate expansion of the lesion and odontogenic Keratocyst because it occasionally may develop in association with an unerupted or impacted tooth follicle. Fine needle aspiration fluid from left molar teeth region was a serosanguinous and creamy-semisolid type. (Figure-3).

Fig-3, Fine needle aspiration from the left mandibular posterior teeth region

The cytosmears were stain with H & E and showed few epithelial cells, RBC, acute and chronic inflammatory cells (Neutrophils) & tissue debris suggestive of odontogenic lesion and total protein estimation value was 5.20 g/dl suggestive of dentigerous cyst.

Radiological investigation through Orthopantomogram, reveal complementation of permanent dentation except complete apical formation of all third molars. Radiolucency seen bilaterally in the mandibular posterior teeth region. On the left side, radiolucency noted in the mandibular posterior teeth region, 2x3 cm in size, involving 37 and formed tooth bud of 38. Extended from the distal root of 36 to the distocoronal surface of 38 antero-posteriorly and superoinferiorly from the external oblique ridge to the inferior border of mandible with well-defined corticated margin. Loss of lamina dura in the periapical region of 37 and on the right side, radiolucency involved alveolar and basal bone of mandible. Epicenter is located superior to the inferior alveolar canal. Well-defined and corticated periphery with

smooth curved septa involving unerupted 2nd molar 3rd molar that had complete formation of coronal third and furcation area. Extended antero-posteriorly from distal root of 1st molar to disto-coronal surface of 3rd molar and superoinferiorly from superior to inferior border of mandible. No external root resorption & displacement of inferior alveolar canal. (Figure- 4)

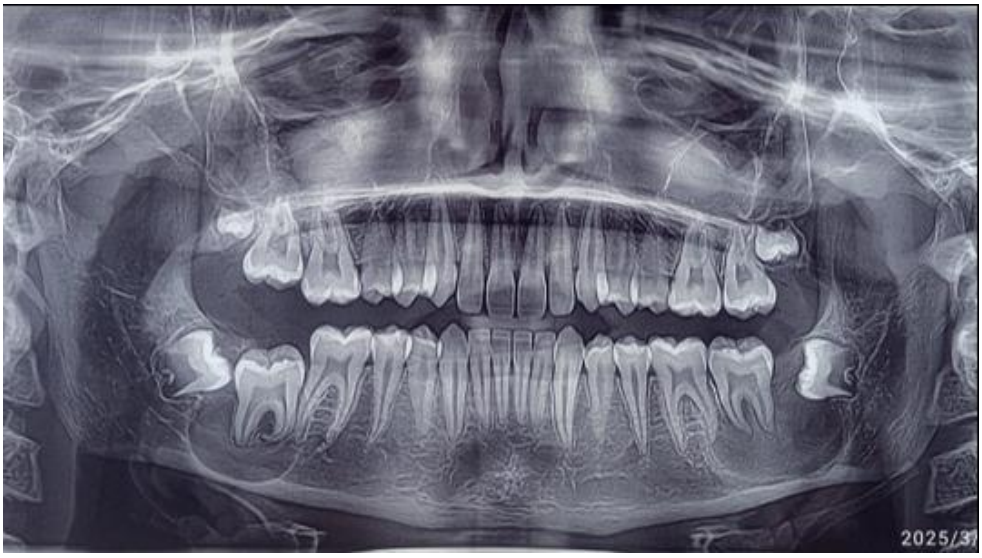
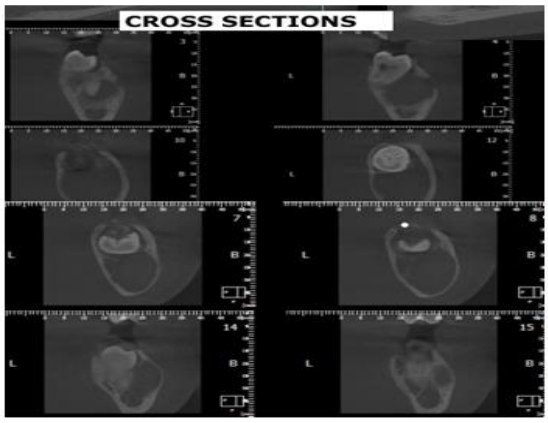
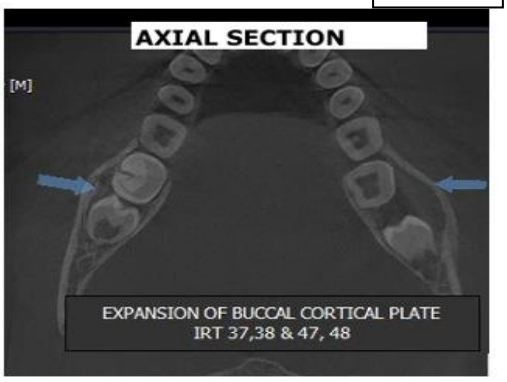


Fig-4, Orthopantomogram

Patient also subjected for Cone beam computed tomography. It revealed, osteolytic expansile lesion of in the mandibular posterior teeth region bilaterally with (28.3x 26.0 x17.5) mm in size on left and right mandibular posterior teeth region shows radiolucency with (23.2 x 25.8 x13.1) in size & buccal cortical plate expansion in the right and left mandibular 2nd & 3rd molars. Internal structures show unerupted 3rd molars and the right and left mandibular 2nd molars. There was no obvious resorption of the teeth root, No notable displacement of inferior alveolar canal, bilaterally. Correlating history, clinical and imaging findings indicated dentigerous cyst as the lesion develops in the peri-coronal site and differential diagnosis as OKC was made. (Figure-5a,5b)



5a



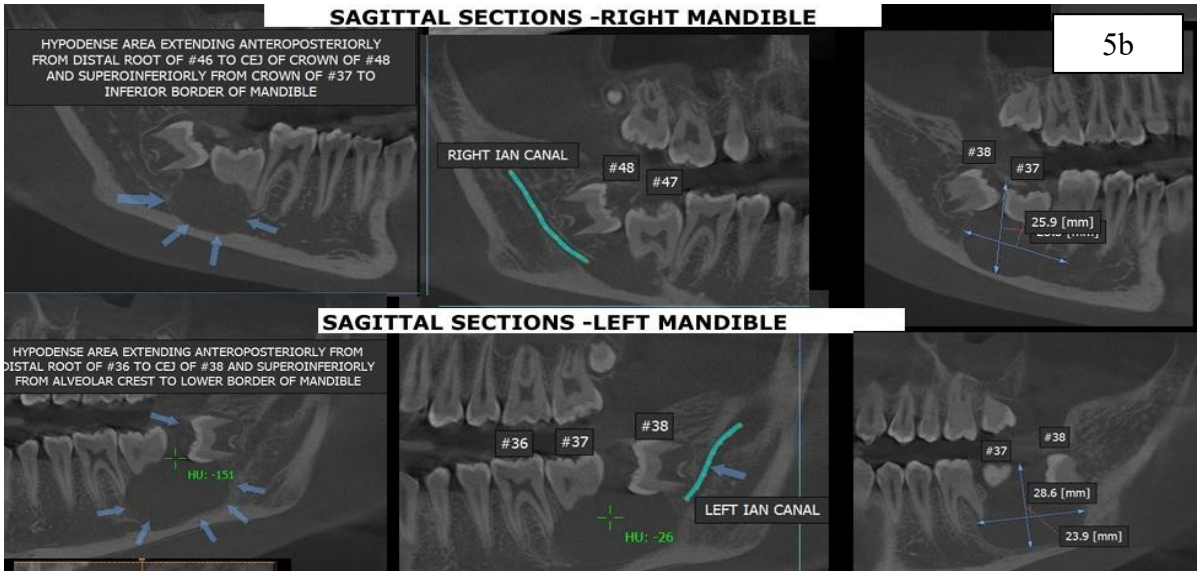


Fig- 5a,5b, CBCT findings

Patient was operated surgically, Cystic lining along with circumscribed mandibular 2nd & 3rd molars was removed bilaterally. (Figure- 6)

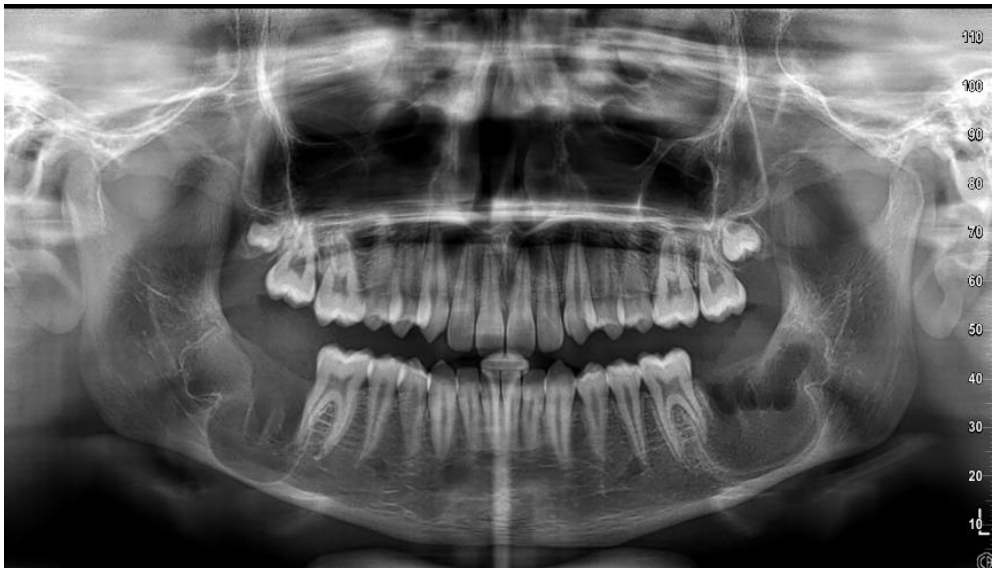


Fig- 6, Post-operative OPG

On histopathological examination, a section of the left cystic lesion showed collagenous wall of the cyst. Denuded lining epithelium. Stained areas show stratified squamous epithelium (with many layers of cells). Stratified squamous epithelium with long rete ridges, associated with thick lymphoplasmacytic infiltrate and hemosiderin deposition. The fragments of rete ridges are incorporated deep into the collagen matrix. The uninflamed areas show a thin squamous epithelium and no keratinization. The focal area shows the Rushton bodies inside the inflamed epithelium. Bone fragments are observed in the deep to collagenous stratum. The absence of mucin-secreting cells in the epithelium and sections of the right-side of the cyst showed a wall of the cyst with a moderate collagenous stroma. Non-keratinizing squamous epithelium lining. Focal areas show elongated rete ridges and mild lymphoplasmacytic infiltration. Epithelial giant cells and absence of mucous cells were observed, which led to the diagnosis of odontogenic Keratocyst based

on histopathological characteristics. Patient was kept under follow-up, after 6 months of surgery satisfactory bone heling was noted in the right & left mandibular posterior teeth region. (Figure- 7)

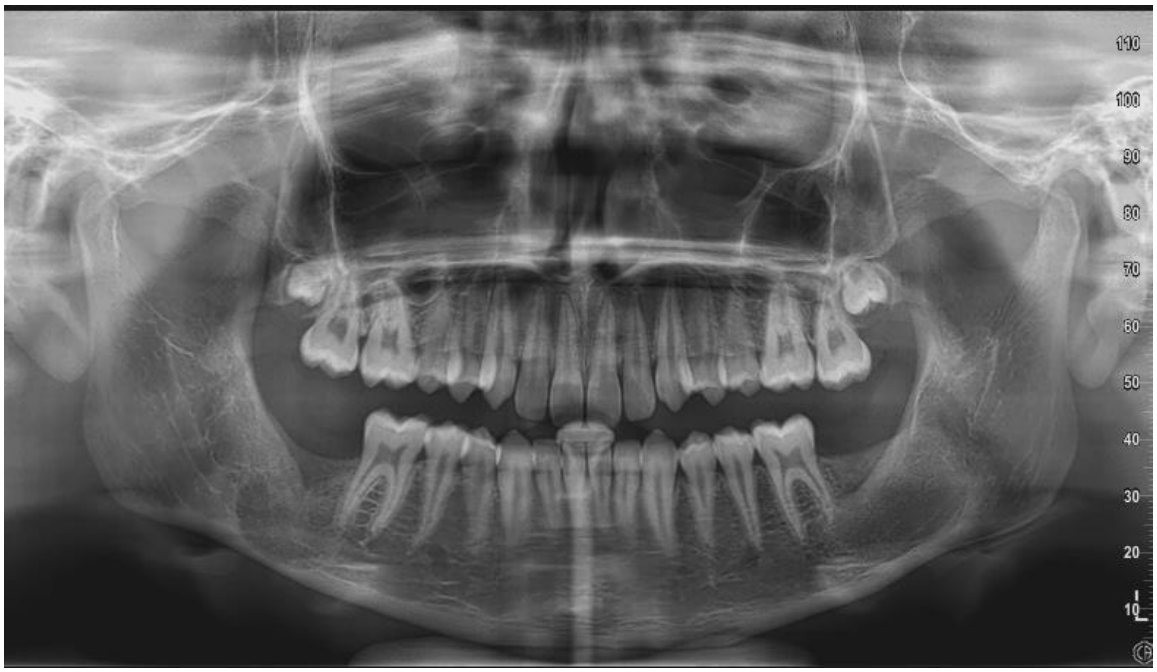


Fig-7, OPG -after 6 months follow-up.

DISCUSSION

"Odontogenic Keratocyst" was first mentioned in a 1956 paper by Philipsen. As demonstrated in the paper by Pimpalkar et al., the occurrence of OKC is a rare phenomenon in and of itself, and its bilateral occurrence is even rarer [4]. The remnants of the tooth germ or dental lamina give rise to the benign but aggressive intra-osseous tumor known as an Odontogenic Keratocyst (OKC). It has a significant potential for tumor development and recurrence [5]. The majority of the time, odontogenic keratocytes are isolated lesions. Nervous Basal Carcinoma Syndrome (NBCCS), Gorlin-Goltz Syndrome, Orofacial Digital Syndrome, Ehler-Danlos Syndrome, Noonan Syndrome, and Golabi-Behmel Syndrome are all frequently associated with multiple odontogenic keratocytes [6]. Multiple odontogenic keratocysts, calcification of the falx cerebri, bifid ribs, and nevoid basal cell carcinomas of the skin are among the characteristics of NBCCS. Skeletal, cutaneous, neurological, ocular, and sexual abnormalities can coexist with odontogenic Keratocyst when NBCCS is present. But in our situation, these characteristics were absent [7]. OKC typically affects adults between the ages of (30 to 50) years, making up 14.3% of all odontogenic tumor cases; however, in our instance, the patient was 16 years old. OKC frequently exhibits no symptoms or manifestations in its early stages. As in our case, swelling is the typical clinical manifestation. In many instances, patients are remarkably free of symptoms until the cysts have reached a large size, this occurs because the odontogenic Keratocyst tends to extend in the medullary cavity and clinically observable expansion of the bone occurs late [7]. However, in our case, cortical platelets were observed to be expanding on the left side. Patients occasionally develop symptoms such as pain, abnormal feeling, discharge of pus and loose teeth [8]. Mandibular Ramus, the anterior mandible, the third molar region in the maxilla, and the canine area are the most frequently occurring sites [9]. (25 to 40) % of the cases involve an unerupted tooth. OKC can

manifest radiologically as small, unilocular radiolucent areas that are round or ovoid. The jaw may become multilocular radiolucent due to larger lesions. Even though bone expansion is rare, it can happen in a significant percentage of cases, particularly at the angle or in the ramus area [10]. It usually appears on radiograph as a dentigerous cyst in the mandible. Rarely, they are connected to the tooth crown and may block the related tooth eruption [11]. Radiographically, the lesions in our case showed up as a dentigerous cyst-like unilocular radiolucency linked to an unerupted tooth. FNAC plays an important role in intraosseous jaw lesions. Cytosmear of OKC shows greasy, pale fluid with keratotic debris and total protein content <4 .0 g %. A cystic lumen lined with six to eight layers of uniformly thick corrugated para-keratinized stratified squamous epithelium is the histopathological characteristic of OKC. The lining epithelium is distinguished by a basal layer of tall columnar cells with reversed polarity and palisading nuclei. The interphase of epithelial connective tissue is flat, and satellite cysts or odontogenic epithelial islands may be present in the connective tissue wall [12]. In addition to these traditional characteristics, the focal area displays Rushton bodies inside inflammatory epithelium. Deep to collagenous stroma, bone fragments were observed. The supra-basal layers have a higher frequency of mitotic figures. OKCs connected to the NBCCS exhibit noticeably higher levels of mitotic activity [13]. The epithelial lining with elevated cell proliferative activity, the epithelial basal layer exhibiting budding, the para-keratinized surface layer, the supra-epithelial split, the subepithelial split, the presence of remnants or cell rests, satellite cysts or daughter cysts, folded epithelium, and thin friable lining are the primary histological characteristics that can be taken into consideration to predict the recurrences of OKC [14]. The goal of OKC management is to minimize the patient's morbidity while simultaneously lowering the chance of recurrence. The optimal treatment approach is currently up for debate. Enucleation, enucleation with cryotherapy, enucleation with application of Carnoy's solution, marsupialization, decompression, and resection are among the treatment modalities for OKC that produce varying recurrence rates [15].

CONCLUSION

Odontogenic Keratocyst is a pathological condition that is recognized worldwide, because of its aggressive behavior. It is necessary to take into account the possibility of NBCCS in any patient with multiple odontogenic Keratocyst. To identify any characteristics linked to this syndrome, a thorough clinical examination and histopathologic analysis must be carried out.

Declaration by Authors

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