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Case Report

Orthokeratinised Odontogenic Cyst: A Diagnostic Havoc

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ABSTRACT

This case report presents a 27 year old male patient who reported with pain in his anterior mandible since a week. History revealed an incidence of trauma on his chin when he was young for which he didn't undergo any treatment. On examination buccal cortical expansion was clinically evident and tenderness was elicited on percussing over the lower anterior teeth. Aspiration was negative initially while it showed serous blood the second time. Radiograph featured an unilocular radiolucency extending from distal of left mandibular first premolar to mesial of right mandibular first molar with an impacted right mandibular canine was seen. Displacement of teeth and inter-radicular scalloping were also evident. With all these clinical and radiographic features we could arrive at a provisional diagnosis of odontogenic keratocyst. A differential diagnosis of traumatic bone cyst and ameloblastoma were also considered. A Complete surgical excision with curettage of the cystic cavity was performed following which histopathological evaluation confirmed the lesion to be an orthokeratinised odontogenic cyst.

Keywords: Diagnostic challenges, Odontogenic keratocyst, Orthokeratinised odontogenic cyst.

INTRODUCTION

Orthokeratinised odontogenic cyst is a rare phenomenon of developmental odotogenic cyst first described by Wright in 1981 with incidence of 7%-17%. Orthokeratinsed odontogenic cyst was considered to be a variant of odontogenic keratocyst by Philipsen in 1945 however WHO stated it to be a separate distinct entity owing to its clinical and pathological presentations [1,2]. The choice of treatment for this lesion is principally enucleation followed by curettage. Prognosis after enucleation is substantially excellent with low recurrence rates of less than 2% [3]. Our case report discusses the assorted diagnostic challenges due to the diverse clinical and radiological presentations mimicking different pathologies occurring in the anterior mandible.

CASE PRESENTATION

A 27 year-old male patient reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of pain in the anterior tooth region of mandible since 7 days. Pain was sudden and intermittent in nature increases during mastication with a feel of loosened teeth. Patient also had a history of trauma on his chin when he was young and he was not treated for the same (**Figure 1**).

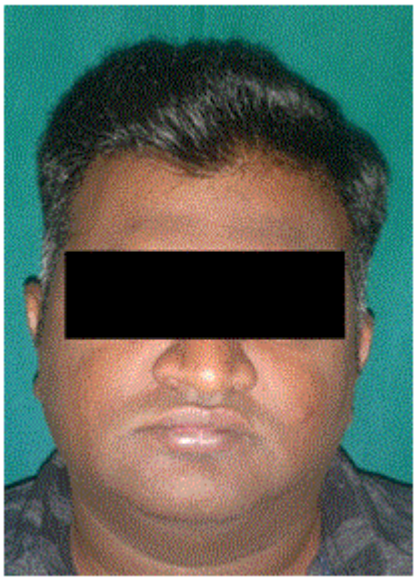

Figure 1: Frontal profile view showing diffuse swelling in right side of face.

Extra orally there was a diffuse swelling in the right side of the face. Intraoral examination showed obliteration of the mandibular vestibule extending from the mandibular right first molar to the left first premolar. On palpation, expansion

of buccal cortical plate was evident. The teeth in the mandibular arch were tender on percussion and vitality test revealed that all the teeth were vital (**Figure 2**).

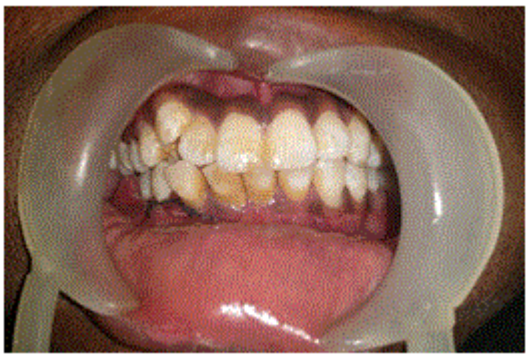

Figure 2: Intra oral view showing obliteration of buccal vestibule.

Orthopantomograph (**Figure 3**) showed presence of a large unilocular radiolucent lesion extending from the lower left first premolar to the lower right first molar. Superoinferiorly it extends from the root apices of the tooth involved in the lesion to the inferior border of mandible. However, the continuity of the lower border of mandible was maintained. The roots of all the teeth involved in the lesion showed resorption. There was an empty aspiration initially and there was serous blood during next aspiration performed in different angulations. On the basis of the clinical and radiological features the lesion was provisionally diagnosed as odontogenic keratocyst. Treatment for the diagnosed lesion was planned for surgical excision of the lesion followed by curettage of cystic cavity.

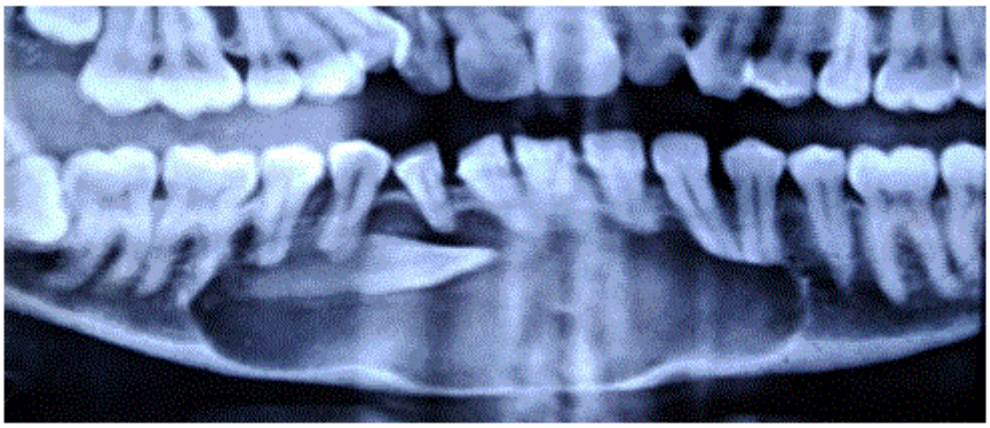

Figure 3: Orthopantamograph showing unilocular radiolucency extending from distal of left mandibular first premolar to mesial of right mandibular first molar with an impacted right mandibular canine, inter-radicular scalloping, and displacement of teeth and resorption of roots.

SURGICAL PROCEDURE

Under Nasotracheal intubation general anesthesia was administered. Standard painting and draping was done. Intra oral betadine irrigation was done and local anesthesia was infiltrated. Crevicular incision was placed with vertical releasing incision in the molar region bilaterally. Mucoperiosteal flap was raised to expose the underlying bone. Thinned buccal cortex was removed to expose the underlying lesion. The lesion seemed to be encapsulated and soft in consistency. A slit was made on the capsule to reveal yellowish-white cheesy material (**Figure 4**). After evacuating the cheesy material, cystic lining was seen which was separated from the bone and removed in toto along with the impacted right mandibular canine (**Figures 5 and 6**). Lingual cortex perforation was evident after complete excision of the cyst (**Figure 7**). The cystic cavity was curetted and washed with betadine. The flaps were sutured with 3-0 vicryl. Patient was extubated and shifted to ICU uneventfully (**Figure 8**).

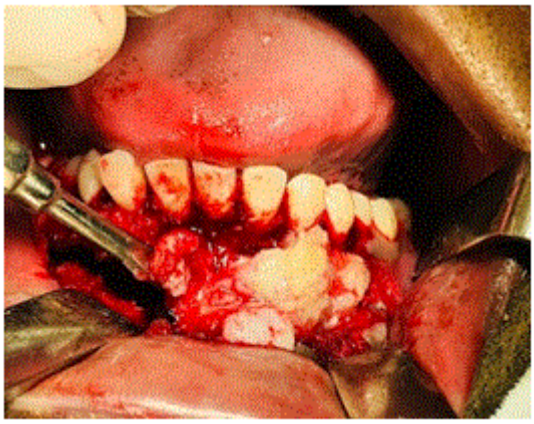

Figure 4: Thinned buccal cortex removed to expose the underlying lesion.

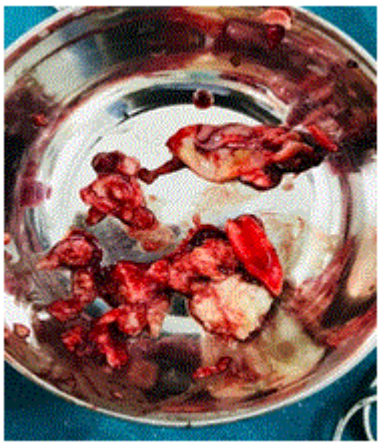

Figure 5: Thick yellowish cheesy content retrieved from the cystic cavity with the impacted tooth.

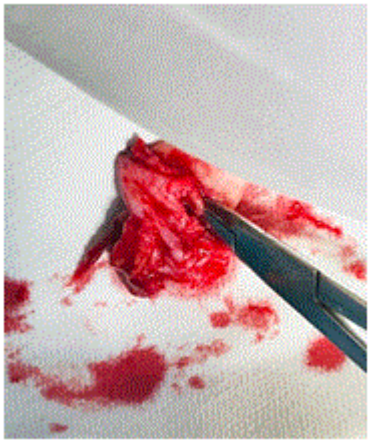

Figure 6: Cystic lining removed in toto.

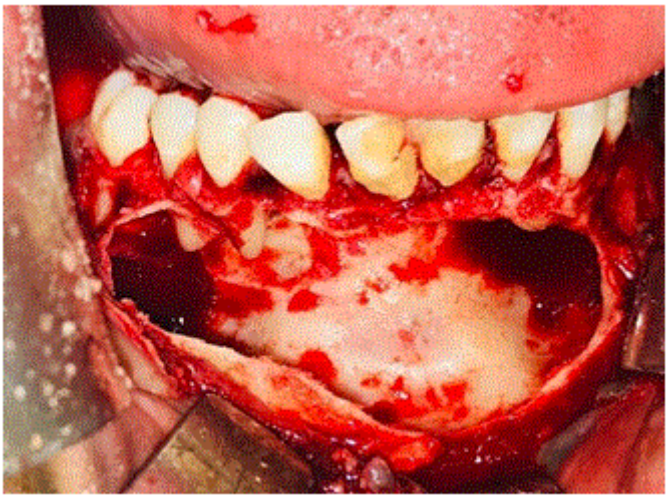

Figure 7: Cystic cavity with thinned out lower border and perforation of lingual cortex.

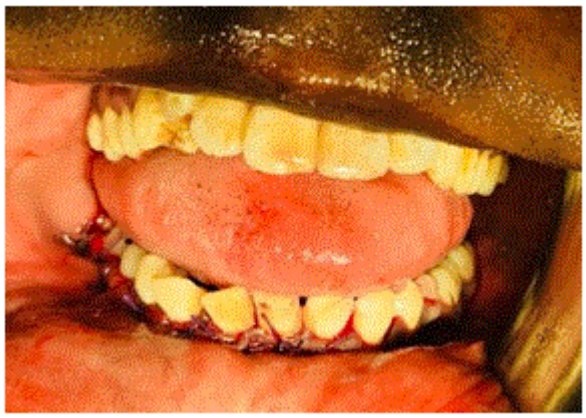

Figure 8: Flap closure with 3-0 vicryl.

Macroscopically, three brownish white soft tissue specimens measuring approximately (28 × 30 × 8) mm^3, (18 × 12 × 6) mm^3 and (14 × 12 × 2) mm^3, five yellowish white hard tissue specimen measuring approximately (6 × 25 × 2) mm^3, (5 × 28 × 2) mm^3, (3 × 15 × 2) mm^3, (7 × 10 × 2) mm^3 and (9 × 23 × 8) mm^3 and one yellowish white soft tissue specimen were submitted for histopathological study.

Histologically (**Figure 9**), hematoxylin and eosin stained soft tissue section showed orthokeratinised stratified squamous epithelium of uniform thickness with prominent granular cell layer. Areas showed separation of epithelium from the adjacent connective tissue wall. Cystic lumen contains keratin flecks giving onion skin appearance. Connective tissue wall is made up of fibrocollagenous tissue infiltrated with chronic inflammatory cells predominantly lymphocytes and plasma cells, suggestive of orthokeratinised odontogenic cyst.

DISCUSSION

Our case report experienced high diagnostic challenges due to varying clinical and radiographic presentation that is similar to various other cystic pathologies in literature. Based on the clinical and radiographic findings elicited in our study we have provisionally diagnosed the case as odontogenic keratocyst.

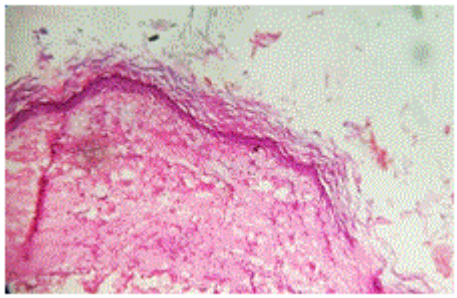

Figure 9: Hematoxylin and eosin section showed orthokeratinised stratified squamous epithelium of 6-8 cell layer thickness and prominent granular cell layer in association with fibrous connective tissue wall.

Traumatic bone cyst is an uncommon and asymptomatic lesion of the jaws which is diagnosed accidentally during radiological examination with posterior mandible being the most common site of occurrence. The etiopathogenesis of the lesion is still debated, though the role of trauma is often associated. Radiographically traumatic bone cyst presents as a unilocular radiolucent with inter-radicular scalloping and thinning of cortical plates. Aspiration is either empty or at times even serous blood can be obtained. The clinical and radiographic findings of traumatic bone cyst were concurrent with features elicited in our case. However, cystic lesion associated with impacted tooth remains a characteristic feature of orthokeratinised odontogenic cyst [4-6].

Dentigerous cysts are the most predominantly occurring developmental odontogenic cysts associated with impacted tooth usually affecting the middle-aged individuals. The cyst remains asymptomatic and discovered unexpectedly through a routine radiographic investigation which is characterized by its attachment to the crown of an unerupted tooth. The exact pathogenesis of dentigerous cyst is uncertain, however it was accepted that the cyst originates from epithelial remnants of tooth-forming organs. Only the radiographic findings of this cyst were consistent with the features of our case. But still histopathological investigations would be a confirmatory diagnosis to rule out dentigerous cyst [6,7].

Central giant cell granuloma is another uncommon destructive and locally aggressive lesion affecting anterior region of mandible and most often seen crossing the midline. It generally affects adults>30 years with female predilection. Clinically it is a painless, slow growing mass with expansion and thinning of cortical plates, and displacement of tooth is noted without resorption of roots. Central giant cell granuloma has high recurrence rate of 15%-20% which is based on the aggressiveness of the lesion [7].

Ameloblastoma, a odontogenic tumor which is benign in nature that presents as a slow growing, painless swelling with expansion of cortical plates, is also associated with paresthesia of lip and loosening of teeth clinically. Common site of this benign odontogenic tumor is posterior mandible affecting 2nd-5th decades of life. Aspiration shows negative but sometimes a straw-colored fluid may also be noted. Our case had a slow growing swelling with expansion of cortical plates leading to asymmetry of the face. The above clinical findings had a positive correlation with features of ameloblastoma and hence it was considered as one of our differential diagnosis. However histopathology was suggestive of orthokeratinised odontogenic cyst [8].

OKC is an aggressive lesion affecting individuals at any age group with mandible (60%-80%) being the most common site of occurrence. Clinically it presents as a painless slow growing swelling that gradually increases in size showing an anteroposterior growth pattern. As the swelling further grows in size, it starts showing a buccolingual growth pattern which then becomes clinically evident owing to gross facial asymmetry. Radiograph shows unilocular or multilocular radiolucency with cortical expansion extending till the inferior border of mandible. The above clinical and radiographic findings had a direct correlation with our current case [9].

Review of literature showed that 93% of orthokeratinised odontogenic cyst are associated with unerupted tooth and radiographically shows unilocular radiolucency. It is believed to be originating from remnants of dental lamina but the pathogenesis of orthokeratinised odontogenic cyst remains unclear. Orthokeratinised odontogenic cyst shows negative expression of BCL2 and low expression of Ki67 and P53 when compared with odontogenic keratocyst. They are also well organized as a cyst and less proliferative cyst when compared with odontogenic keratocyst [10]. Considering the recurrence rates and aggressiveness of cystic lesion, orthokeratinised odontogenic cyst had low recurrence rates (2%) on comparing it with Odontogenic keratocyst (43%) [11,12].

CONCLUSION

Diagnosing a cystic lesion remains challenging and chaotic for both the clinicians and pathologists. Unusual presentation of cyst such as orthokeratinised odontogenic cyst, a unique entity must be correlated with varying radiolucent lesions of jaw associated with impacted tooth to arrive at a confirmatory diagnosis.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

DECLARATION OF PATIENT CONSENT

The authors certify that they have obtained written informed patient consent for the surgery under general anesthesia and has given his consent for images and other clinical information to be reported in the journal with an understanding that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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