

Odontogenic Keratocyst mimicking Dentigerous Cyst - A Case Report

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Citation of this Article: Dr. Nandesh Shetty, Dr. Aishwarya Jayachandra, Dr. Jeff K. Zacharia, Dr. Tejkiran Shetty, “Odontogenic Keratocyst mimicking Dentigerous Cyst- A Case Report”, IJDSIR- February - 2021, Vol. – 4, Issue - 1, P. No. 561 – 567.

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Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

The odontogenic keratocyst (OKC) is considered as a distinctive lesion because of its varied origin, distinctive histology, proliferation characteristics, and its propensity to recur^{1,2}. They are odontogenic benign lesions within the bone, most often found in the mandible. Remarkable parallelism exists between odontogenic keratocyst and dentigerous cyst concerning mean age of occurrence, site predilection and radiographic features. This paper reports a case of a 21-year-old male patient with OKC occurring in the ramus of the mandible associated with an impacted mandibular third molar. The lesion, which was initially

diagnosed as a dentigerous cyst, was later confirmed to be an odontogenic keratocyst on histopathological examination. The initial diagnosis was made based on the clinical and radiographic features with differential diagnosis of OKC and Ameloblastoma. The clinical and radiological presentation of this cyst along with its histopathological features and management have been discussed.

Keywords: Odontogenic keratocyst, odontogenic cysts, enucleation, chemical cauterisation, case report

Introduction

The odontogenic keratocyst (OKC), is one of the most controversial pathologies in maxillofacial pathology. It

is a cyst derived from the remnants (rests) of the dental lamina, showing a typical lining of six to ten cells in thickness, it exhibits a basal cell layer of palisaded cells and a surface of corrugated parakeratin. It is also known to exhibit a biological behaviour similar to a benign neoplasm¹. These cysts are known to behave aggressively, show a high rate of recurrence and mimic other odontogenic cysts and tumours in clinical and radiographic presentation making histopathological examination vital for confirmatory diagnosis^{1,2}. Recurrence occurs in cases of incomplete removal of cyst lining and its contents, persistent epithelial islands after enucleation, a high mitotic index of the epithelial cells, a large sized cyst and also when the lesion is located in a site with difficult surgical access^{1,3}.

Case History

A 21-year-old male presented to the Department of Oral and maxillofacial surgery, with complaints of pain and swelling in the lower right back tooth region in the last three weeks. The patient was normal 3 weeks before, after which he gradually developed pain associated with a swelling over the right lower third of the face. The pain was mild to moderate in intensity, continuous in nature, radiating to the head and ear and was relieved on taking analgesics. Patient had no complaints of paraesthesia.

The patient had no systemic illness and was non-syndromic. On extra oral examination, the patient presented with a diffuse swelling over the right side of the face involving the ramus region, measuring about 4 x 4 cm, extending superiorly to the ala tragal line and inferiorly up to the lower border of the mandible [Figure 1]. The skin over the swelling appeared normal. On palpation, the swelling was firm in consistency and tender with a local rise in temperature.

Right submandibular lymph nodes were tender on palpation.



Figure 1 : Extraoral view showing gross facial asymmetry (swelling noted over the right side of the face)

Intraoral examination showed reduced mouth opening; region posterior to the lower right first molar was tender on palpation with pus discharge noted.



Figure 2: Intraoral picture of the patient showing purulent discharge Posterior to the second molar region.

Aspiration yielded a yellowish straw-coloured fluid mixed with blood and had a foul smell, which was consistent with the diagnosis of an infected cystic lesion. Routine laboratory parameters were normal.

The patient was subjected to radiological examination, by conducting an orthopantomograph and cone-beam computed tomogram. The investigation showed an impacted mandibular third molar placed horizontally in the middle of the right mandibular ramus, with a significant envelope of a radiolucency attached to the cervicoenamel junction of the impacted tooth, extending to the mesial of the mandibular right first molar. It was surrounded by a corticated border, having an intact alveolar border. The mandibular canal was pushed downwards on the right side [Figure 3], [Figure 4]. The radiologist gave a provisional diagnosis of dentigerous cyst since the radiolucency was attached to the coronal head of the impacted tooth.

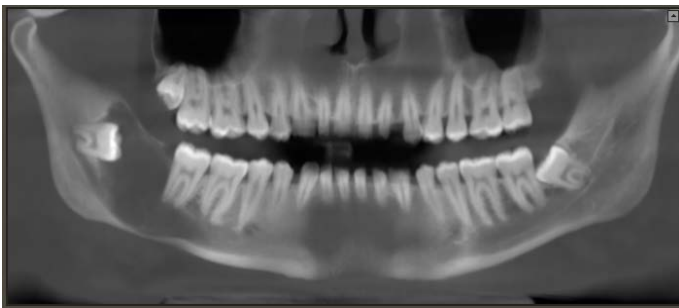


Figure 3: Preoperative Orthopantomograph showing extent of lesion involving impacted right mandibular third molar.



Figure 4 : [A and B] 3-D reconstruction cuts showing the lesion involving

The impacted right mandibular third molar.

Informed consent was obtained, the patient was posted for cyst enucleation under general anaesthesia. Markings were made over the right submandibular region. Local infiltration (lignocaine + adrenaline) was given, incisions were placed, dissection was done to expose the mandibular ramus. Facial artery and vein were identified and ligated. A bone window of about 5x5mm was made over the mandibular ramus, the friable cyst lining was identified, the cyst, which was seen involving the mandibular canal was enucleated along with the impacted mandibular molar [Figure 5].



Figure 5: Intraoperatively, a bone window created over the mandibular.

Ramus to localize the cyst.

The contents appeared creamy with cheesy consistency; Following enucleation and thorough debridement, the specimen was sent for histopathological examination. Peripheral osteotomy was done to achieve clean bone margins. Also, as a precautionary measure, Carnoy's solution (100% ethanol, chloroform and glacial acetic acid in a 6:3:1 ratio with added ferric chloride) was placed for chemical cauterization. This was followed by reinforcing the mandible with a 2.5mm, 11-hole reconstruction plate at the lower border of the mandible to prevent inadvertent

fracture [Figure 6]. Haemostasis was achieved and closure was done in layers.



Figure 6: Intraoperatively, chemical cauterization and peripheral ostectomy was done after enucleation and lower border of the mandible was reinforced with 2.5mm,11-hole stainless steel reconstruction plate

Histopathological examination reported stratified squamous epithelium, parakeratinized and thickness varying from five to ten layers. Basal layers were tall columnar. The lumen of the lesion was comprised of keratin material. Underlying connective tissue showed collagen fibers arranged parallel to the epithelium. A dense amount of inflammatory cells and cholesterol clefts were evident. The above features were suggestive of an Odontogenic Keratocyst. [Figure 7], [Figure 8]

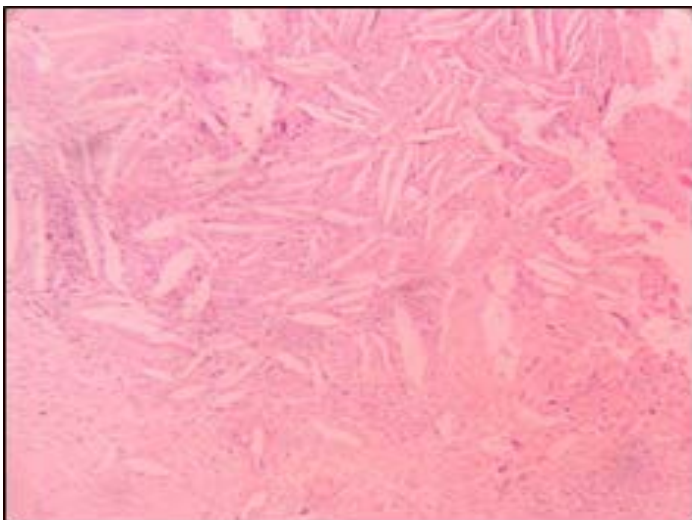


Figure 7: H and E-stained section of the lesion showing

Cholesterol clefts.

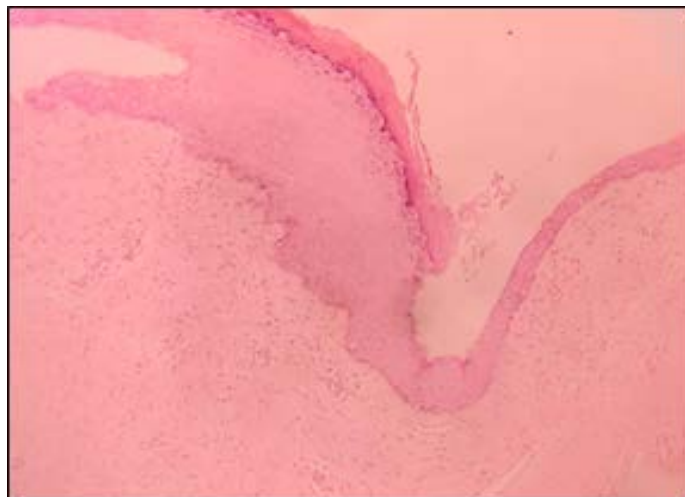


Figure 8: H and E section showing the lining epithelium which is corrugated with palisading basal layer and loosely bound to the cystic wall. Cystic lumen filled with Keratin (10 X magnification)

Healing was uneventful. However, patient had complaints of paresthesia over the right lower lip region during his follow up. Immediate Post op and late post op photographs were taken [Figure 9 and 10] along with a post-operative orthopantomograph was taken [Figure 11] .



Figure 9: Postoperative Orthopantomograph

Discussion

The Odontogenic Keratocyst is one among the commonly occurring odontogenic cysts of the jaws. The term ‘Odontogenic Keratocyst’, introduced by Philipsen in 1956, was renamed as Keratocystic Odontogenic Tumour (KCOT) by the WHO (2005).

However, in 2017, the WHO reinstated the original terminology of OKC as the pathology was again considered as an odontogenic developmental cyst⁴. Hence, we chose to use the term OKC to describe the cyst in this case report.

The OKC represents between 12% to 14% of all odontogenic cysts and is hypothesized to arise from the rests or remnants of the dental lamina exhibiting biological features similar to a benign neoplasm according to Toller in 1976. Recent studies also suggest the role of the PTCH (patched) gene in both syndrome-associated and sporadic OKCs⁵.

According to a case series published by Brannon, which is the largest and representatives of the lesion, the incidence of OKC had a varied age range, with incidence peaking in the second and third decade of life, and a predilection for occurrence in males⁶. The mandible is almost always affected more frequently than the maxilla in over 65% of the cases. The ramus-third molar region, followed by the first and the second molar area are the most commonly involved sites within the mandible. They invariably occur within the bone, although few cases of peripheral OKCs have also been reported⁷. The case described is that of a male patient aged 21 years, presenting with a cyst occurring in the mandibular ramus.

Patients may present with common clinical features including pain, soft tissue swelling, drainage and various neurological manifestations like paraesthesia of the lip or teeth. However, the signs and symptoms seldom develop until the cyst has grown to a large extent. This is owing to the distinctive ability of OKC to grow in the anteroposterior direction without causing a remarkable swelling that can be diagnosed at an earlier phase. A bony swelling is caused by periosteal bone formation, however OKCs do not give to such a

swelling because they tend to follow the path of minimum resistance, thereby occupying the medullary cavity⁸. Thus, most OKCs are discovered incidentally during routine radiographic examination. The present case showed a cyst extending from the mesial of the mandibular right first molar to the middle of the mandibular ramus on the ipsilateral side, with symptoms occurring only 3 months before the patient sort consultation.

Literature shows that 27% of the OKC cases are associated with an impacted tooth, the mandibular third molar being the most involved⁹. The patient in this report presented with an impacted 46.

Although OKCs can occur as solitary lesions, most cases are associated with multiple lesions, especially in patients with Nevoid Basal Cell Carcinoma Syndrome (NBCCS) or Gorlin-Goltz syndrome. In addition to multiple OKCs, NBCCS is characterized by multiple basal cell carcinomas, palmar or plantar pitting, bilamellar calcification of the falx cerebri, bifid ribs, frontal bossing, epidermoid cysts and medulloblastoma¹⁰. Since the patient did not fulfil the major or minor criteria, NBCCS was ruled out.

On radiographic examination, most OKCs are unilocular, with a few exceptions of mulilocular lesions. They present a well-defined peripheral rim. Scalloping of the border is a common finding, representing variations in the growth pattern of the cyst¹. OKCs are often mistaken for dentigerous cysts when they contain the crown of an impacted tooth within its lumen. Other differentials include Ameloblastoma, Adenomatoid odontogenic tumour and bone cyst. Initially, the present case was also misdiagnosed as a dentigerous cyst which was later diagnosed as OKC after histopathological confirmation.

OKCs show unambiguous features on histopathological examination. These include a uniform cyst lining with

thickness of six to ten cells and a basal layer of hyperchromatic palisaded cells. The surface of the cyst shows wavy, corrugated parakeratin production, features of which were similar in the present case. However, these classic histopathological features of the cyst may often be obscured due to inflammation and infection, posing diagnostic difficulties which can lead to an incorrect diagnosis¹¹.

Unlike other odontogenic cysts, OKCs demonstrate a high rate of recurrence between 25% and 60%. In 1976, Brannon proposed 3 mechanisms for KCOT recurrence: inadequate or incomplete removal of the cyst lining, satellite cysts giving rise to new lesions and development of a new OKCs in the area adjacent to parent lesion that is interpreted as a recurrence⁷. Due to this high recurrence, treatment of OKCs continues to be a challenge to a clinician. The present case was followed up for 9 months; however, a follow up period of at least 5 years is required owing to the recurrence rate.

Morgan and colleagues categorize surgical treatment methods for OKC as either conservative or aggressive. Conservative treatment generally includes simple enucleation, with or without curettage, or marsupialization¹². Aggressive treatment on the other hand includes enucleation along with peripheral ostectomy, chemical curettage using Carnoy's solution, and resection. Aggressive modalities are recommended for cases associated with Nevoid Basal Cell Carcinoma Syndrome, large cysts and recurrent lesions. In the present case, the patient underwent enucleation, chemical cauterisation with Carnoy's solution, followed by peripheral ostectomy. Literature shows that treatment of an Odontogenic keratocyst with peripheral ostectomy, with or without the use of Carnoy's solution, had a significantly lower rate of recurrence (3.8%)^{5,13}.

Conclusion

Significant parallelism exists between odontogenic cysts associated with an impacted tooth due to similar clinical and radiographic features. However, it is of utmost importance to differentiate OKCs from the others due to its aggressive behaviour and high rate of recurrence, which would require a more aggressive treatment modality. Therefore, histopathological examination combined with the clinical and radiographic examination is a key to accurate diagnosis and appropriate management.

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