The Silent Cyst: A Case Report

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Abstract

The Odontogenic keratocyst (OKC) is a cystic lesion that is a non – inflammatory developmental and a chronic cystic lesion which may be seen as a unilocular or multi locular lesion on a radiograph. OKC is usually asymptomatic and has an odontogenic origin. It shows an aggressive clinical behaviour and is known to have a high recurrence rate and has a tendency to invade adjacent soft tissues and bone. The same may be undiagnosed and only seen incidentally on a radiograph. Diagnosis is based on clinical history, clinical appearance, radiographs and histology. This case of OKC of the mandible was incidentally discovered on a radiograph involving the ramus of the mandible and is presented in this article.

Keywords: Odontogenic Keratocyst; Asymptomatic cyst; impacted third molar; 5 flurouracil **Article text**

Introduction

Phillipsen first described OKC in 1956.OKC is a cyst of singular odontogenic origin. This can be potentially destructive and has a high recurrence rate. OKC comprises of about 11% of all odontogenic cysts.^[1] It is the third most common cyst of the jaws and has a slight male

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predilection.^[2,3] As per literature OKC is known to originate from the dental lamina¹, but can also originate from the cells of the overlying epithelium.^[7] Several lesions can mimic an OKC hence it is necessary to establish a differential diagnosis which can include central giant cell granuloma, odontogenic myxoma, ameloblastoma, traumatic bone cyst, residual cyst, dentigerous cyst if the OKC is associated with an impacted tooth. A histopathological examination is required to establish a definitive diagnosis OKC tends to grow in an anteroposterior direction within the medullary cavity of the bone without causing obvious bone expansion. ^[4-6]

This case report is of an odontogenic Keratocyst in a 22 year old patient which involved the left posterior mandible.

Patient information

A 22 year old male patient was referred to the OPD due to an incidental finding on the panoramic radiograph in the lower left posterior region revealing an osteolytic lesion. The radiograph was taken due to a missing third molar in lower left posterior region. No associated history of swelling, fever, or pus discharge. The patient could not recollect any past history of trauma.

Past dental history revealed a gradual onset and gingival inflammation on the distal aspect of 37 due to trauma from the opposing tooth. The pain was occasional for one year for which he was treated by a private dental practitioner and was prescribed antibiotics and analgesics post which the gingival swelling would subside. Subsequently the patient was advised panoramic imaging. His family,medical and social history was non-contributory.

Clinical findings

In the extra oral examination the face appeared bilaterally symmetrical. No surface abnormalities were noted. No neurosensory deficit was elicited and the lymph nodes were non palpable bilaterally.

On intraoral examination the 38 and 48 were partially impacted. The buccal and lingual vestibule in 36 and 37 region showed no obliteration and the mucosa appeared intact. [figure 1 (a) and (b)]





Image legends

Figure 1 (a)pre operative intraoral image showing missing 38

figure 1 (b) missing 38 and inflamed gingiva distal to 37

On palpation, there was no pain or tenderness or expansion of the buccal and ingual cortical plates. No associated mobility of the adjacent teeth.

The panoramic image revealed a well-defined multilocular radiolucency with well-defined corticated scalloped borders measuring approximately 5 cm \times 2.5 cm in its largest dimensions extending from the cervical $1/3^{rd}$ of the impacted 38 up to 1cm inferior to the coronoid notch supero inferiorly and mesiodistally the radiolucency extends from the medial border of the ramus to 0.5cm medial to the lateral border of the ramus of the mandible. Antero-posteriorly seen extending from the mesial aspect of 36 to 1 cm below the sigmoid notch. Inferior alveolar nerve was downwardly displaced. Another round well defined radiolucency is noted in the periapical area of 36 and 37 which appears to be in continuation with the former radiolucency. The internal structure appeared to be uniformly radiolucent. [figure 2]



Image legends

Figure 2: Panoramic radiography showing extensive radiolucent lesion.

A CBVI (Cone Beam CT) was taken for the area of 36 37 38 and the left ramus of the mandible area. The CBVI revealed a hypodensity extending anteriorly from the distal root aspect of 36 to the ascending ramus of the mandible posteriorly. The epicentre is located superior to the IANC. The lesion is well defined and has a corticated border and a scalloped outline. Homogenous area of hypodensity is seen throughout the lesion along with the impacted 38.

Buccal and lingual cortical plate thinning noted. The inferior alveolar nerve canal appeared intact. [figure 3 (a), (b), (c)]

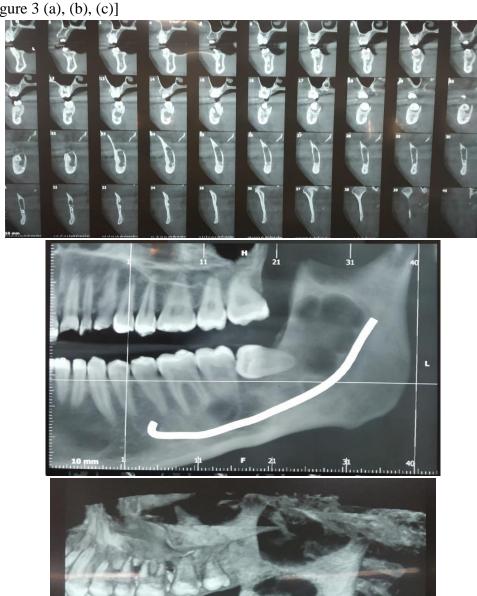


Image legends

Figure 3 (a) large hypodense lesion extending from 36 into the ramus of the mandible and has scalloped margins and an intact nerve canal

figure 3(b) axial section showing no evidence of cortical bone expansion

Figure 3 (c) sagittal sections

Diagnostic assessment

Radiographic features were suggestive of Odontogenic cyst/ tumor. A differential diagnosis of dentigerous cyst, odontogenic Keratocyst and ameloblastoma were considered. Further an incisional biopsy was performed.

The patients routine hematological investigations and blood sugar evaluation were within normal limits.

Therapeutic Intervention

The patient underwent extraction of 38 followed by cystic enucleation and packing of the surgical cavity with placement of a sterile ribbon gauze coated with an antimetabolite agent 5 flurouracil. Closure was done and a small end was left exposed into the oral cavity. [figure 4]

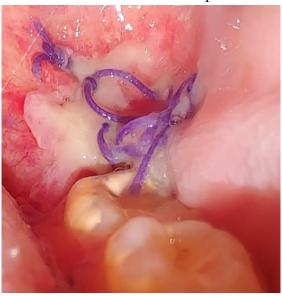


Image legend

Figure 4: Post operative and treatment with 5 fluorouracil

Diagnosis was confirmed as odontogenic Keratocyst after histopathologic evaluation.

Follow up and outcomes

The patient is under follow up since 2 years and is asymptomatic with healing taken place.

Discussion

This case report presents the parakeratotic type which is the most frequent type(80%). This type has a more aggressive clinical presentation compared to the orthokeratinized variant. The luminal content can vary in consistencies from "straw-colored fluid," or "thick pus-like" material to a caseous, thick, cheesy, milk white mass. OKCs mostly grow intraosseously and in a longitudinal direction hence causing minimal expansion, as seen in this case. Due to rapid growth the bone marrow is replaced and there is not enough time for the periosteum to lay down new bone.^[7]

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OKCs may present radiographically with the following features which may help distinguish it from other osteolytic lesions which are as follows 1) Border is often Well corticated and scalloped; 2) longitudinal and medial expansion; 3) resorption, extrusion or displacement of teeth; and 4) internal structure is mostly a radiolucent occasionally can appear cloudy or milky. [8]

Resection is the most aggressive treatment of OKC but is not accepted as a routine treatment as it is aggressive and is known to have high morbidity and hence may be considered in only severe cases. Studies show that recurrence can be avoided with resection involving surrounding 5 mm margin of healthy bone.^[9]

In the present case considering the patients age and the extent of the lesion an enucleation was performed with adjunctive therapy of 5- fluorouracil (5-FU, antimetabolite). It was used in place of the traditional Carnoy's solution due to the presence of a carcinogenic component(chloroform). The modified Carnoy's solution has shown high chances of recurrence. Other adjunctive therapies used are Cryotherapy, osteotomy. 5-Fluorouracil (5-FU) has been used in the treatment of hepatocellular cancer, and different malignant diseases, like Basal Cell Carcinoma, for topical application. It acts by inducing aptosis by inhibiting Sonic Hedgehog (SHH). Leddehroff suggested a targeted approach in the treatment of OKC based on its molecular genetics. [10]

Patient perspective

The patient had been suffering with lesion for last one year and was undiagnosed. But when the same was diagnosed clinically and radiographically the present treatment given was effective and the cyst healed completely. There was no discomfort with any medication or treatment that was prescribed. The treatment considered was highly conservative and retained aesthetics and hence a very major and aggressive surgery was avoided.

Conclusion

Developing an appropriate treatment modality for OKCs is still a matter of research because of its genetic and molecular pathogenesis. No consensus exists on a uniform treatment plan and surgical managements vary from enucleation to marsupialization to en bloc resection. Treatment plan depends on several factors like the extent of lesion, age etc. Due to high recurrence rates all cases even after surgery require a clinical and radiographic follow-up for years.

Declaration of patient consent

A written informed consent was obtained from the patient for publication of the case report and the accompanying images

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figure 3(b) axial section showing no evidence of cortical bone expansion

Figure 3 (c) sagittal sections

Figure 3 (d) Computed tomography showing the presence of a large hypodensity with scalloped borders and no evidence of cortical bone expansion.

Figure 4: Post operative and treatment with 5 fluorouracil