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ABSTRACT

Odontogenic keratocyst (OKC), previously known as keratocystic odontogenic tumour (KCOT), is a benign cystic lesion originated from dental lamina involving the mandible or maxilla. It is locally aggressive and has high recurrence rate due to its infiltrative behavior. OKC can occur in a wide age range but peak at second to third decade of life. It is most commonly seen at the angle of mandible. We report an unusual presentation of OKC at anterior maxilla in a 67-year old gentleman who had history of trauma to his maxillary anterior teeth, presented with non-vital tooth 11 and a well-defined radiolucent lesion. Following completion of root canal treatment for tooth 11, enucleation and peripheral ostectomy were performed with intra-operative use of Carnoy’s solution. Histopathological examination of the specimen confirmed it as OKC. Currently, the patient was put under strict 6 monthly periodical follow-ups. Clinicians should be aware of such atypical presentation of OKC to avoid misdiagnosis.
INTRODUCTION

Odontogenic keratocyst (OKC), a term first used by Philipsen in 1956, is a benign developmental cyst which originate from residual dental lamina within the alveolar bone. Despite being a benign lesion, it is characterized by its locally aggressive behavior and high recurrence rate. These are attributed to its infiltrative behavior. It is because of these characters that this lesion was classified as a benign neoplasm in 2005 giving its name keratocystic odontogenic tumour (KCOT). In the recent review in 2017, the World Health Organization (WHO) has reclassified it as a cyst due to lack of evidence to support its neoplastic nature, thus reverting to its earlier name of odontogenic keratocyst. Other seldom used names for this lesion are primordial cyst and odontogenic keratocystoma. OKC is mostly presented at the 2nd and 3rd decades and predominantly affect male. It affects mandible more than maxilla and is frequently found at the angle of mandible. It can be solitary or multiple. Multiple OKCs in the jawbone is one of the features of naevoid basal cell carcinoma syndrome. We report a rare case of OKC in a 67 years old man involving anterior maxilla which mimicked a radicular cyst.

CASE REPORT

A 67-year old male presented with a swelling at the labial aspect of maxillary central incisors. He first noticed the swelling about 2 months ago when there was purulent discharge. The purulent discharge resolved following a course of antibiotic given by his dentist. However, the swelling persisted albeit painless. He had a fall when he was young which caused displacement of his upper front teeth. Otherwise, his medical history was unremarkable. Upon extra oral examination, the face was
symmetry and no obvious swelling seen over the face. Intraorally, there was a swelling at the upper labial sulcus of tooth 11 to 22 measuring 2cm x 2cm with 11 displaced labially, overlying mucosa was normal and there was no palatal expansion. Periodontal pocket charting was insignificant. There was no discoloration of the maxillary incisors. His maxillary incisors were asymptomatic except for slight tender to percussion on tooth 11. Upon palpation, the swelling was soft, non-tender with no discharge. Electrical pulp sensibility test revealed negative response from tooth 11 only. An upper occlusal radiograph was taken and showed a well-defined unilocular radiolucency extending from tooth 12 to 22 with no root resorption. Based on clinical and radiological findings, a provisional diagnosis of radicular cyst associated with non-vital 11 tooth was given. After completion of root canal treatment for tooth 11, enucleation of the cyst with apicectomy and retrograde filling were performed under general anesthesia. During the surgery, we noticed thick cheesy content extruding from the cyst. Suspecting it could be OKC, we performed peripheral ostectomy and Carnoy’s solution was applied over the bone defect. Histopathology examination revealed the cystic lining was composed of parakeratinised stratified squamous epithelium with palisaded basal layer. The epithelial lining exhibited corrugated appearance and flat interface of the epithelial connective tissue while the underlying connective tissue was moderately collagenous and inflamed. A dense infiltration of predominantly lymphocytes and plasma cell was seen. A final diagnosis of OKC was given. Healing following surgery was uneventful. Patient was put under regular 6 monthly follow up.

**DISCUSSION**

OKC is generally thought to be derived from either the epithelial remnants of the dental lamina or the basal cell layer of the surface epithelium. The latter may account for the development OKC at the angle of mandible as the area posterior to
the last molar is unlikely to have dental lamina\textsuperscript{2,3}. Role of Patched (PTCH) gene mutations in etiopathogenesis of OKCs have been reported in both syndromic and non-syndromic OKCs, with higher incidence documented in the former\textsuperscript{1,4}. It can occur at any age but is predominant in the second and third decades of life with male predilection. OKC is most often found at the angle of the mandible. In the maxilla, it is commonly reported at the canine area. In the present case, the age at presentation and location of the OKC were unusual. OKC is usually asymptomatic when the lesion is small. However, as the cyst enlarges, it may present with swelling, pain and purulent discharge if infected as seen in this case. Resorption of the roots of adjacent teeth and bone expansion is often not observed in OKC, as it tends to grow in an anterior posterior direction within the medullary cavity of the bone\textsuperscript{5}. Various theories were put forward to explain the expansion of OKC including intraluminal hyperosmolality, active epithelial proliferation, collagenolytic property of the cyst wall and synthesis of interleukin 1 and 6 by keratinocytes\textsuperscript{6}.

Being a slow growing cyst, OKC presents as a radiolucency lesion with well-defined border. Diagnosis through radiographs is difficult as its radiographic features are non-specific. However, presence of multilocular radiolucency may highly indicate OKC\textsuperscript{7}. Being originated from dental lamina, OKC can present at lateral periodontal or lateral follicular region. This can lead to misdiagnosis as lateral periodontal cyst, radicular cyst, residual cyst and dentigerous cyst\textsuperscript{8}. In this case, the provisional diagnosis given was radicular cyst based on the history of trauma, clinically non-vital\textsuperscript{11} and radiographic presentation of a well-defined radiolucency. Definitive diagnosis relies on histopathological findings. Under the microscope, OKC presents with a thin capsule of fibrous connective tissue, lacking inflammatory infiltrate and covered by parakeratinized stratified epithelium with superficial corrugation and a basal layer of hyperchromatic cells in palisade\textsuperscript{8}. 
Treatments of OKC remain controversial due to its possibility of recurrence. Managements of OKC can be generally divided into conservative such as marsupialization or decompression and radical such as enucleation and resection. While resection, may it be marginal or segmental, has the lowest rate of recurrence, it shall be kept as last option. This is because of the benign nature of the cyst and its resulting morbidities such as facial asymmetry following resection. Johnson et al also concluded that simple enucleation carries high rate of recurrence of 30%. Marsupialization alone contributes to a recurrence rate of 18%. However, enucleation can be performed for small cyst provided that adjunctive measure such as Carnoy’s solution is used. This treatment regime greatly reduced recurrence rate to about 8%. Carnoy’s solution is a chemical cauterization agent with moderate tissue penetration, rapid local fixation and hemostatic action. It consists of 60% ethanol, 30% chloroform, 10% glacial acetic acid with 0.1g ferric chloride. It has been shown to eradicate epithelial rest from the cyst wall with the average depth of about 1.5mm of penetration after 3 minutes of application. For larger lesion, it is recommended to perform marsupialization followed by enucleation with adjunctive measure to reduce surgical injury. Combination of peripheral ostectomy had been reported to be successful. In our case, we performed enucleation combined with peripheral ostectomy and application of Carnoy’s solution. As OKC is notorious for its recurrence, patient must be followed up periodically with radiographic examination. Pogrel recommended follow-up, with radiographs, every 6 months for 2 years, every year for 5 years, and every 2 years for 10 years in asymptomatic patients.
CONCLUSION

This case showed a rare presentation of OKC at anterior region of maxilla, which mimic a radicular cyst. Provisional diagnosis was misled by the history of trauma, non-vital tooth and radiographic presentation. Aspiration of the cyst content should be performed as it may provide clue to the diagnosis. This was not performed in our case and we deeply regretted. Another lesson learnt from this case was to be versatile while performing surgery and always be ready to alter surgical plan in case of unexpected intra-operative findings.

Figure 1. Intraoral swelling obliterated the labial sulcus from 12 to 22 area with 11 displaced labially
Figure 2. Upper occlusal radiograph shows unilocular well-defined radiolucency

Figure 3. Photomicrograph showing cystic lining composed of parakeratinised stratified squamous epithelium with palisaded basal layer
Conflict of interest

The authors declare that there are no conflicts of interest regarding the publication of this article.

Consent

Informed consent had been obtained from the patient.
REFERENCES

