Case Report

Is odontogenic keratocyst an endodontic enigma? A rare case report of management of odontogenic keratocyst in anterior mandible

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Abstract

Odontogenic keratocyst (OKC), a rare, locally aggressive developmental cyst, is found incidentally on dental radiographs, most commonly in the posterior mandible. When it occurs in the periapical region in association with nonresponding teeth to pulp sensibility tests, it is often misdiagnosed as other endodontic lesions such as radicular cyst, lateral periodontal cyst and dentigerous cyst. This case report describes the rare occurrence of OKC in the anterior mandibular region managed successfully with endodontic treatment and re-interventional surgery in a conservative approach. This case report also emphasizes on the importance of histopathological examination of the surgically excised tissue specimens as the clinical, radiographic and histopathological correlations are essential for successful treatment.

Keywords: Biopsy, Carnoy's solution, endodontic lesion, odontogenic keratocyst, periapical surgery

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INTRODUCTION

Periapical lesion with significant destruction of alveolar bone and lack of a response to tooth sensibility test indicates the need for endodontic therapy. The most common differential diagnosis of such endodontic lesions would be radicular cyst, lateral periodontal cyst and periapical granuloma. Odontogenic keratocyst (OKC) is often misdiagnosed as other developmental cysts. It is a rare and benign but locally aggressive developmental cyst[1] and make up around 19% of jaw cysts.[2]

In the WHO/IARC classification of head-and-neck pathology, this clinical entity which had been known for years as the OKC was reclassified as keratocystic odontogenic tumor (KCOT) from 2005 to 2017.[3,4] In 2017, it reverted to the earlier name, as the new WHO/IARC classification reclassified OKC back into the cystic category.[5] The peak incidence of OKC is seen during the second and third decades of life. At least 50% of OKCs are found in the posterior part and the lower ramus of the mandible.[6] However, most of the times, OKCs may be asymptomatic and found incidentally on dental radiographs.[7] Rarely, if symptoms can arise, they are due to infection or expansion of the bone.[2,6]

This is a rare case report describing the occurrence of OKC in the anterior mandibular region, which was

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managed successfully with endodontic treatment and re-interventional surgery in a conservative approach.

CASE REPORT

A 29-year-old male patient reported to the department of conservative dentistry and endodontics with a chief complaint of sensitivity in the lower front teeth for 8 months. Here, the severity of sensitivity increased on occluding teeth. Few months later, he noticed swelling in the lower front jaw, which had slight tenderness on touch, not associated with fever or any discharge.

Extraoral examination revealed no gross asymmetry, discharge or sinus tract. On palpation of the lower chin, no tenderness was present.

Intraoral examination [Figure 1a] revealed tenderness on percussion with respect to tooth nos. 32, 31 and 41. A solitary diffuse swelling was observed with respect to the lower anterior region, with vestibular obliteration extending mediolaterally from 33 to 42 regions. The overlying mucosa was inflamed as compared to the surrounding mucosa with no sinus tract observed. The mouth opening was approximately 35 mm. To palpate, the swelling was soft in consistency and compressible due to labial cortical perforation. Cold test and electric pulp test were performed. Negative response was obtained on testing with respect to 31, 32, 33 and 41. Radiovisiograph revealed a well-corticated, multiple coalescent radiolucency with respect to the apical regions of 33, 32, 31, 41 and 42 [Figure 1b]. A provisional diagnosis of radicular cyst associated with 33, 32, 31 and 41 was made. Treatment protocol planned was endodontic treatment followed by surgery.

Endodontic therapy phase

Access opening was done for 33, 32, 31 and 41. On negotiating two canals, one buccal and one lingual canals were located for all teeth.

Working length was established, and cleaning and shaping was done up to ISO 30/0.02 size.

For disinfection, calcium hydroxide medicament was placed for 4 weeks [Figure 1c]. As there were no signs of healing of the periapical lesion noticed, periapical surgery was planned after obturation of 33, 32, 31 and 41 [Figure 1d].

Periapical surgery phase

Enucleation of cyst with respect to 33, 32, 31 and 41 was done under local anesthesia. A full-thickness mucoperiosteal flap was elevated from the regions of 34–43 [Figure 1e]. Cystic lining was separated from the bone and dissected from the underlying mucosa and removed in toto [Figure 1f and g]. Apicectomy and retrograde filling was done with glass ionomer cement with respect to 33, 32, 31 and 41. Sharp bony margins were reduced, and the socket was debrided with betadine and saline solution. 4-0 vicryl sutures were placed to close the surgical site. Pressure pack and postoperative instructions were given. The patient was recalled after 1 day for follow-up and after 7 days for suture removal.

Biopsy phase

The cyst enucleated in toto was sent to the department of oral pathology for histological examination [Figure 1h]. The histopathological report revealed that the section showed lining of corrugated parakeratinized stratified squamous epithelium with basal cell layer, showing typical tombstone appearance. The cystic lumen showed keratin flakes. The epithelium was lifted away from the underlying connective tissue capsule at few areas. The connective tissue showed dense inflammatory infiltrate with areas of vascularity.

An impression of OKC was made by the histopathological report [Figure 1i].

Revisit of surgical site

Considering the aggressive nature and high chance of recurrence of OKC, a decision to revisit the surgical site was taken. Vestibular incision was given from the distal aspect of 33 to the distal aspect of 42 region after administration of local anesthesia. A full-thickness mucoperiosteal flap was elevated. Curettage of granulation tissue was done within the cystic cavity followed by peripheral ostectomy and Carnoy's solution application. Socket toileting was done with betadine-saline solution. 3-0 interrupted silk sutures were used to close the surgical site. Pressure pack and postoperative instructions were given. The patient was recalled after 1 day for follow-up and after 7 days for suture removal.

Suture removal was done after 7 days.

The patient was recalled after 1 month, 3 months, 6 months and 1 year for follow-up. The radiographs revealed complete resolution of the periapical lesion, with the teeth being asymptomatic [Figure 2].

DISCUSSION

The term OKC was first used by Philipsen in 1956.[8] It is one of the most aggressive odontogenic cysts of the oral cavity. OKC is known for its rapid growth and its tendency to invade the adjacent tissues including bone.[9,10]

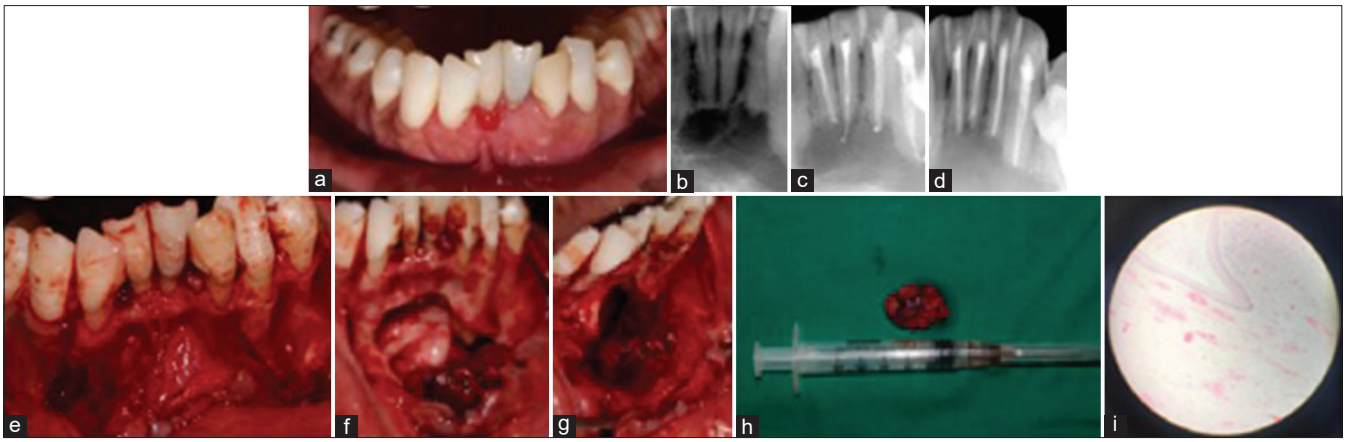

Figure 1: (a) Preoperative clinical intraoral view. (b) Preoperative radiovisiograph. (c) Calcium hydroxide intracanal medicament placed with relation to 33, 32, 31 and 41. (d) Obturation of 33, 32, 31 and 41. (e) Flap reflection. (f and g) Cyst isolated (h) cyst removed in toto and sent for histopathological examination. (i) H and E staining of the tissue sample showing characteristics of odontogenic keratocyst

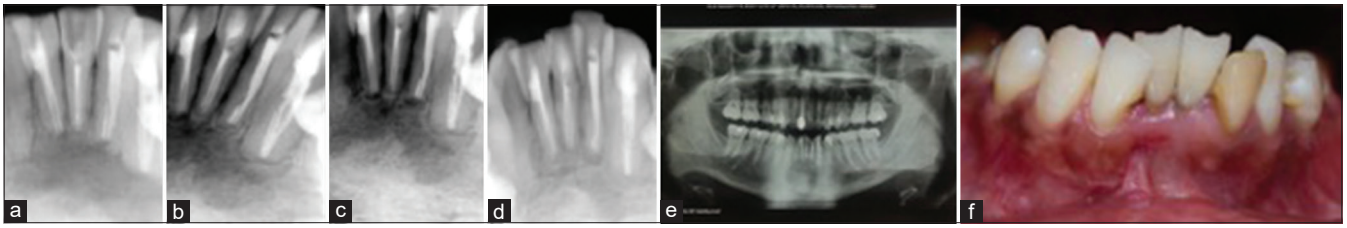

Figure 2: Follow-up at (a) 1 month; (b) 6 months; (c) 1 year and (d) 1 year, 6 months. (e) Orthopantomogram showing complete resolution of periapical lesion at 1-year follow-up. (f) 18-month follow-up clinical intraoral view

Histologically, OKCs arise from the dental lamina and are constituted by a cystic space containing desquamated keratin, lined with a uniform parakeratinized squamous epithelium of 5–10 cell layers, with a distinct basal layer of palisaded columnar or cuboidal cells, whose nuclei tend to be vertically oriented. The interface with the adjacent connective tissue is normally flat with a potential for budding of the basal layer and the formation of small satellite cysts.[11] The mitotic activity is higher than that of other cysts of odontogenic origin.[12]

The treatment method for OKCs remains controversial. There is no evidence in the literature that would guide surgeons in selecting the best treatment options. Clinicians continue to rely on their own experience when deciding for the most appropriate treatment. Surgical approaches vary between conservative and more aggressive treatments. Enucleation is the most commonly used treatment method and is associated with a high recurrence rate. Decompression and marsupialization has been used as a conservative treatment of OKCs. Some clinicians do not approve these techniques because the potential remnant cystic tissues left behind can facilitate the recurrence. A more aggressive approach may, therefore, lower the risk of recurrence. It has been suggested that aggressive resection should be limited to OKCs that have recurred more than twice or to those that have undergone malignant transformation. In addition, Worrall recommended radical excision as the treatment of choice for OKCs that had cortically perforated, whereas it is also been reported that enucleation plus curettage with Carnoy's solution results in a recurrence rate, statistically comparable to that of resection excision.[13]

In our case report, a 29-year-old male patient had sensitivity in the mandibular anteriors and swelling in the lower front jaw which was slight tender on touch. This was associated with negative response to pulp sensibility testing and intraoral periapical radiograph showing periapical radiolucency in relation to mandibular anterior teeth. Hence, a provisional diagnosis of radicular cyst was made, and endodontic therapy followed by periapical surgery with apicectomy and retrograde filling was planned. The cyst lining and contents were subjected to histopathological examination for obtaining the final diagnosis. The histologic examination revealed that the section showed lining of corrugated parakeratinized stratified squamous epithelium, with basal cell layer showing typical tombstone appearance. The cystic lumen showed keratin flakes. The epithelium was lifted away from the underlying connective tissue capsule at few areas. The connective tissue showed dense inflammatory infiltrate with areas of vascularity. Impression of OKC was made by the histopathological

report. Thus, the surgical site was revisited, and curettage of granulation tissue was done within the cystic cavity followed by peripheral ostectomy and Carnoy's solution application. Carnoy's solution fixative (ethanol, chloroform and acetic acid), which is usually used in conjunction with excision and curettage, intended to remove any residual epithelial cells to a depth of 1–2 mm.[13]

OKC, being an aggressive odontogenic cyst, has 2.5%–62.5% frequency of recurrence after surgical intervention.[14] Hence, the definitive diagnosis is important. This was achieved with the help of biopsy of the cystic lesion. The study revealed that the concordance between clinical and histopathological diagnoses of all lesions was 80.5%. It was reported that many of the diagnostic disagreements were in the developmental/inflammatory/reactive lesion group. KCOTs mimicking cystic lesions were reported by several studies published in recent years. Previous literature shows that in a study, 38 cases of KCOT were diagnosed as inflammatory cystic lesions or dentigerous cysts provisionally.[15] Hence, KCOTs should be included in the differential diagnoses of cystic lesions due to their aggressive behavior and recurrence in spite of complete removal.[16] The emphasis on identifying whether the lesion is a cyst or KCOT is important for surgical procedures. Radicular and dentigerous cysts can completely be cured with simple enucleation, while a simple enucleation of KCOT can have recurrence rate of up to 27.8%.[17]

In a systematic review of the literature, Johnson *et al*. showed that association of lesion's enucleation with adjunctive technique of chemical cauterization with Carnoy's solution, significantly reduced the recurrence rates to about 8%.[17] A similar surgical approach was followed in this present case report.

CONCLUSION

This is a rare case of OKC occurring in a 29-year-old male patient in the anterior mandible managed successfully with conservative approach, with special emphasis on the importance of histologic examination of the surgically excised specimen. The clinical, radiographic and histopathological correlations are essential for proper patient treatment and follow-up. This will avoid the further complications as OKCs are highly aggressive and have high recurrence rate.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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