International Surgery Journal
Dua P et al. Int Surg J. 2024 Sep;11(9):1555-1558
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902

DOI: https://dx.doi.org/10.18203/2349-2902.isj20242461

Case Report

Unmasking the enigma - recurrent odontogenic keratocyst: a continual challenge

Piyush Dua*, Shayan Ghosh, Sania Khalid, Muskan Grover, Rishabh Jaiswal, Amit Gupta

Department of Oral and Maxillofacial Surgery, I.T.S Centre for Dental Studies and Research, Muradnagar, Uttar Pradesh, India

Received: 03 July 2024
Accepted: 05 August 2024

***Correspondence:**
Dr. Piyush Dua,
E-mail: drduapiyush@gmail.com

ABSTRACT

Odontogenic keratocysts (OKCs) represent 10% to 14% of all jaw cysts and are notable for their high recurrence rate compared to other odontogenic cysts. This case study details a recurrent OKC in a young woman, highlighting the critical need for diligent long-term follow-up and effective management strategies. A 25-year-old female presented with a recurrent OKC in the maxilla. The treatment involved surgical enucleation of the cyst, apicectomy of affected teeth, and application of Carnoy's solution to reduce recurrence risk. Follow-up imaging and clinical evaluations were conducted periodically to monitor healing and detect potential recurrences. Postoperative recovery was uneventful, with no immediate complications. Serial radiographs and clinical examinations during the follow-up period showed satisfactory healing and no signs of recurrence. The patient reported no discomfort or functional impairment. This case underscores the importance of comprehensive surgical intervention and long-term follow-up in managing recurrent OKCs. The combined approach of cyst enucleation, apicectomy, and Carnoy's solution application proved effective in preventing recurrence in this patient. Ongoing monitoring remains essential to ensure early detection and management of potential recurrences, ultimately improving patient outcomes.

Keywords: Cyst, OKC, Keratocystic odontogenic tumor, Cryotherapy

INTRODUCTION

In 1956, Philipsen first introduced the term OKC.[1] The OKC is a frequently occurring cyst in the jaws, accounting for about 10% to 14% of all cystic lesions affecting the jaws.[2] An OKC is a cyst of developmental origin believed to arise from the remnants of the dental lamina.[3] It is widely acknowledged that OKCs exhibit a higher recurrence rate compared to other forms of odontogenic cysts.

Several etiological hypotheses have been proposed, including: incomplete enucleation of the primary cystic lining comprised of highly metabolically active epithelial cells, distinct from those found in other odontogenic cysts; proliferation of a secondary cyst from residual satellite cysts or odontogenic remnants following surgical intervention; or suspicion of recurrence based on the emergence of a new cyst in an adjacent maxillofacial region.[4] The recurrence rate of KCOT varies widely, ranging from 2.5% to as high as 62%.[5]

OKCs, or OKCs, commonly exhibit a predilection for the mandibular third molar region and ascending ramus. However, they can also appear in the dentate segments of both the maxilla and mandible, mimicking typical odontogenic cysts in appearance.[6]

This paper presents a case of a young woman diagnosed with recurrent OKC, a condition affecting the jaw, underscoring the importance of long-term patient follow-up and its management. Emphasis is also placed on various management strategies for treating OKC.

CASE REPORT

A 28-year-old female, on a routine follow-up after being operated on for the management of an OKC five years ago, presented with a chief complaint of maxillary right-sided maxillofacial pain persisting for the past month. Pain was dull, intermittent, and non-radiating in nature. Extraoral examination revealed no discernible signs of any facial edema or asymmetry. There was no nasal blockage/paraesthesia observed. On an intra-oral examination, a slight diffuse swelling was noted over the palatal aspect of lateral incisor on right side (Figure 1). On palpation tenderness was present over the buccal vestibular region from central incisor extending till 1st molar in first quadrant. There was also tenderness in region of palatal mucosa adjacent to lateral incisor.

Radiographic examination was done using CBCT which revealed foci of lesion begins at the periapex of lateral incisor as a well demarcated radiolucency that extends superiorly to involve anterior border of right maxillary sinus in coronal view (Figure 2). In axial view disruption of labial cortex and obliteration of the palatal cortex that measures approximately 20.0×8.9×14.00 mm in size in its greatest dimension was also noted (Figure 3).

Figure 1: Slight diffuse swelling over the palatal aspect of lateral incisor on right side.

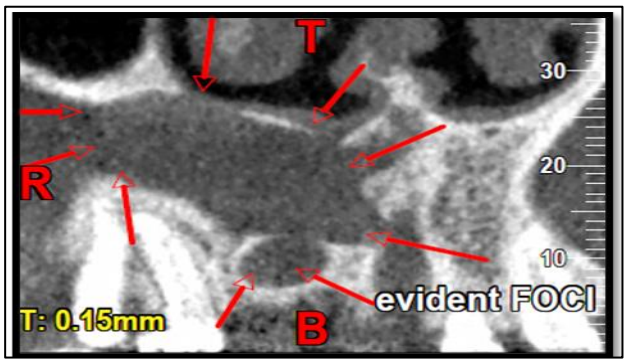

Figure 2: Coronal section of CBCT demonstrating the foci of lesion begins at the periapex of lateral incisor as a well demarcated radiolucency that extends superiorly to involve the anterior border of right maxillary sinus.

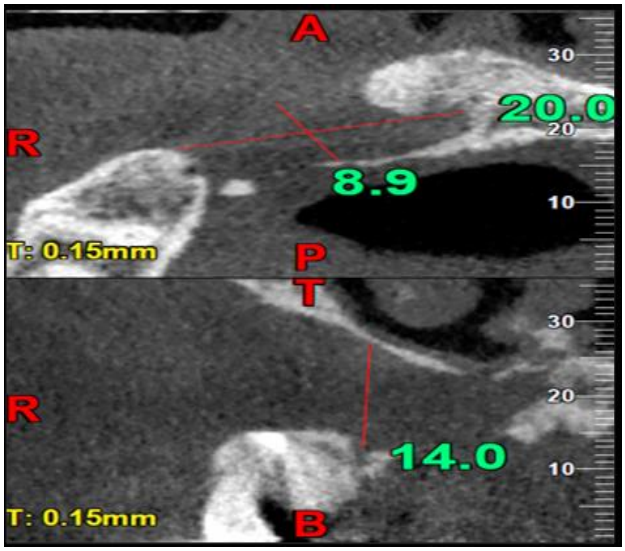

Figure 3 (A and B): Axial section of CBCT revealing the disruption of labial cortex and obliteration of the palatal cortex that measures approximately 20.0×8.9×14.00 mm in size.

Surgical intervention

An antibiotic prophylaxis dose of Augmentin 1.2 gm IV was administered one hour prior to incision. Under local anaesthesia, a crevicular incision was made from midline extending till the mesial of first molar region with a vertical extension on both the sides and a trapezoidal flap was raised (Figure 4). A bony window was created buccally, and the cystic lesion was separated by packing wet cotton gauze to isolate it from the maxillary floor between the region of 12 and 15. Additionally, cystic enucleation was performed from the palatal aspect of 12, and an apicectomy of 12 was carried out. Cotton pellets soaked in a modified Carnoy's solution were placed in the cavity for five minutes following enucleation, and a peripheral ostectomy was performed (Figure 5). Closure of defect site was done using 4-0 prolene suture. (Figure 6) Specimen was sent for histopathological examination. On the 7th day follow-up, suture removal was done.

Figure 4: A trapezoidal flap was raised.

Figure 5: Enucleation with peripheral ostectomy followed by Carnoy's application and apicectomy of 12 was done.

Figure 6: Closure of defect site using 4-0 prolene suture.

Histopathological examination

According to histological examination, submitted H and E-stained sections showed cystic lining overlying a fibro-cellular connective tissue stroma. The epithelium exhibited uniform thickness of para-keratinized stratified squamous epithelium, which was about 6-8 cell layers thick and without rete ridges. The basal layer consisted of tall, columnar cells with round to oval hyperchromatic palisaded nuclei situated away from the basement membrane. The suprabasal layer showed polyhedral cells. The surface epithelium showed a corrugated para-keratin surface. Focally, sub-epithelial hyalinization was evident. The underlying connective tissue stroma showed dense bundles of thick and thin collagen fibers interspersed with spindle-shaped fibroblasts. Varying shapes and sizes of blood vessels engorged with red blood cells along with haemorrhagic areas were seen. Diffuse mild chronic inflammatory infiltrate composed of lymphocytes was seen. And a final impression of OKC was given.

DISCUSSION

According to the world health organization (WHO), this is a benign intraosseous tumor originating from the odontogenic tissues (specifically the dental lamina and its remnants). It typically presents as a unilocular or multilocular cyst and is characterized by a lining of para-keratinized stratified squamous epithelium. Despite its benign nature, it has the potential for aggressive and infiltrative behavior. The WHO recommends the term keratocystic odontogenic tumor (KCOT) to describe this condition due to its neoplastic characteristics.[7] In the 2017 WHO classification, OKC was reclassified from a neoplastic entity back to OKC due to inadequate evidence supporting its neoplastic origin. Several studies demonstrated PTCH gene mutations in other non-neoplastic lesions such as the dentigerous cyst. Additionally, the resolution of many cases following marsupialization did not align with typical tumor behavior.[8] There is ongoing debate surrounding the optimal surgical approaches for managing OKCs, as uncertainty persists regarding which methods minimize recurrence rates (RR) while minimizing associated morbidity. In the management of OKCs, treatment strategies are categorized into conservative, aggressive, or combined approaches. Conservative methods encompass simple enucleation and marsupialization or decompression. Adjuvant techniques such as peripheral ostectomy, cryotherapy using liquid nitrogen, and application of Carnoy's solution are considered aggressive interventions, showing more favorable outcomes. Radical approaches primarily involve resection, offering the lowest recurrence rate but often resulting in significant morbidity.[9] Although KCOTs are typically benign and show minimal potential for becoming malignant, the necessity of radical surgeries like jaw resection is debatable from both medical and ethical perspectives. Consequently, a variety of supplementary techniques have emerged for managing KCOTs. The key principle in treatment planning is achieving an effective reduction in recurrence risk while opting for the least invasive surgical approach tailored to each patient.[10] The selection of treatment modality should be determined by the cyst size, recurrence history, and radiological findings indicating cortical perforation.[11] The adjunctive application of adjuvants such as Carnoy's solution, along with the utilization of liquid nitrogen in cryotherapy, has been recommended, particularly for managing cysts that are challenging to excise due to their anatomical location or multilocular nature. The primary objective of employing Carnoy's solution or cryotherapy is to mitigate the risk of cyst recurrence. The RR for Carnoy's solution and cryotherapy are reported at 14.5% and 11.5%, respectively. Carnoy's solution functions by inducing superficial tissue necrosis, thereby aiding in the eradication of residual tumor cells. The underlying rationale for using Carnoy's solution parallels that of cryosurgery, with both methodologies aiming to ablate epithelial remnants of the dental lamina at the osseous margin. An advantage of cryotherapy with liquid nitrogen

is the preservation of bone architecture, which promotes subsequent osteogenesis. The application of liquid nitrogen in cryotherapy achieves temperatures below -20°C, facilitating cellular necrosis. This necrotic process is driven by the formation of ice crystals, which disrupt osmotic pressure, leading to protein denaturation and vascular stasis.[12] More recently, topical application of 5-fluorouracil (5-FU) has been introduced because of its antimetabolic effect, which results in cellular apoptosis.[13] The follow-up period for OKCs is not clearly defined in the literature, but aggressive monitoring is recommended during the first five years following primary surgery, as recurrences have been predominantly reported within this period.[14]

CONCLUSION

OKC, a highly aggressive odontogenic cyst known for its high recurrence rate, has been treated with various surgical approaches. The existing literatures lacks a consensus on the optimal surgical approach for the treatment of OKC. Treatment options vary widely, ranging from conservative methods such as marsupialization and enucleation to more aggressive management, which includes the use of adjunctive therapies like the application of Carnoy's solution, cryotherapy, peripheral ostectomy, or a combination of Carnoy's solution application with peripheral ostectomy, as well as radical treatments like en bloc resection.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Dua P, Ghosh S, Khalid S, Grover M, Jaiswal R, Gupta A. Unmasking the enigma - recurrent odontogenic keratocyst: a continual challenge. Int Surg J 2024;11:1555-8.