ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)

IJDSIR : Dental Publication Service

Available Online at:www.ijdsir.com

Volume – 8, Issue – 3, June – 2025, Page No. : 12 - 16

Maxillary Odontogenic Keratocyst with Secondary Infection: Diagnostic Dilemma and Surgical Management

[1]Shivani Chopra, Department of Oral and Maxillofacial Surgery, KLE VK Institute of Dental Sciences, Nehru Nagar, Belagavi, Karnataka-590010, India

[2]Arati Neeli, Department of Oral and Maxillofacial Surgery, KLE VK Institute of Dental Sciences, Nehru Nagar, Belagavi, Karnataka-590010, India

Corresponding Author: Arati Neeli, Department of Oral and Maxillofacial Surgery, KLE VK Institute of Dental Sciences, Nehru Nagar, Belagavi, Karnataka-590010, India

Citation of this Article: Shivani Chopra, Arati Neeli, "Maxillary Odontogenic Keratocyst with Secondary Infection: Diagnostic Dilemma and Surgical Management", IJDSIR- June – 2025, Volume – 8, Issue – 3, P. No. 12 – 16.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Odontogenic Keratocyst (OKC) is a benign but locally aggressive cystic lesion of odontogenic origin, most commonly found in the mandible. Presentation in the maxilla, particularly with secondary infection, is less frequent and may mimic other cystic lesions. This case report describes a 25-year-old female presenting with a history of swelling in the right half of the palate following extraction of a grossly decayed maxillary first molar. Clinical and radiographic findings led to a provisional diagnosis of odontogenic keratocyst, which was confirmed by histopathology following surgical enucleation. The post-operative course was uneventful. This case underscores the importance of considering OKC in the differential diagnosis of persistent palatal swellings, particularly in the setting of recent dental extractions, and highlights the role of histopathology in the definitive diagnosis.

Keywords: computed tomography, cyst enucleation, maxilla, odontogenic keratocyst

Introduction

Odontogenic keratocyst (OKC) is a developmental cyst arising from odontogenic epithelium, characterized by aggressive growth and a high recurrence rate. Originally described by Philipsen in 1956,[1] OKC was reclassified as a keratocystic odontogenic tumor (KCOT) by the World Health Organization in 2005, but this designation was reverted to OKC in 2017.[2] Clinically and radiographically, OKCs can closely mimic other odontogenic cysts, such as dentigerous cysts, particularly when associated with unerupted teeth, making histopathological analysis crucial for accurate diagnosis.[3] OKCs occur twice as frequently in the mandible, typically in the posterior region, than in the maxilla.[4] Root resorption is an uncommon feature, reported in 1.3% to 11% of cases.[5] Radiographically, OKCs typically present as well-defined unilocular or

multilocular radiolucencies, with the multilocular form being more prevalent in the mandible.[6]

Case Presentation

A 25-year-old female patient reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of swelling on the right half of the palate since 15 days. The patient also reported extraction of the right maxillary first molar one week back in view of gross decay. No significant personal, medical, or family history was reported. Clinical examination revealed mild swelling on the right side of the mid-face extending from the nasolabial fold to the malar prominence. (Fig. 1A) The swelling was soft to firm in consistency and non-tender on palpation. Intraoral examination revealed a prominent swelling on the right anterior two-thirds of the hard palate, extending from the right maxillary first molar, continuing anteriorly till the left maxillary central incisor. (Fig. 1B) A healing extraction socket was noted in relation to 16. On palpation, the swelling was soft to firm, non-tender, and non-compressible. No active pus discharge was seen. Vitality testing revealed that teeth 12 and 13 were non-vital. An IOPA and occlusal radiograph was advised, which revealed a well-defined unilocular radiolucency surrounding the roots of the maxillary right central incisor to the right second premolar, with displacement of roots of 12,13 and 14. (Fig. 2A and 2B) Computed tomography (CT) showed bony erosion with expansion and thinning of both buccal and palatal cortical plates on the right half of the maxilla. (Fig. 3A, 3B and 3C) Based on clinical and radiographic findings, differential diagnosis of Odontogenic Keratocyst, Central giant cell granuloma, and Unicystic Ameloblastoma was made. A wide-bore aspiration biopsy was performed, which was suggestive of Subacute inflammation. Surgical enucleation of the lesion was planned under general anesthesia.

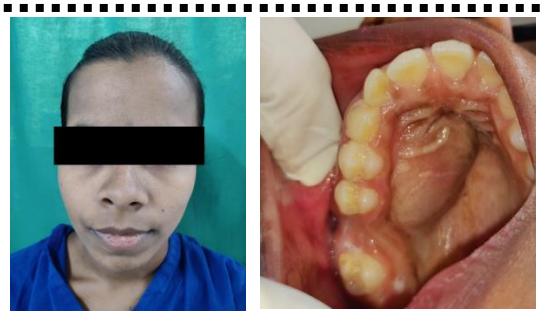

Figure 1: Pre-operative clinical pictures of the patient. (A) Extraoral. (B) Intraoral

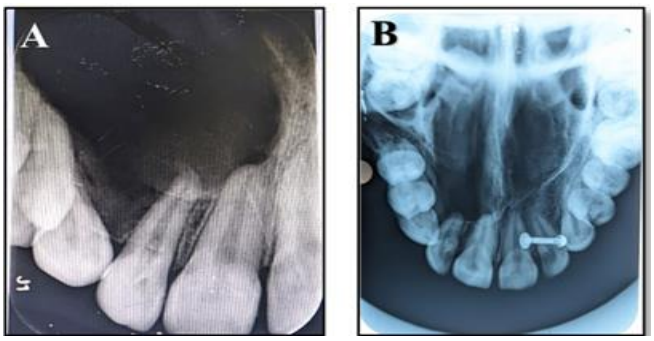

Figure 2: Pre-operative IOPA (A) and Occlusal (B) radiographs of the patient

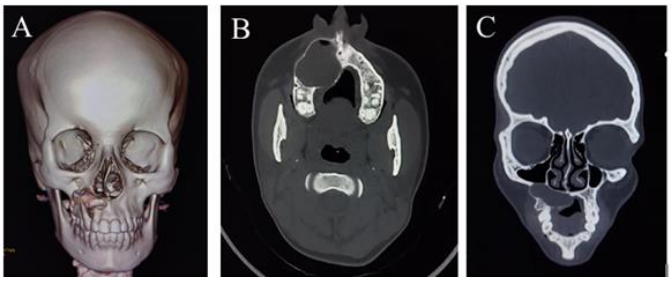

Figure 3: Pre- operative computed tomography (CT) scans of the patient. (A) 3D view. (B) Axial section. (C) Coronal section

Surgical Procedure

The procedure was performed under general anesthesia with nasal intubation. A crevicular incision was made from 21 to 17 teeth region along with an anterior releasing incision. Mucoperiosteal reflection was done carefully to expose the cystic lining. Upon reflection, due to the bony erosion, a bony defect was already present, revealing the cystic cavity. The contents of the cyst were aspirated, and the cystic epithelium was enucleated in-toto. (Fig. 4A, 4B and 4C) Extraction of

tooth 12 was done due to loss of bony support. The excised sample was sent for histopathological evaluation. Carnoy's solution was applied for five minutes to reduce recurrence risk, followed by thorough irrigation with saline and a 5% povidone-iodine solution. (Fig. 5A and 5B) The reflected flap was re-positioned over healthy bone, and closure was achieved through 3-0 vicryl sutures. A follow-up was done at 6 months which showed satisfactory healing with adequate bone formation.

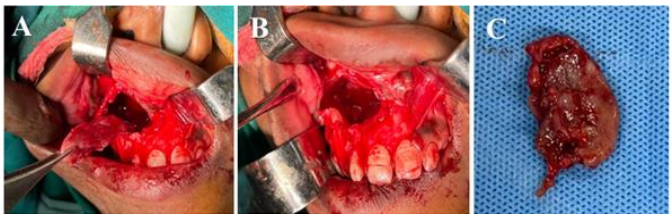

Figure 4: Intra-operative pictures of the patient. (A) Removal of the cystic lining from the bony cavity. (B) Bony cavity after enucleation. (C) Excised lesion

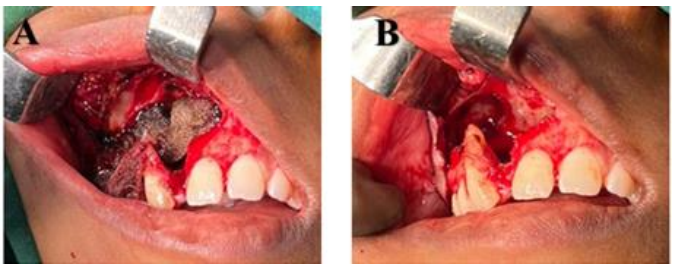

Figure 5: (A) Chemical cauterization with Carnoy's solution. (B) Bony cavity after cauterization

Histopathology

Gross examination of the specimen included multiple tissues, greyish brown in colour, soft in consistency. Following fixation in 10% neutral buffered formalin, the specimens were subjected to routine processing and staining. Histopathologically, the specimen showed a cystic space lined by 2-4 layers of stratified squamous parakeratinized epithelium showing a corrugated appearance with predominantly flat interface. Focal areas showed areading pattern of the epithelium. The surrounding connective tissue was loose, fibrillar to dense, with parallelly arranged collagen fibres with plump to spindle-shaped fibroblasts suggestive of capsule. (Fig. 6A) The capsular stroma was diffusely inflamed with chronic inflammatory infiltrate predominantly consisting of lymphocytes, plasma cells and few eosinophils. The presence of daughter cysts in the focal area was noted. (Fig. 6B) Based on the histopathology, a final diagnosis of an infected Odontogenic Keratocyst was made.

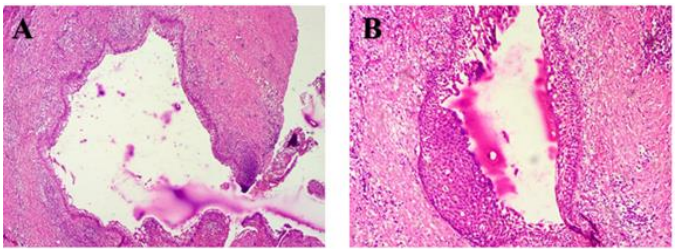

Figure 6: Histopathological pictures of the case. (A) Cystic space lined by 2-4 layers of stratified squamous parakeratinized epithelium having a corrugated appearance with surrounding loose connective tissue with parallelly arranged collagen fibres. (B) Presence of daughter cysts

Discussion

Odontogenic keratocysts (OKCs) are developmental cysts derived from the remnants of the dental lamina and represent the third most common type of odontogenic cyst.[7] They are characterized by locally aggressive behavior, a high recurrence rate, and a predisposition to secondary infection.[4,8] While OKCs predominantly affect the posterior mandible, involvement of the maxilla is relatively rare, especially when accompanied by infection.[9,10]

The non-resolution of swelling post-extraction, coupled with the non-vitality of adjacent teeth, suggested a persistent odontogenic lesion in this case. Infected odontogenic keratocysts (OKCs) frequently exhibit atypical clinical, radiographic, and histological presentations due to the presence of secondary

inflammation, which can obscure their characteristic features.[11] Histologically, OKCs are typically lined by parakeratinized stratified squamous epithelium, and they tend to expand along the medullary cavity of the bone with minimal cortical expansion, often remaining asymptomatic until they reach a considerable size.[12]

Studies have emphasized that maxillary OKCs are less common but tend to behave more aggressively due to thinner surrounding bone and proximity to vital structures like the maxillary sinus and orbit.[3]

The clinical presentation of a non-tender, non-fluctuant palatal swelling with adjacent non-vital teeth in our case was consistent with OKC, but overlapped with other odontogenic cysts such as radicular and globulomaxillary cysts. FNAC findings of subacute inflammation further complicated the diagnosis, highlighting the importance of histopathological confirmation, which remains the gold standard for diagnosis. In this case, Histopathological examination confirmed the diagnosis of an infected odontogenic keratocyst, demonstrating a parakeratinized stratified squamous epithelial lining, infiltrated with inflammatory cells. Infection often leads to epithelial lining disruption and may result in granulation tissue replacing the cyst lining.[1]

The treatment of OKC ranges from enucleation and curettage to segmental resection. Given its aggressive behaviour and high recurrence rate, the primary aim of treatment is to achieve total eradication. Nick et al. reported that Carnoy's solution is effective in routine cases, particularly when patient follow-up is feasible, whereas decompression or marsupialization followed by enucleation is a suitable approach for managing large odontogenic keratocysts, with a relatively low risk of recurrence. The final decision of treatment depends on key factors like the patient's age, cyst size, location, behaviour, appearance on radiographs, presence of perforation, or soft tissue involvement, commitment to anatomical structures, and possible complications. Since the present case was seen in a young female and owing to its aggressive nature, we had opted to perform enucleation and curettage with adjuvant therapy of Carnoy's solution. Long-term follow-up is essential in the management of odontogenic keratocysts (OKCs) due to their potential for late recurrence. Long-term follow-up is crucial in managing odontogenic keratocysts, with the highest risk of recurrence occurring within the first year post-treatment. Regular follow-ups every two years for a minimum of 25 years is recommended to detect any delayed recurrences that may develop over time.[4,9] Even with the use of Carnoy's solution, recurrence rates ranging from 1% to 8.7% have been reported,[13] and hence the patient has been kept under periodic follow-up.

Conclusion

Maxillary OKCs are less common in occurrence, and every case has a varied presentation. This case demonstrates the necessity of considering odontogenic keratocyst in the differential diagnosis of persistent palatal swellings, especially following dental extractions. Accurate diagnosis relies on a combination of clinical, radiographic, and histopathological findings. Early surgical management is essential for favourable outcomes.

References

1. Belmehdi A, Chbicheb S, Wady WE. Odontogenic Keratocyst Tumor: A Case Report and Literature Review. Open J Stomatol. 2016 Jul 14;6(7):171–8.
2. El-Naggar AK, Chan JKC, Takata T, Grandis JR, Slootweg PJ. The fourth edition of the head and neck World Health Organization blue book: editors' perspectives. Hum Pathol. 2017 Aug;66:10–2.

3. MacDonald-Jankowski DS. Keratocystic odontogenic tumour: systematic review. Dento Maxillo Facial Radiol. 2011 Jan;40(1):1–23.
4. Shear M, Speight P. Cysts of the Oral and Maxillofacial Regions [Internet]. 1st ed. Wiley; 2007 [cited 2025 May 24]. Available from: https://onlinelibrary.wiley.com/doi/book/10.1002/9780470759769
5. Bataineh AB, al Qudah M. Treatment of mandibular odontogenic keratocysts. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1998 Jul;86(1):42–7.
6. Browne RM. The odontogenic keratocyst. Histological features and their correlation with clinical behaviour. Br Dent J. 1971 Sep 21;131(6):249–59.
7. Daley TD, Wysocki GP. The small dentigerous cyst. A diagnostic dilemma. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1995 Jan;79(1):77–81.
8. Madras J, Lapointe H. Keratocystic odontogenic tumour: reclassification of the odontogenic keratocyst from cyst to tumour. J Can Dent Assoc. 2008 Mar;74(2):165–165h.
9. Blanas N, Freund B, Schwartz M, Furst IM. Systematic review of the treatment and prognosis of the odontogenic keratocyst. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2000 Nov;90(5):553–8.
10. Chirapathomsakul D, Sastravaha P, Jansisyanont P. A review of odontogenic keratocysts and the behavior of recurrences. Oral Surg Oral Med Oral Pathol Oral Radiol Endodontology. 2006 Jan 1;101(1):5–9.
11. Ahlfors E, Larsson A, Sjögren S. The odontogenic keratocyst: a benign cystic tumor? J Oral Maxillofac Surg Off J Am Assoc Oral Maxillofac Surg. 1984 Jan;42(1):10–9.
12. Shear M. The aggressive nature of the odontogenic keratocyst: is it a benign cystic neoplasm? Part 1. Clinical and early experimental evidence of aggressive behaviour. Oral Oncol. 2002 Apr;38(3):219–26.
13. Dashow JE, McHugh JB, Braun TM, Edwards SP, Helman JI, Ward BB. Significantly Decreased Recurrence Rates in Keratocystic Odontogenic Tumor With Simple Enucleation and Curettage Using Carnoy's Versus Modified Carnoy's Solution. J Oral Maxillofac Surg Off J Am Assoc Oral Maxillofac Surg. 2015 Nov;73(11):2132–5.