

Unusual Site for a Common Pathology: Odontogenic Keratocyst of the Maxilla-A Case Report

¹Dr. K.S. Manjunath, Professor and HOD, Department of Oral and Maxillofacial Surgery, Sri Hasanamba Dental College and Hospital, Vidyanagar, Hassan, Karnataka

²Dr. Shashidhara Kamath K, Professor, Department of Oral and Maxillofacial Surgery, Sri Hasanamba Dental College and Hospital, Vidyanagar, Hassan, Karnataka

³Dr Chitra G S, Postgraduate, Department of Oral and Maxillofacial Surgery, Sri Hasanamba Dental College and Hospital, Vidyanagar, Hassan, Karnataka

Corresponding Author: Dr Chitra G S, Postgraduate, Department of Oral and Maxillofacial Surgery, Sri Hasanamba Dental College and Hospital, Vidyanagar, Hassan, Karnataka.

Citation of this Article: Dr. K.S. Manjunath, Dr. Shashidhara Kamath K, Dr Chitra G S, “Unusual Site for a Common Pathology: Odontogenic Keratocyst of the Maxilla-A Case Report”, IJDSIR- July – 2025, Volume – 8, Issue – 4, P. No. 191 – 194.

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Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Odontogenic keratocyst is an aggressive cystic lesion and a common type of tooth derived cyst due to presence of odontogenic epithelial remnants in different regions of jaw. In majority of cases, it is located in mandibular posterior region. But it can also be found in the maxilla especially in the canine region. OKCs of the maxilla are particularly rare with less than 1% of cases reported in the literature. We present a rare case of OKC in alveolar process of right maxilla at the roots of molar tooth protruding into right maxillary sinus. There can be malignant transformation of this benign condition towards squamous cell carcinoma or ameloblastoma. So an early and accurate diagnosis of odontogenic keratocyst is a challenge for pathologists.

Keywords: Odontogenic Keratocyst, Carnoy’s solution

Introduction

Odontogenic keratocyst (OKC) is the third most common odontogenic cyst. The odontogenic keratocyte (OKC) is a developmental cyst arising from dental lamina remnants, first characterized by Philipsen in 1956. In the most recent WHO classification (2017), it is again classified as a cyst rather than a tumor, despite its locally aggressive behavior and high recurrence rate. OKCs represent approximately 10–15% of all odontogenic cysts, with a predilection for the posterior mandible, particularly the ramus and angle region. Radiographically, they typically present as well defined, unilocular or multilocular radiolucency with smooth, corticated margins. Histologically, OKCs are lined by a uniform Para keratinized stratified squamous epithelium, 6–8 cells thick, often with a palisaded basal layer and a corrugated

surface. Although OKCs account for roughly 10–15% of all odontogenic cysts, the vast majority develop in the posterior mandible; involvement of the maxilla is comparatively rare, representing fewer than 1% of cases in some series. OKCs of the maxilla often present unique challenges due to the region's thin cortical bone, proximity to the maxillary sinus, and potential for rapid extension into adjacent anatomic spaces. Radiographically, maxillary OKCs may appear as well circumscribed unilocular or multilocular radiolucency, sometimes causing opacification or expansion of the maxillary sinus and displacement of the sinus floor.

Case presentation

A 50-year-old female reported with a complaint of pain and pus discharge with respect to the right upper back teeth region for 3 months. The pain was intermittent, dull aching, of moderate intensity and was radiating to the ear and eye. It was also associated with pus discharge with respect to the upper right first, second premolar and first molar region for 3 months. The patient visited a local doctor a week before where they were given some medications, but there was no relief from pain. Intra oral examination revealed, the swelling was soft to firm in consistency and was tender on palpation with respect to mucobuccal fold, alveolar mucosa in the upper right posterior region. Hard-tissue examination showed missing 14,15 and 16. Orthopantomogram revealed complete maxillomandibular complex with incomplete set of teeth, a diffused radiolucency noted in right side maxillary region in relation to 14,15 and 16, involving maxillary sinus (figure-2).

Incisional biopsy was carried out, microscopic examination revealed fragments of fibro collagenous tissue few are partially lined stratified squamous epithelium, wall shows mixed inflammatory cells with congested blood vessels, which was suggestive of OKC

Case. After 15 days of incisional biopsy, surgical treatment carried out under General anesthesia. Preoperative photo shown in figure-1.

Patient painted and draped under standard aseptic precautions under general anaesthesia. Local anaesthesia achieved on the right upper vestibular region containing 2% lidocaine with 1:800000 adrenaline. crevicular incision and crestal incision placed from 22 to 18 and anterior releasing incision placed distal to 22 (figure-3). A full thickness mucoperiosteal flap is reflected to expose the cystic lining. Thickened lining of cyst removed in fragments and complete curettage of cystic cavity done (figure-4). Chemical cauterization bony cavity done with modified carnoy's solution done for 5 minutes for 2 cycles (figure-5). Bony irregular margins smoothed with acrylic bur. PRF (platelet rich fibrin) placed in the bony cavity. Haemostasis achieved and wound closure done with absorbable 3-0 vicryl (fig-6). Analgesics and antibiotics were prescribed. Recalled after 10 days and follow up was done at 4 weeks, 6 months and one year.



Figure 1:



Figure 2:



Figure 3:



Figure 4:

Discussion

Odontogenic keratocysts (OKCs) of the maxilla are exceedingly rare, accounting for less than 1% of all OKCs, yet they exhibit aggressive behaviour and a high recurrence tendency¹. OKCs arise from remnants of the dental lamina (rests of Serres), reflecting their developmental origin²³. Radiographically, maxillary OKCs present as well defined unilocular or multilocular radiolucencies that may cause maxillary sinus opacification, cortical thinning, or sinus floor elevation⁴⁵. Clinically, these lesions often remain asymptomatic until they expand sufficiently to produce facial swelling, nasal obstruction, or oroantral communication; secondary infection can mimic sinusitis with pain and purulent discharge⁶⁷.

Histologically, OKCs are characterized by a uniform Para keratinized stratified squamous epithelial lining, 6–10 cell layers thick, with a palisaded basal layer and corrugated surface; daughter (satellite) cysts within the fibrous capsule contribute to their aggressive nature⁸⁹.

Mitotic activity and molecular alterations in the PTCH1/hedgehog pathway further underlie their growth potential¹⁰. Reported recurrence rates range from 25% to 60%, driven primarily by residual epithelial remnants and satellite cysts left after simple enucleation^{11,12}. Enucleation alone carries a recurrence risk up to 56%, underscoring the need for adjunctive measures¹³.

Adjunctive chemical cauterization with modified Carnoy's solution after enucleation has been shown to reduce recurrence rates to as low as 0%–8.7%, without the neurotoxicity associated with standard Carnoy's solution^{14,15}. Conservative two stage management—initial decompression or marsupialization to reduce cyst volume and thicken the lining, followed by enucleation with peripheral ostectomy—balances morbidity and efficacy, lowering recurrence by approximately 30% compared with enucleation alone^{16,17}. In maxillary lesions, this approach preserves sinus integrity and adjacent anatomy while facilitating complete removal^{5,18}.

Given that recurrences most often occur within the first five years but may present up to 10 years post treatment, rigorous long term follow up with periodic clinical and radiographic evaluation (every 6 months for two years, then annually) is essential^{11,19}. In the present case, cystic enucleation followed by chemical cauterization with modified Carnoy's solution achieved complete removal and, at four year follow up, no recurrence has been detected.



Figure 5:



Figure 6:



Figure 7: Post-operative OPG after 6 months

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