



Case Report

Odontogenic keratocyst with atypical *Klebsiella* colonization in the maxillary sinus: A case report

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Abstract

Odontogenic keratocyst (OKC) of the maxillary sinus is very rare and occupies less than 1% of the total number of cases reported in the previous literatures with a highly susceptible to secondary infection and recurrence. This case report presents a female patient with swelling and persistent discharge of pus from the left upper posterior dental region. Patient underwent the necessary basic and advanced microbiological and radiological investigations showed an unusual instance of OKC of maxillary sinus with *Klebsiella* infection. The patient underwent surgical intervention following cyst enucleation with chemical cauterization under General anaesthesia resulting in successful management and resolution of symptoms. The case report provides additional insights into the infected odontogenic keratocyst of maxillary sinus drawing upon the existing body of literature on previously reported cases.

Keywords: Dontogenic keratocyst, Maxillary sinus, Cyst enucleation, Carnoy's, *Klebsiella* infection.

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1. Introduction

Odontogenic keratocysts, are distinctive cystic lesions arising from dental lamina remnants.¹ Characterized by their aggressive nature and high recurrence rates which resulted in a change in their classification from a cyst to a tumour by WHO in 2005 which classified them as keratocystic odontogenic tumours (KCOT) but due to lack of evidence for the same they were reclassified to cyst in 2017.² OKCs pose significant clinical challenges while predominantly occurring in the mandible, maxillary OKCs particularly those extending into the sinus are rare and present unique diagnostic and therapeutic considerations.^{3,4} The mandible is significantly more likely to be affected by OKC compared to the maxilla, with a prevalence rate of 73% versus 27%.⁵

2. Case Presentation

A 59-year-old hypertensive female sought medical attention for persisting discharge of pus from the left upper posterior tooth region, since 20 days. Initial symptoms included

unpleasant odour and a salty taste. Despite antibiotic treatment (Augmentin 625mg for 5 days) symptoms persisted but with no gross changes in facial symmetry (**Figure 1**).



Figure 1: Pre-op (Extraoral and intraoral)

Clinical examination revealed a 5mm periodontal pocket mesial to tooth 28, with pus discharge distally. Radiological investigations including panoramic radiography and CT scan (**Figure 2**) demonstrated an expansile lytic lesion with scalloped thin corticated borders in the left maxilla extending into the maxillary sinus. The cystic lesion measured

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approximately 3 x 2.6 x 4 cm exhibiting soft tissue density with patchy hyper densities.

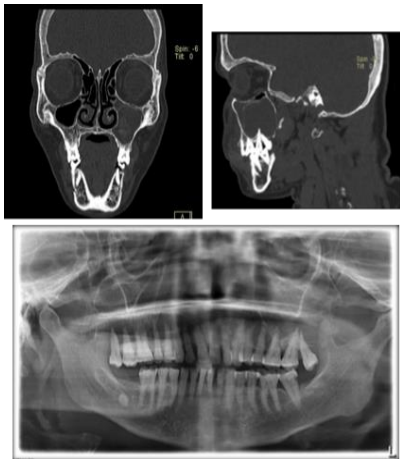


Figure 2: Pre-op radiological investigations

Microbiological analysis of pus culture collected from the maxillary sinus which was accessed through the anterior wall via Caldwell-luc approach under local anaesthesia identified *Klebsiella* species, a finding of particular interest given the rarity of such infections in maxillary sinuses.

3. Management and Outcome

Under general anaesthesia, the patient underwent meticulous surgical intervention (**Figure 3**). The procedure involved cyst enucleation and chemical cauterization using Modified Carnoy's solution. Teeth 26, 27, and 28 were extracted and the cystic lining was thoroughly removed (**Figure 4**). Post-enucleation, the cavity was irrigated with saline, betadine, and gentamicin followed by Modified carnoy's solution application for 5 minutes. The cavity was filled with Abgel and closed using 3-0 vicryl suture material along with placement of a splint.



Figure 3: Intra-op



Figure 4: Enucleated cystic lining

Postoperative care included a tailored medication regimen comprising antibiotics (Inj PIPTAZ 2.25gm IV TDS and Inj METROGYL 500 mg IV BD), anti-inflammatory drugs (Inj DEXA 8 mg IV BD), gastric protection (inj. PAN 40 mg IV BD) and pain management (Inj DICLO 75 mg IM) along with placement of Bactigrass dressing in the splint changing it every 2 days for 2 weeks. The patient was advised to maintain optimal oral hygiene and adhere to a soft diet for a time period of 2 weeks (**Figure 5**). Follow-up examination demonstrated resolution of symptoms and post 1 year follow up (**Figure 6**). There is absence of recurrence along with patient still under follow-up underscoring the efficacy of the treatment approach.



Figure 5: Immediate post-operative

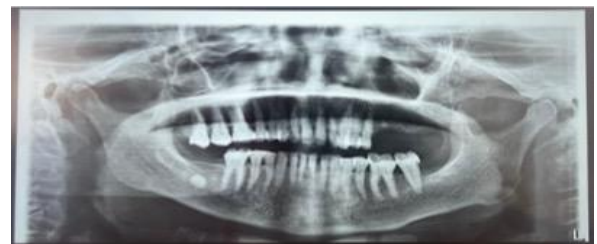


Figure 6: 1yr post-op OPG

4. Discussion

This case highlights the rare occurrence of an OKC with *Klebsiella* infection in the maxillary sinus. OKCs constituting 10-12% of jaw cysts are known for their aggressive growth and high recurrence rates.⁶ Odontogenic keratocysts in the maxilla, once considered rare were explored in a case report by Absi et al, revealing sinus invasion.⁷

The presence of *Klebsiella* species in maxillary sinus is noteworthy. While the maxillary sinus is not sterile the prevalence of *Klebsiella* in chronic rhinosinusitis has increased over the past decade often associated with antibiotic resistance.⁸ This underscores the importance of microbiological analysis in managing such cases. The successful outcome in this case can be attributed to the comprehensive approach combining surgical intervention with appropriate antibiotic therapy.

Type VI secretion system (T6SS) in *Klebsiella* which contributes to bacterial competition, cell invasion, and in vivo colonization highlighting its role in pathogenicity.⁹ Similar bacterial strains of *Klebsiella* species were also found in a

study by Jiang et al investigating the normal bacteriology in the maxillary sinus.¹⁰ This underscores the importance of microbiological analysis in managing such cases. The employment of Modified Carnoy's solution for chemical cauterization likewise aids in diminishing the likelihood of recurrence a prevalent matter in the oversight of OKCs.^{11,12}

5. Conclusion

A multidisciplinary approach is demonstrated to be crucial in the treatment of complex odontogenic lesions as exemplified by this case report. The concurrent presence of an odontogenic cyst and a *Klebsiella* infection within the maxillary sinus represents a distinctive clinical scenario with significant diagnostic and therapeutic implications. To the authors knowledge such a combination has not been previously described in the literature. Thorough radiological assessment, microbiological analysis, and tailored surgical intervention are crucial for optimal outcomes. Regular follow-up is essential to monitor for potential recurrence given the known aggressive nature of OKCs.

6. Source of Funding

None.

7. Conflict of Interest

None.

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