OPEN ACCESS

Saudi Journal of Pathology and Microbiology
Abbreviated Key Title: Saudi J Pathol Microbiol
ISSN 2518-3362 (Print) |ISSN 2518-3370 (Online)
Scholars Middle East Publishers, Dubai, United Arab Emirates
Journal homepage: https://saudijournals.com

Case Report

Odontogenic Keratocyst in Maxillary Sinus- A Rare Case Report

Dr. Sabu Paul[1], Dr. Latha Mary Cherian[2], Dr. Hafeefa, M[3*], Dr. Vally, M[3], Dr. Aishwarya Menon, P[3], Dr. Rasla, P. C[3]

[1]Associate Professor, Department of Oral Pathology and Microbiology, Government Dental College, Kottayam, Kerala 686008
[2]Professor and HOD, Department of Oral Pathology and Microbiology, Government Dental College, Kottayam, Kerala 686008
[3]Postgraduate Student, Department of Oral Pathology and Microbiology, Government Dental College, Kottayam, Kerala 686008

DOI: 10.36348/sjpm.2024.v09i01.002 | **Received:** 03.12.2023 | **Accepted:** 09.01.2024 | **Published:** 13.01.2024

***Corresponding author:** Dr. Hafeefa, M
Postgraduate Student, Department of Oral Pathology and Microbiology, Government Dental College, Kottayam, Kerala 686008

Abstract

Odontogenic keratocyst is a common and aggressive cystic lesion derived from tooth remnants in the jaw. It is frequently found in the posterior mandible and less common in the maxilla. This report presents a rare case of an odontogenic keratocyst in maxillary sinus which associated with ectopic third molar. Due to its symptomatic resemblance to other maxillary sinus lesions such as sinusitis or antral polyp, pathologists often face challenges in accurately and promptly diagnosing odontogenic keratocyst.

Keywords: Ectopic tooth, Maxillary sinus, Odontogenic keratocyst.

INTRODUCTION

The Odontogenic Kerato Cyst (OKC) is one of the more aggressive developmental odontogenic cysts affecting the jaw because of its high rate of recurrence and propensity to penetrate adjacent tissues [1]. Approximately 12% of all jaw cysts comprise of it [2]. As per the OKC incidence rate, the mandible occurs at a higher frequency (73%) than the maxilla (27%). OKC found slightly more common in males than females and around 60% cases diagnosed in the age of 10 -40 years. OKCs of the maxillary sinus are even rarer; of which just over one percent have been reported in the literature [2]. Oral surgeons and pathologists worldwide have been interested in OKCs due to their unusual behaviour, diverse origin, disputed development, discussion of treatment options, and high recurrence rate. The 24-year-old female patient in this case study had an uncommon presentation with an aggressive right maxillary sinus OKC.

CASE REPORT

A 24-year-old female patient reported to our oral medicine department with a chief complaint of pain and pus discharge from the right upper back teeth region of the jaw for the past four months. Pain was intermittent, dull aching, of moderate intensity and was radiating to ear. No nasal blockage or paraesthesia was noted. History of pain and occasional pus discharge was present 8 years back. Patient first reported to a private clinic but no treatment was given then referred to our institute.

The patient did not give any history of recent trauma on the right side of the face or the tooth. She was afebrile. Her pulse rate was 87 beats/min, and blood pressure was 120/84 mm Hg. On examination there was no gross facial asymmetry noted. There was no palpable intra oral swelling present. Erythematous mucosa was noted in relation to 14-17 region and distal to it. Tenderness present in vestibule 16, 17. Clinically missing 18. No tooth mobility was noted. Orthopantogram and computed tomography of facial bones were taken.

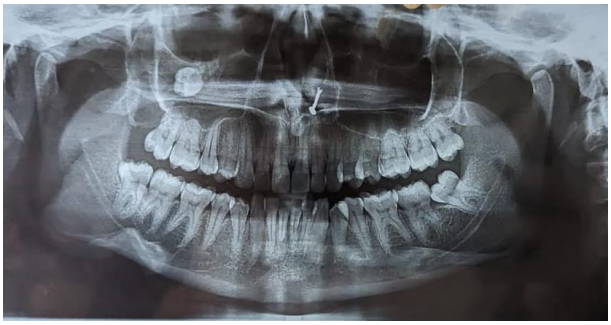

Figure 1: Orthopantomogram showing ectopically placed 18 in the maxillary sinus

On radiographic examination revealed an ill-defined, radiolucent lesion associated with an impacted third molar displaced to the right maxillary sinus. Computed tomography report shows a cystic lesion with incomplete bony wall and tooth in situ noted in the right maxillary sinus measuring 2.4x2.6x2.9 cm with bony defect inferiorly and mild bulge with erosions of medial wall of right maxillary sinus.

Figure 2: Aspiration of cystic fluid revealed a high viscous white creamy fluid

Aspiration was done using a 15-gauge disposable syringe. A highly viscous, white, creamy fluid was obtained. Analysis revealed the presence of squames and abundant eosinophilic material, which was suggestive of an odontogenic keratocyst. Therefore, enucleation of the cyst along with removal of impacted third molar was done by caldwel luc approach under general anesthesia, and the specimen was sent for histopathological examination.

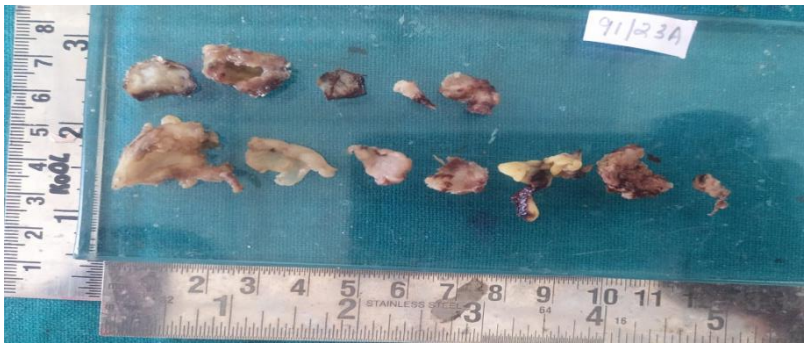

Figure 3: Multiple cyst wall like soft tissue specimens.

Gross examination showed multiple thin cyst walls, which on microscopic analysis revealed moderately collagenous cyst wall lined by a thin, parakeratinized epithelium that was 5-8 cell layers thick, without rete ridges. The parakeratin surface was corrugated, and the basal layer was palisaded, with hyperchromatic nuclei and focal areas showing reversed nuclear polarity. Therefore, the histopathological diagnosis of OKC of the maxillary sinus was made.

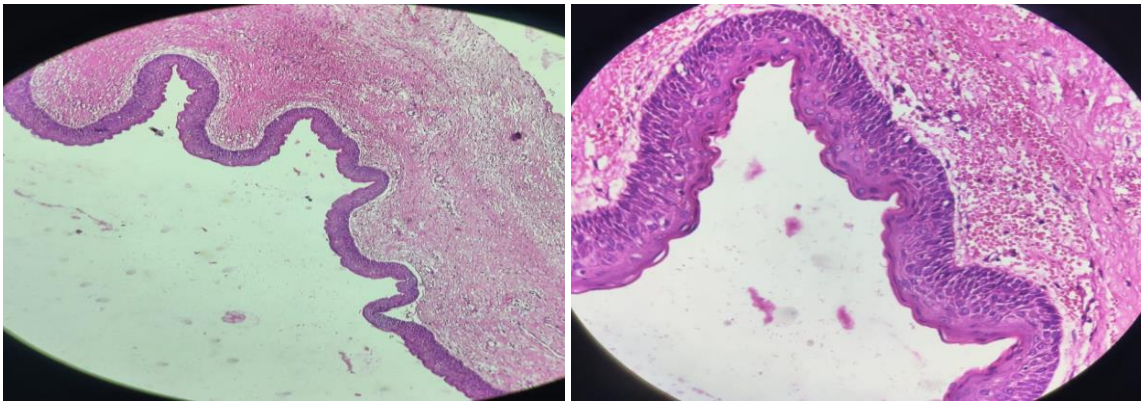

Figure 4: Epithelial lining was uniformly thin, ranging from 8-10 cell layer thickness without rete ridges.Basal layer exhibits a palisaded pattern with polarised and intensely stained nuclei. The luminal epithelial cells are parakeratinised and produce a corrugated appearance

After four months, the patient was examined and showed no symptoms consistent with the lesion. Intraorally, the site appeared to have returned to its normal architecture. The patient was kept in periodic follow-up, and an OPG will be repeated every six months. A CECT will be repeated at the one-year post-surgery visit.

DISCUSSION

The odontogenic keratocyst was first described by Philipsen in the 1956 WHO Classification of Head and Neck Tumours [3]. Pindborg and Hansen later established the histological criteria for diagnosing odontogenic keratocysts in 1962. Initially, it was believed to be a "Primordial cyst" due to its origin in the tooth primordium. Shear referred to it as "keratocystoma" and in 2004 Reichart and Philipsen called it as a "keratinising cystic odontogenic tumour." Philipsen redefined it as a "keratocystic odontogenic tumour" in 2005 [4]. However, in the 2017 classification of developmental odontogenic cysts, the term "keratocystic odontogenic tumour" was moved from the neoplastic category (2005) to the cyst category [5].

OKC is a common developmental odontogenic cyst that accounts for 10%–12% of all jaw cysts [6]. OKC is more common in men than women. It affects more persons in their second and third decades of life and gradually declines after that. The mandible is the preferred site compared to the maxilla [7, 8]. Though it can occur in any part of the mandible, in 70% of cases it arises in the posterior body and 6.9% at the symphyseal region of the mandible. In the maxilla, it is most commonly seen in the canine area, followed by the third molar tuberosity and anterior maxilla [1]. It appears that the maxilla with sinus involvement accounts for fewer than 1% of all instances of OKC.

The protein patched homolog (PTCH) gene mapped to chromosome 9q22.3 plays an important role in the development of odontogenic keratocyst. Toller and Browne believed it as a cyst derived from the dental lamina or its remnants and basal cells of the overlying epithelium. The origin of OKC in the maxillary sinus is a matter of debate. However, it is likely the result of the odontogenic epithelium becoming trapped within the sinus because of the close anatomic relationship between the floor of the sinus, dental lamina and developing antrum [9]. One interesting aspect in our presented cases is that the OKCs were completely restricted in the maxillary sinus without alveolar bone involvement. The most commonly affected site in the maxilla is between the canine and lateral incisor. It is rare for this condition to be associated with the impacted molar.

Approximately half of the patients do not experience any symptoms from the lesion. However, in some cases, patients may experience pain, swelling, expansion, drainage, and bone perforation. These lesions can grow larger than any other odontogenic cyst and are more likely to penetrate the bone rather than expand it, growing in an anterior to posterior direction. According to Philipsen (2005), even though the lesions may appear smaller in the maxilla, they are extensive due to the cancellous nature of the maxillary bone.

Radiographically, OKC appears as well-defined radiolucency which may be unilocular or multilocular. As OKC appearance in the maxillary sinus is rare, its radiographic image in such situation may be misinterpreted due to overlapping of various structures. Some authors believe that CT is superior since it able to display the important features such as size, shape, margins, bony changes, and extension of the lesion [11].

In the present case report, the radiographic examination and the computed tomography scan showed obliteration of the right maxillary sinus with ectopic third molar in it. The surgical exploration has unveiled the true pathological status with a classical histopathological picture of OKC. Its presentation with impacted third molar tooth and location made us to clinically consider the case as an infected dentigerous cyst. Hence it is important for the clinician to consider OKC in the differential diagnosis for such lesions when they occur in a younger patient. Involvement of maxillary sinus must be carefully assessed because the risk of orbital damage and infection spreading that could lead to the local and/or systemic complication. Lesion can easily expand bigger in the maxillary sinus due to the less dense structure [14].

Some study performed aspiration of cyst for diagnosis assessment. The luminal content can have different consistencies as a "straw-coloured fluid"; "thick pus like" material; or a caseous, thick, cheesy, milk white mass. The varying consistencies reflect various densities of keratinaceous debris [15]. Histopathological assessment through an incisional biopsy is the best way to diagnose the OKC before surgery when suspicion has arisen from the clinical and radiographic presentation. However, the incisional biopsy may cause inflammation to the OKC and interfere with the histopathological analysis of the post-surgical specimen. The cyst wall is fibrous and lined by a thin, parakeratinized epithelium 5-8 cell layers thick, without rete ridges. The parakeratin surface is corrugated, and the basal layer is well defined and often palisaded, with hyperchromatic nuclei and focal areas showing reversed nuclear polarity. However, as mentioned before, inflammation of the cyst may render a false-negative result secondary to the metaplasia of the cyst wall. The features of the parakeratinized epithelium and basal layer may have completely disappeared.

Several factors believed to be considered to reach the successful treatment like size, location, uni- or multi locularity, soft tissue involvement and cortical perforation [16]. There is no universally accepted treatment for OKC. Various treatment alternatives based on surgical approaches have been suggested, such as marsupialization, enucleation, enucleation with Carnoy's solution, enucleation with cryotherapy, curettage and resection. Considering its aggressive nature and history of recurrence, the primary aim of treatment is to achieve total eradication. Simple

enucleation was associated to a higher recurrence rate, while resection and enucleation with bone curettage presented lower rates.

When the lesion occurs in the maxillary sinus, Carnoy's solution is not recommended since it can penetrate the bone to a depth of 1.54 mm. Sub labial incision approach can provide direct view to the sinus ensuring the clear enucleation and curettage. One-piece enucleation with eradication of whole epithelium will give lower recurrence compared to several pieces.

Since cyclopamine, a steroidal alkaloid derived from plants, inhibits the sonic hedgehog (SHH) pathway, it may be used as a "mechanism-based" therapeutic agent to treat human tumours whose pathophysiology includes excessive SHH pathway activity. For the effective treatment of OKC, antagonists of SHH signalling factors may also be employed. The proposed tactics encompass the reintroduction of the wild-type form of the tumour suppressor gene PTCH, the inhibition of the oncogene SMO molecule through synthetic antagonists, and the suppression of the transcription factors downstream of the SHH pathway. Future treatment options with the most potential include intra cystic injection of an SMO protein-antagonist [16].

Recurrences can happen in the first two years following treatment, and some authors have reported that they have happened after ten years. Because of this, long-term follow-up and routine radiographic monitoring are crucial components of the treatment plan. A patient's functional and aesthetic appearance should be assessed as well for a higher quality of life.

CONCLUSION

The present report describes OKC in the maxillary sinus is a rare occurrence, and it usually does not present characteristic clinical and radiographic features as its central counterpart within the jaw bone. The difference between OKC and other jaw cysts is its potential aggressive behaviour and recurrence. Diagnosis of maxillary sinus OKC is challenging. CT scan and/or Magnetic Resonance Imaging (MRI) were important to differentiate the cyst with other Sino nasal neoplasms. In addition, long-term follow-up must be done to detect any recurrence associated with the lesion when it occurs in the maxillary sinus.

REFERENCES

1. Neville, B. W., Damm, D. D., & Brock, T. (1997). Odontogenic keratocysts of the midline maxillary region. *Journal of oral and maxillofacial surgery*, *55*(4), 340-344.
2. Silva, G. C. C., Silva, E. C., Gomez, R. S., & Vieira, T. C. (2006). Odontogenic keratocyst in the maxillary sinus: Report of two cases. *Oral Oncology Extra*, *42*(6), 231-234.
3. El-Naggar, A. K., Chan, J. K. C., Grandis, J. R., Takata, T., & Slootweg, P. J. (ed). (2017). International Agency for Research on Cancer.
4. International Agency for Research on Cancer, 2017.
5. Shear, M. (2003). Odontogenic keratocysts: natural history and immunohistochemistry. *Oral and Maxillofacial Surgery Clinics*, *15*(3), 347-362.
6. Shear, M., & Speight, P. M. (2007). Cysts of the Oral and Maxillofacial Regions. 4th ed. Oxford: Blackwell Munksgaard.
7. Browne, R. M. (1971). The odontogenic keratocyst: histological features and their correlation with clinical behaviour. *Brit Dent J*, 131, 249-259.
8. Brannon, R. B. (1976). The odontogenic keratocyst: A clinicopathologic study of 312 cases. Part I. Clinical features. *Oral Surgery, Oral Medicine, Oral Pathology*, *42*(1), 54-72.
9. Cioffi, G. A., Terezhalmy, G. T., & Del Balso, A. M. (1987). Odontogenic keratocyst of the maxillary sinus. *Oral Surgery, Oral Medicine, Oral Pathology*, *64*(5), 648-651.
10. Mohan, K. R., mathew Fenn, S., Thangavelu, R. P., Jeyavel, M. J., & Pancharethinam, D. (2022). An Unusually Large Parakeratinised Odontogenic Keratocyst in the Maxilla With Extension Into the Floor of the Maxillary Sinus. *Cureus*, *14*(1), e21002. DOI 10.7759/cureus.21002
11. Wright, J. M., & Vered, M. (2017). Update from the 4th edition of the World Health Organization classification of head and neck tumours: Odontogenic and maxillofacial bone tumors. *Head Neck Pathol*, 11, 68-77.
12. Mehra, P., & Jeong, D. (2008). Maxillary sinusitis of odontogenic origin. *Curr Infect Dis Rep*, 10, 205-210.
13. Pavelić, B., Levanat, S., Crnić, I., Kobler, P., Anić, I., Manojlović, S., & Šutalo, J. (2001). PTCH gene altered in dentigerous cysts. *Journal of oral pathology & medicine*, *30*(9), 569-576.
14. Wright, J. M., & Vered, M. (2017). Update from the 4th edition of the World Health Organization classification of head and neck tumours: Odontogenic and maxillofacial bone tumors. *Head Neck Pathol*, 11, 68-77.
15. Goto, M., Ueda, S., Miyabe, S., Watanabe, S., Sugita, Y., & Nagao, T. (2020). A rare case of odontogenic keratocyst extending into the sphenoid bone from the maxilla. *International Journal of Surgery Case Reports*, *71*, 132-138. 10.1016/j.ijscr.2020.05.003
16. Zhang, L., Sun, Z. J., Zhao, Y. F., Bian, Z., Fan, M. W., & Chen, Z. (2006). Inhibition of SHH signaling pathway: molecular treatment strategy of odontogenic keratocyst. *Medical hypotheses*, *67*(5), 1242-1244.