

Orthokeratinized versus parakeratinized odontogenic keratocyst: Our institutional experience

Jyoti Ravi, Vijay Wadhwan, Suhasini Palakshappa Gotur

Department of Oral Pathology and Microbiology, Subharti Dental College and Hospital, Swami Vivekanand Subharti University, Meerut, Uttar Pradesh, India

Abstract

Introduction: The odontogenic keratocyst (OKC) is quite unique among odontogenic cysts in its specific histological features and in clinical characteristics. The OKC has two variants orthokeratinized odontogenic cyst (OOC) and parakeratinized odontogenic cyst (POC), and POC is considered to be more aggressive and has a high recurrence rate. Therefore, OKC has generated considerable controversy with regard to its true nature.

Aim: The purpose of this study was to determine the differences between POC and OOC variants of odontogenic cysts on the basis of clinical as well as radiological features.

Materials and Methods: A total of 85 cases were included in the present study. There was a marked difference in the age of occurrence, sex, location and radiological presentation of both these entities.

Results: Our institutional experience suggests that POC is more common than OOC and both the variants were commonly found in males. Most of the cases of POC were found in the third decade of life and most of the cases of OOC were frequently seen in the second, fourth and fifth decades. Mandibular posterior area was frequently occurring in both variants. Left side involvement was seen in maxilla and right side in mandible in both variants. Most of the cases were clinically diagnosed as OKC.

Conclusion: The treatment and prognosis varies in both the variants of OKC, hence such epidemiological studies are helpful to ensure proper treatment for these distinct entities.

Keywords: Odontogenic keratocyst, orthokeratinized, parakeratinized

Address for correspondence: Dr. Jyoti Ravi, Department of Oral Pathology and Microbiology, Subharti Dental College and Hospital, Swami Vivekanand Subharti University, Meerut, Uttar Pradesh, India.

E-mail: ravijyoti0511@gmail.com

Submitted: 07-Dec-2020, **Accepted:** 03-Mar-2021, **Published:** 31-Mar-2022

INTRODUCTION

Odontogenic keratocyst (OKC) is the third most common odontogenic cyst and it comprises about 12% of all the cysts occurring in the maxillofacial region.^[1] The designation OKC was first employed by Philipsen in 1956 and later used by Pindborg and Hansen in 1963.^[2] It is a controversial cyst that has

undergone conceptual and terminological changes in recent decades.^[3]

OKC is so named because it is odontogenic in origin and the epithelium produces keratin that accumulates in cystic lumen.^[4] In 2005, OKC was classified as a tumor and renamed a keratocystic odontogenic tumor because of

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Ravi J, Wadhwan V, Gotur SP. Orthokeratinized versus parakeratinized odontogenic keratocyst: Our institutional experience. J Oral Maxillofac Pathol 2022;26:60-4.

Access this article online

Quick Response Code:



Website:

www.jomfp.in

DOI:

10.4103/jomfp.jomfp_498_20

its aggressive behavior, high recurrence rates and specific histological characteristics.^[5] The 2017 classification reverted back to the original and well-accepted terminology of OKC because many papers showed that the PTCH gene mutation could be found in nonneoplastic lesions, including dentigerous cysts, and furthermore, many researchers suggested that resolution of the cyst after marsupialization was not compatible with a neoplastic process.^[6] There is a lack of distinction between orthokeratinized and parakeratinized lesions. These two entities exhibit different clinicopathological characteristics,^[7] so that it is mandatory to distinguish between orthokeratinized odontogenic cyst (OOC) and parakeratinized odontogenic cyst (POC) variants due to the difference and prognosis.

Although several similar studies were carried out in other countries and regions of India, there are no demographic studies on variants of OKCs in North India. Hence, the purpose of the present study was to compare the clinical parameters such as age, sex, location and radiographic features of orthokeratinized and parakeratinized variants of OKCs.

MATERIALS AND METHODS

The data were retrieved from the Department of Oral and Maxillofacial Pathology and Oral Microbiology, Subharti Dental College, Meerut, from January 2010 to December 2019. The cases with missing clinical details, and the cases which showed a mixture of both variants, were excluded because the aim of our study is to differentiate the two separate entities. H&E-stained slides were re-examined and were re-segregated into the POC [Figure 1] and OOC variants of OKCs [Figure 2]. We differentiated OKCs as POC and OOC variants based on the lining and type of keratin production. The demographic and clinical data

such as age, sex, anatomic location with area involvement, side involvement, practitioner's clinical impression and association with an impacted tooth were obtained from the submitted biopsy form.

The criteria for determining area involvement of the lesions were as follows: (1) anterior, (2) premolar, (3) molar, (4) ramus, (5) tuberosity and the side involvement either left or right or midline. Any cyst involved in two or more areas was assigned to the location approximating the center of the lesion. Clinical impression of the practitioner, i.e., provisional diagnosis as dentigerous cyst, ameloblastoma, radicular cyst and OKC, was recorded as mentioned in biopsy requisition form.

The data were analyzed, and descriptive statistics were employed for gender, age, anatomical location and the association with impacted tooth.

RESULTS

A total of 2929 biopsy records were scanned from the archives of department, and a total of 85 OKC cases were retrieved. Among them, we found 72 cases (84.70%) of POC and 13 cases (15.30%) of OOC. In our study, of all the OKCs, 49 (57.65%) cases were observed in males and 36 (42.35%) cases were seen in females with the ratio of 1.36:1 [Table 1].

OKCs showed a wide range of age distribution (9–75 years). Most of the POCs with 25 (29.41%) cases and 12 (14.12%) cases were reported in the third and fourth decades of life, respectively, while OOC showed equal distribution, with 3 (3.53%) cases being diagnosed in the second, fourth and fifth decades of life [Table 2].

The maxilla was affected in 32 (37.65%) cases and mandible in 53 (62.35%) cases with the ratio of 1:1.65. In maxilla, the

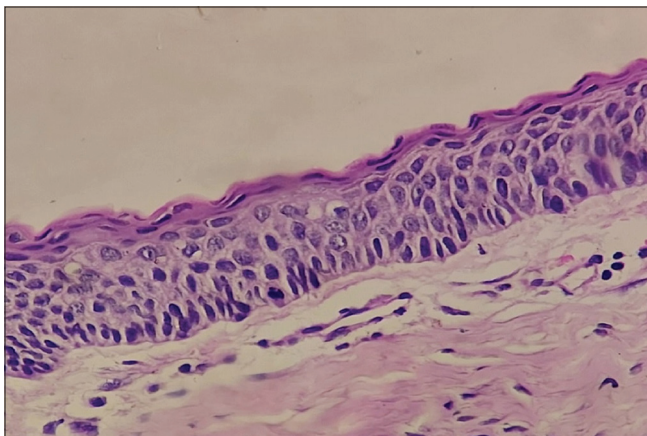


Figure 1: H and E stained photomicrographs of parakeratinized odontogenic cyst, X100

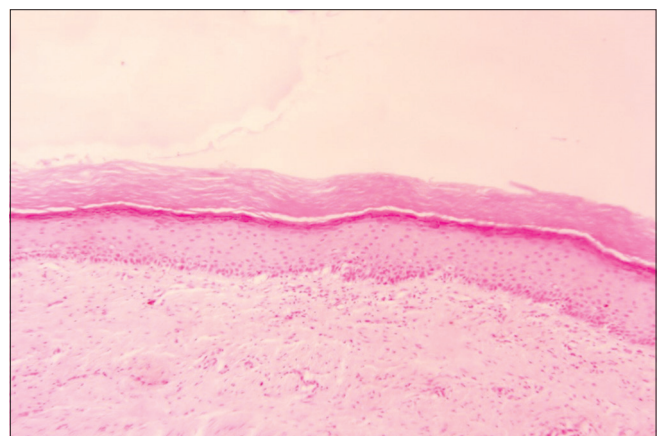


Figure 2: H and E stained photomicrographs of orthokeratinized odontogenic cyst, X100

most common site was molar area followed by tuberosity area, and in mandible, molar area was the most common site of occurrence followed by ramus area in POC. However, OOC showed equal distribution in maxilla with respect to the all-area and molar area predilection in mandible [Table 3].

Left side was commonly involved in maxilla and right side was commonly involved in mandible in both variants and very few cases crossed the midline [Table 4].

Most cases were clinically diagnosed as OKC 53 (62.36%) by surgeons at the time of surgery and misdiagnosed as dentigerous cyst 12 (14.12%) followed by ameloblastoma 10 (11.76%) and radicular cyst 10 (11.76%) [Table 5]. POC cases showed more association with impacted third molar as compared to OOC [Table 6].

Table 1: Frequency and gender distribution of odontogenic keratocyst

Types	Total (n), n (%)	Sex		Ratio
		Male, n (%)	Female, n (%)	
Orthokeratinized (OOC)	13 (15.30)	9 (10.60)	4 (4.70)	2.25:1
Parakeratinized (POC)	72 (84.70)	40 (47.05)	32 (37.65)	1.25:1
Total	85 (100)	49 (57.65)	36 (42.35)	1.36:1

OOC: Orthokeratinized odontogenic cyst, POC: Parakeratinized odontogenic cyst

Table 2: Age distribution

	Age (years)							
	0-9, n (%)	10-19, n (%)	20-29, n (%)	30-39, n (%)	40-49, n (%)	50-59, n (%)	60-69, n (%)	70-79, n (%)
OOC	0	3 (3.53)	1 (1.18)	3 (3.53)	3 (3.53)	1 (1.18)	2 (2.35)	0
POC	1 (1.17)	6 (7.06)	25 (29.41)	12 (14.12)	11 (12.94)	9 (10.59)	6 (7.06)	2 (2.35)

OOC: Orthokeratinized odontogenic cyst, POC: Parakeratinized odontogenic cyst

Table 3: Area distribution

	Maxilla					Mandible					Ratio
	Anterior, n (%)	Premolar, n (%)	Molar, n (%)	Tuberosity, n (%)	Total, n (%)	Anterior, n (%)	Premolar, n (%)	Molar, n (%)	Ramus, n (%)	Total, n (%)	
OOC	1 (1.18)	1 (1.18)	1 (1.18)	1 (1.18)	4 (4.72)	2 (2.35)	1 (1.18)	4 (4.70)	2 (2.35)	9 (10.58)	1:2.25
POC	5 (5.90)	5 (5.90)	11 (12.95)	7 (8.23)	28 (32.94)	4 (4.70)	6 (7.05)	22 (25.90)	12 (14.12)	44 (51.76)	1:1.57
Total			32 (37.65)					53 (62.35)			1:1.65

OOC: Orthokeratinized odontogenic cyst, POC: Parakeratinized odontogenic cyst

Table 4: Side comparison of odontogenic keratocyst

	Maxilla				Mandible			
	Right, n (%)	Left, n (%)	Midline, n (%)	Total, n (%)	Right, n (%)	Left, n (%)	Midline, n (%)	Total, n (%)
OOC	1 (1.18)	3 (3.53)	0	4 (4.72)	7 (8.23)	1 (1.18)	1 (1.18)	9 (10.58)
POC	10 (11.76)	16 (18.82)	2 (2.35)	28 (32.94)	23 (27.04)	17 (20.00)	4 (4.72)	44 (51.76)

OOC: Orthokeratinized odontogenic cyst, POC: Parakeratinized odontogenic cyst

Table 5: Comparison of surgeon's clinical diagnosis

Clinical misdiagnosis→ Type↓	Ameloblastoma	Dentigerous Cyst	Odontogenic Keratocyst	Radicular Cyst
OOC-13 (15.30%)	2 (2.35%)	2 (2.35%)	7 (8.23%)	2 (2.35%)
POC-72 (84.70%)	8 (9.41%)	10 (11.76%)	46 (54.12%)	8 (9.41%)
Total 85 (100%)	10 (11.76%)	12 (14.12%)	53 (62.36%)	10 (11.76%)

DISCUSSION

OKC is generally thought to be derived from either the epithelial remnants of the tooth germ or the basal cell layer of the surface epithelium or hamartomatous proliferation of odontogenic epithelium.^[8] The histogenesis of POCs and OOCs may vary and needs further investigation. From the results of this study, it would appear that the OOC variant of the OKC is histopathologically and clinically distinct from POC variant. According to Wright,^[9] histologic examination demonstrated several striking differences between the epithelial lining of OOC and POC. Although the typical POC exhibits a highly cellular parakeratinized epithelial lining with surface corrugations and a palisaded layer of basal cells, the OOC lacks these features. Instead, the thin, uniform, orthokeratinized lining epithelium was characterized by onion-skin-like luminal surface keratinization, prominent stratum granulosum and low cuboidal or flattened basal cell layer with little tendency of nuclear palisading.^[10]

The present study represents a large clinicopathological study from North India in western Uttar Pradesh population. The results of this present study showed that orthokeratinized and parakeratinized variants of OKC are distinct histopathologically and clinically.

Table 6: Associated with impacted teeth

	Associated with impacted teeth, n (%)
OOC	3 (3.53)
POC	9 (10.58)
Total	12 (14.12)

OOC: Orthokeratinized odontogenic cyst, POC: Parakeratinized odontogenic cyst

The prevalence of POC is more than OOC which is in agreement with other studies from Indian population.^[10] However, the prevalence rate varies between the two studies owing to the large sample size in the present study. In this study, the POC comprised 84.70% of all OKCs and 15.30% OOC reported in the department which is in accordance with a study conducted by Brannon^[11] who described the histological features of 312 OKCs and found that 83.2% were parakeratinized, 9.7% exhibited orthokeratinized and 7.1% had features of both.

Most of the previous studies^[10,12] have shown male prediction which was consistent with our study, with the male–female ratio of 1.36:1. In our study, the male prediction in OOCs is more than twice in males, with the male-to-female ratio being 2.25:1, similar to the data of other studies.^[9,12,13] However, another^[14] study reported a female preponderance.

In our study, most of the OOC cases were frequently seen in the second, fourth and fifth decades of life and most of the POC cases were reported in the third decade followed by the fourth decade of life. The number of cases decreased with advancing age after the fourth decade. This finding is in contrast to other studies which showed a bimodal age distribution with an additional peak in the fifth and sixth decades,^[15] while another study^[10] reported the peak incidence in the third decade of life, with a mean age of occurrence in parakeratinized OKC being 30 years and OOC being 29.1 years.

In general, all the OKCs in the present study showed a site predilection toward mandibular posterior area in accordance with many studies.^[10,12,16] However, OOCs did not show any site predilection in maxilla and its clinical significance is unknown. The current study also showed more cases of POC occurring in the midline in contrast to another study.^[13]

The difficulty in radiographically differentiating between OKC and any other odontogenic cyst and tumors was confirmed in our study and another study.^[17] In contrast with other studies,^[9,13] the current study showed that POC is more often associated with an impacted tooth than OOC. However, the reason for this finding could not be explained.

CONCLUSION

Our institutional experience provides a baseline epidemiological information regarding the incidence of OKCs. The results of the present study support the difference in the clinical behavior between POC and OOC, although the follow-up information regarding the recurrence in our record is limited. The remaining findings were comparable to previous studies of OKCs. The aggressive nature of POC requires site- and size-based treatment, ranging from simple enucleation or marsupialization, with Carnoy's solution up to radical excision while OOC, on the other hand, is less aggressive and requires conservative treatment preferably. Thus, the knowledge of these data will be helpful to ensure proper treatment for these distinct entities.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Sheethal HS, Rao K, Umadevi HS, Chauhan K. Odontogenic keratocyst arising in the maxillary sinus: A rare case report. *J Oral Maxillofac Pathol* 2019;23:S74-7.
2. Nayak MT, Singh A, Singhvi A, Sharma R. Odontogenic keratocyst: What is in the name? *J Nat Sci Biol Med* 2013;4:282-5.
3. Ribeiro-Júnior O, Borba AM, Alves CA, Gouveia MM, Deboni MC, Naclério-Homem MD. Reclassification and treatment of odontogenic keratocysts: A cohort study. *Braz Oral Res* 2017;31:e98.
4. Passi D, Singhal D, Singh M, Mishra V, Panwar Y, Sahni A. Odontogenic Keratocyst (OKC) Or Keratocystic Odontogenic Tumor (KCOT). Journey of OKC from cyst to tumor to cyst again: Comprehensive review with recent updates on WHO classification. *Int J Curr Res* 2017;9:54080-6.
5. Joshi A, Anthwal N, Shah A, Zaidi MA, Singh S. OKC or KCOT – From diagnosis to treatment. *Ann Int Med Dent Res* 2020;6:2395-822.
6. Pavelić B, Levanat S, Kobler PC, Manojlović SA, Sutalo J. PTCH gene altered in dentigerous cysts. *J Oral Pathol Med* 2001;30:569-76.
7. Hadziabdic N, Dzinovic E, Udovicic-Gagula D, Sulejmanagic N, Osmanovic A, Halilovic S, et al. Nonsyndromic examples of odontogenic keratocysts: Presentation of interesting cases with a literature review. *Case Rep Dent* 2019;2019:9498202.
8. Mukherjee D, Gopal Pati A. Odontogenic keratocyst involving mandible - A case report. *Int J Adv Res* 2018;6:635-41.
9. Wright JM. The odontogenic keratocyst: Orthokeratinized variant. *Oral Surg Oral Med Oral Pathol* 1981;51:609-18.
10. Selvamani M, Devi AY, Basandi PS, Madhushankari GS. Prevalence and clinicopathological comparison of keratocystic odontogenic tumor and orthokeratinized odontogenic cyst in South Indian sample population: A retrospective study over 13 years. *J Pharm Bioall Sci* 2014;6:127-30.
11. Brannon RB. The odontogenic keratocyst: A clinicopathologic study of 312 cases. *Oral Surg* 1976;42:54-72.
12. Dong Q, Pan S, Sun LS, Li TJ. Orthokeratinized odontogenic cyst: A clinicopathologic study of 61 cases. *Arch Pathol Lab Med* 2010;134:271-5.
13. Crowley TE, Kaugars GE, Gunsolley JC. Odontogenic keratocysts:

- A clinical and histologic comparison of the parakeratin and orthokeratin variants. *J Oral Maxillofac Surg* 1992;50:22-6.
14. Chirapathomsakul D, Sastravaha P, Jansisyanont P. A review of odontogenic keratocysts and the behavior of recurrences. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006;101:5-9.
 15. Rachanis CC, Shear M. Age-standardized incidence rates of primordial cyst (keratocyst) on the Witwatersrand. *Community Dent Oral Epidemiol* 1978;6:296-9.
 16. Kotwaney S, Shetty P. Orthokeratinized odontogenic cyst: A milder variant of an odontogenic keratocyst. *Univ Res J Dent* 2013;3:101-3.
 17. Myoung H, Hong SP, Hong SD, Lee JI, Lim CY, Choung PH, *et al.* Odontogenic keratocyst: Review of 256 cases for recurrence and clinicopathologic parameters. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2001;91:328-33.