

# Odontogenic Keratocysts in a Malaysian Population: Clinical, Radiological and Histological Considerations

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## ABSTRACT

This paper represents a study of 35 cases of odontogenic keratocyst (OKC) diagnosed in a Malaysian population. The clinical, radiological and histopathological features, treatment modalities and recurrence of this lesion were analysed. There was a higher prevalence rate among Malaysian Chinese (51.42 percent), while the Malays and Indians accounted for 22.86 percent and 25.72 percent respectively. The male to female ratio was 16 : 19. Age at presentation ranged from nine to 71 year, with about 70 percent occurring between ages ten to 39 years. A total of 40 cysts were analysed. Of these, 67.5 percent occurred in the mandible and 32.5 percent in the maxilla. About 88 percent of OKC presented as unilocular radiolucencies and 42.0 percent were associated with unerupted or impacted teeth. Histologically, the parakeratinized OKC was the predominant variant found (75.8 percent). The majority of cases were treated by enucleation (85 percent), and a recurrence rate of 20 percent was recorded.

**Key Words:** *Odontogenic keratocysts, radiological features, histological analysis, treatment and recurrence.*

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## INTRODUCTION

The odontogenic keratocyst (OKC) is an unusual odontogenic cyst because of its unique clinical behaviour and high incidence of recurrence. It is a well studied subject, but controversy still exists with regards to its pathogenesis, aggressive nature and the best approach to management of this lesion. In the context of the Malaysian population, there has been only one study done six years ago (1). The present study will attempt to give an update and a more detailed study of the clinical, radiological, histopathological and treatment aspects of this lesion as it occurs in the Malaysian population. In addition, present findings were compared with those of similar studies.

## MATERIALS AND METHODS

Clinical records, radiographs and microscopic slides of all patients with odontogenic keratocyst treated in the Faculty of Dentistry, University of Malaya from 1976 to 1992 (a total of 17 years) were retrieved and studied. The results obtained were tabulated, analysed and discussed.

Radiographs were analysed with respect to margin characteristics of the lesion, lower border and buccolingual expansion of the mandible, and association of the lesion with unerupted or impacted teeth.

Histological sections of each cyst were routinely stained in haematoxylin and eosin and examined by one of the authors (CHS), with the use of the Olympus BH-2 light microscope. These sections were studied without reference to their original biopsy reports. A diagnosis of odontogenic keratocyst was made based on the histological criteria of Kramer et al (2). Specific

changes in the lining epithelium, cyst wall and lumen were analysed and the results obtained were tabulated (Tables 4a and 4b).

## RESULTS

Prevalence and site distribution:

In 17 years, i.e between 1976 to 1992, a total of 35 cases of odontogenic keratocysts were recorded. Data on the prevalence of this lesion by sex, race and age are detailed in Table 1 and by site in Table 2.

**Table 1** Prevalence of odontogenic keratocysts by sex, race and age

		No. of cases:
- by sex	MALE	: 16 (45.71%)
	FEMALE	: 19 (54.29%)
- by race	CHINESE	: 18 (51.42%)
	MALAY	: 8 (22.86%)
	INDIAN	: 9 (25.72%)
- by age	9 years old	: 1
	10 - 19	: 10
	20 - 29	: 8
	30 - 39	: 7
	40 - 49	: 3
	50 - 59	: 4
	60 - 69	: 1
	71 years old	: 1

Age ranged from nine to 71 years. (median : 28 years) Twenty-five out of 35 cases (about 70%) occurred between ages ten to 39 years.

**Table 2** Distribution by site

	No. of cases:
<b>MAXILLA ONLY</b>	
unilateral	: 10
bilateral	: 0
<b>MANDIBLE ONLY</b>	
body only - unilateral	: 8
body only - across midline	: 4
3rd molar - ramus	: 8
bilateral (3rd molar - ramus)	: 2
<b>MAXILLA &amp; MANDIBLE (3rd molar to ramus)</b>	
unilateral	: 1
bilateral	: 1
unknown	: 1

4 cases (10%) had more than 1 cysts at presentation.

Total no. of cysts = 40

Maxillary cysts = 13 (32.50%)

Mandibular cysts = 27 (67.50%) - body = 12 (30.00%)  
ramus = 15 (37.50%)

**Table 3** Radiological findings

	No. of cases	mand.	max.
<b>1) MARGIN CHARACTERISTICS</b>			
UNILOCULAR: Smooth	: 21 (63.64%)	15	6
Scalloped	: 4 (12.12%)	3	1
Indistinct	: 4 (12.12%)	1	3
MULTILOCULAR: Scalloped	: 4 (12.12%)	4	0
<b>2) LOWER BORDER OF MANDIBLE</b>			
Eroded	: 11 (47.8%)		
Expanded	: 1 (4.3%)		
<b>3) BUCCO-LINGUAL EXPANSION OF MANDIBLE</b>			
From lower occlusal view	: 3		
Buccal only	: 1		
Lingual only	: 1		
Buccal & lingual	: 1		
From clinical information	: 9		
Buccal only	: 9		
<b>4) ASSOCIATION WITH UNERUPTED/ IMPACTED TEETH</b>			
	: 14 (42.42%)		
<b>5) ROOT RESORPTION</b>			
	: 2 (6.06%)		

**Symptoms:**

The most commonly recorded symptom was a swelling (57.14 percent of cases) and 50 percent of these swellings were painful. Recurrent discharge was the chief complaint in eight cases (22.86 percent). Lower labial numbness was uncommon and was noted in two out of 27 mandibular cysts (7.41 percent).

**Table 4** Histological findings : Epithelial characteristics

	No. of cases
<b>A) Keratin layer</b>	
i. Localized corrugation	: 25 (75.8%)
ii. Generalized corrugation	: 4 (12.1%)
iii. No corrugation	: 4 (12.1%)
<b>B) Thickness of epithelium (spinous layer)</b>	
i. 5-6 cells	: 14 (42.4%)
ii. 7-10 cells	: 8 (24.3%)
iii. 5-6 to 7-10 cells	: 9 (27.3%)
iv. >7-10 cells	: 1 (3.0%)
v. <5-6 cells	: 1 (3.0%)
<b>C) Rete ridges</b>	
i. Present	: 13 (39.4%)
ii. Absent	: 20 (60.6%)
<b>D) Basal cell layer</b>	
i. Columnar	: 2 (6.1%)
ii. Cuboidal	: 3 (9.1%)
iii. Columnar - cuboidal	: 28 (84.8%)
<b>E) Epithelial separation</b>	
i. Present	: 29 (87.9%)
ii. Absent	: 4 (12.1%)
<b>F) Epithelial infolding</b>	
i. Present	: 17 (51.5%)
ii. Absent	: 16 (48.5%)

**Provisional diagnoses:**

In 15 cases (42.8 percent), a provisional diagnosis of odontogenic keratocyst was made. Other diagnoses in decreasing frequency included dentigerous cyst (20 percent), radicular cyst (11.4 percent), residual cyst (11.4 percent), cystic lesion (8.6 percent), ameloblastoma (2.9 percent) and chronic osteomyelitis (2.9 percent). In only five cases (14.3 percent) was a diagnosis established histologically prior to definitive treatment.

**Radiological findings:**

Radiographs were available for study in 28 cases, out of which a total of 33 cysts were examined and findings detailed in Table 3. The size of cysts as measured from radiographs varied from 10mm square to 110mm x 40mm. However, it is difficult to standardize measurement of cysts from radiographs because of variable magnification. Comparison of radiographic findings of primary cysts which recurred with those which did not showed no significant difference.

**Histological findings:**

Out of the 35 cases studied, there were two primary cysts without typical features of keratocysts (probably because of infection or inflammatory reaction).

In the remaining 33 cases, the cysts bore features that fulfilled the diagnostic criteria of an OKC. Results of the histological analysis of these 33 primary cysts are detailed in Tables 4a and 4b. Comparison of the histology of the primary and recurrent odontogenic keratocysts showed no significant difference.



**Treatment modalities and recurrence:**

A total of 40 cysts were treated and treatment modalities included :

1. Marsupialization followed by enucleation and primary closure five to twelve months later ( five cysts or 12.5 percent).
2. Enucleation and primary closure (22 cysts or 55 percent).
3. Enucleation and the cavity packed with Whitehead varnish ribbon gauze, allowing granulation from the depth of cavity (12 cysts or 30 percent).
4. One case of partial maxillary resection (2.5 percent).

Follow-up was generally not satisfactory. Twenty-one cases ( 60 percent ) were lost to follow-up following successful initial treatment. Three patients (8.6 percent) were well when seen six months later but failed subsequent follow-up. Eleven (31.4 percent) were followed up for a period of one to nine years (mean: 4 1/2 years). Seven cases presented with recurrence, of which one case had recurrence at two different sites. Of the eight recurrent cysts, three were asymptomatic, four presented with swelling and one was painful. The time lapse between recurrence and operation ranged from one to five years. Analysis of recurrence by treatment modalities is as follows : marsupialization - two out of five cysts (40.0 percent); enucleation with primary closure four out of 22 cysts (18.2 percent) and enucleation with open packing of cavity - two out of 12 cysts (16.7 percent). There was no follow-up for the sole case of partial maxillary resection.

**DISCUSSION**

Analysis of this series of OKC in terms of sex, age and race distribution revealed some interesting features. Generally OKC is reported to be found more frequently in males than females (1,3-7). The present series however shows a slight female preponderance with a female to male ratio of 1.2 : 1. It was also generally accepted that OKC may occur over a wide age range with a peak incidence in the second and third decades. However, some series had demonstrated a bimodal age distribution with first peak in second and third decades and a second lower peak in the fifth decade (7-10). In this series, age at presentation ranged from nine to seventy-one years (median : 28 years). The peak incidence was in the second to fourth decades, with 70 percent of cases occurring between ages ten to thirty-nine years, and no second peak. There was a notably high incidence of occurrence of OKC in Malaysian Chinese (51.42 percent) giving a ratio by race in decreasing frequency of Chinese : Indian : Malay = 2 : 1 : 0.89. (According to 1988 survey, the population ratio of these three races in the same order is approximately Chinese : Indian : Malay = 3.7 : 1 : 5.8.). This preponderance of Malaysian Chinese was also reported by Siar et al (1). The possibility of a racial predilection had also been suggested by other researchers. Rachanis and Shear (7) and Brannon (3) had reported a considerably higher incidence of OKC in Caucasians than blacks.

Odontogenic keratocysts tend to involve the mandible much more frequently than the maxilla. The frequency of mandibular involvement reported ranged from 65 percent to 83 percent, with about 70 percent of these cysts occurring in the third molar to ramus region (3,4,11). Mandibular involvement of 67.5 percent in this series falls within this range but a smaller percentage (56 percent) occurred in the third molar to ramus region.

Odontogenic keratocyst may be discovered incidentally or the patients may complain of swelling, pain, recurrent discharge or lower labial numbness (12). Analysis of symptoms by frequency was rarely done. The present results could only be compared with another local study by Siar et al (1) who reported that 66 percent of the cases in their series complained of swelling with 22.9 percent of the swelling being painful, and 45.7 percent complained of chronic discharging sinus. This series however showed that 57.14 percent complained of swelling with half of these swelling being associated with pain, and 22.9 percent complained of recurrent discharge. Lower labial numbness was uncommon and was noted in two cases (7.41 percent) in the present series, and one case (less than two percent) in the series reported by Siar et al (1).

Based on clinical features and radiological characteristics, it is quite often possible to make a diagnosis of OKC. But OKC can also mimic other odontogenic cysts, especially dentigerous cysts or even ameloblastoma. Haring et al (4), reported in their series of 60 patients, that only 23 percent of the provisional diagnosis were correct and that the most common diagnosis made was that of a dentigerous cyst. In the present study, 42.86 percent were correctly diagnosed as OKC and 20 percent diagnosed as a dentigerous cyst.

Forsell et al (6) described the characteristic radiological features of a typical mandibular OKC as unilocular with a scalloped or smooth margin, or multilocular and tending to show distinct borders (Fig. 1). There is little or no bony expansion, and expansion if present is more often lingual than buccal. The lower border tends to be eroded rather than expanded. Displacement of unerupted or impacted teeth is common but root resorption is rare. The present series

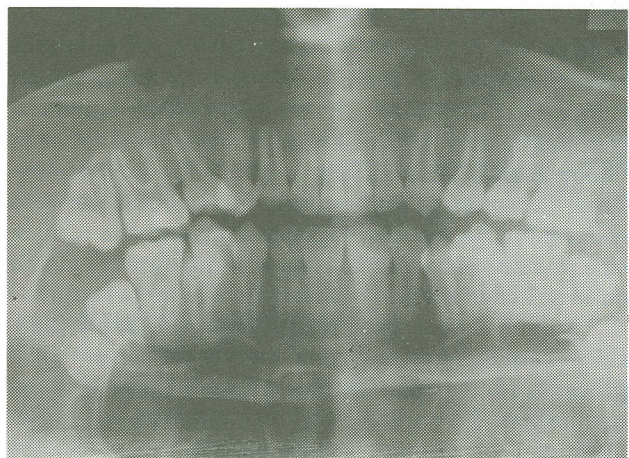
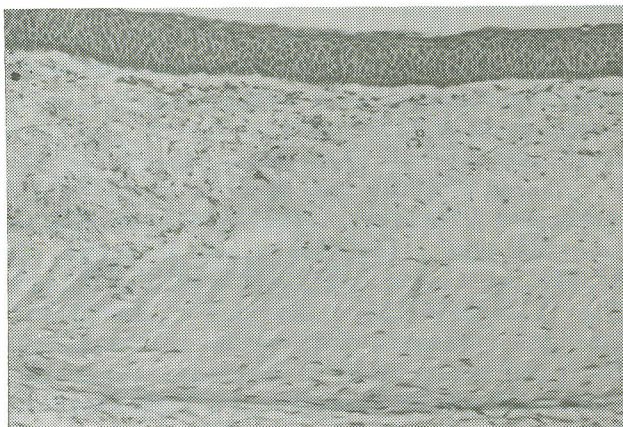


Figure 1. Bilateral lesions in the ramus of mandible, showing typical radiological features of OKC.



showed that the majority (63.64 percent) were unilocular with smooth margin. The lower border of mandible was eroded in 47.8 percent of cases. There were ten mandibular cysts (43.5 percent) presenting with buccal expansion. This finding is in agreement with Browne (11) who reported that about one-half of the mandibular lesions produced buccal expansion. About 42.4 percent of the OKC in the present study were associated with unerupted or impacted teeth. This is close to the findings of Forssell (41 percent) (9), but is higher than that reported by Haring and Van Dis (26.7 percent) (4). Such frequent association of OKC with unerupted teeth makes it likely for the lesion to be mistaken for a dentigerous cyst. Root resorption is uncommon. The present figure of 6.06 percent compares favourably with the five percent frequency of root resorption reported by Haring and Van Dis (4).

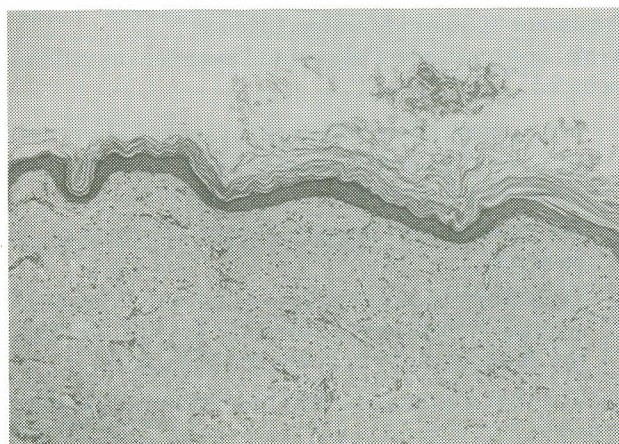
Histological analysis of the 33 OKC disclosed certain similarities and discrepancies in findings when compared with those of published reports. The recorded percentage of parakeratinized odontogenic keratocysts (Fig. 2) was higher than one other local series (72.3 percent) (1) but lower than those in Caucasian series (83.2 percent to 98.3 percent) (4,13-15). On the other hand, odontogenic keratocysts with features of both ortho- and parakeratin was highest in this series compared to both local (18.5 percent) (1) and Caucasian



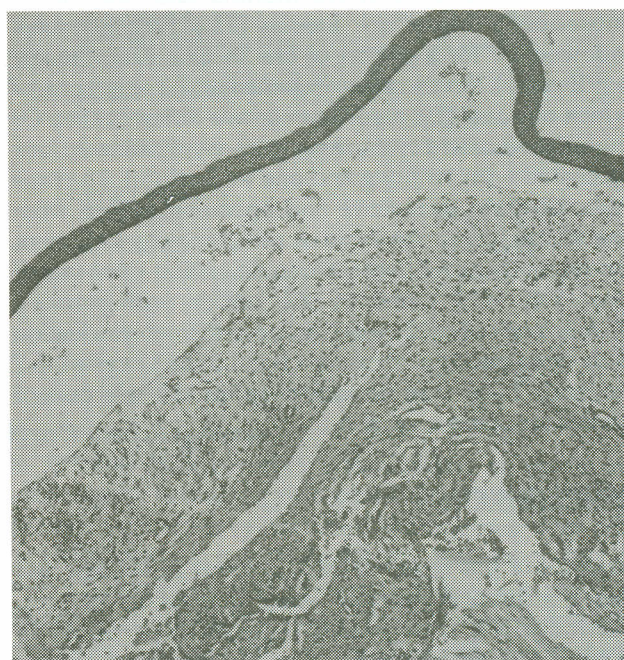
**Figure 2.** Parakeratinized odontogenic keratocyst. Note palisading of basal columnar cells, absence of rete ridges and underlying uninfamed fibrous connective tissue wall. H & E X 100

studies (1.6 percent to 7.1 percent) (13,14). However orthokeratinization (Fig. 3) was low in this series in comparison with one other local report (9.2 percent) (1) but falls within the documented range for Caucasian odontogenic keratocysts (0 percent to 12.2 percent) (4,14). These variations in findings may be attributed partly to geographic factors and partly to differences in sample size (the present series of cysts is comparatively small) and subjective method of assessment of these features.

Epithelial separation (Fig. 4) as noted in this series is low compared to published reports (94 percent to 98 percent) (4,13). The precise cause for this distinct tendency of the lining epithelium to detach away from the underlying connective tissue remains to be elucidated but factors including preparation artefact (16), absence of rete ridges (17), poorly-developed basement



**Figure 3.** Orthokeratinized odontogenic keratocyst with surface orthokeratin corrugation. Note absence of rete processes. H & E X 40



**Figure 4.** Detachment of epithelial lining from underlying connective tissue. Note the flat epithelial-connective tissue interface. H & E X 40

membrane (17) and collagenolytic activity (18) have been implicated. The significance of this feature is that remnants of detached lining may be left behind following surgical removal of an odontogenic keratocyst and this may contribute to a recurrence (13).

In Brannon's series, findings on epithelial infoldings were graded as prominent (86.9 percent), mild (7.4 percent) or absent (5.8 percent) (13). Within this same study, 21.8 percent of odontogenic keratocyst had satellite cysts and 13.5 percent contained epithelial rests (13). In contrast, all these three features (Fig. 5 and 6) were uncommon in this series. The reported frequency of satellite cysts in odontogenic keratocysts may range from seven percent to 33.3 percent (5,13,15,16) while epithelial rests were found in 13.5 percent to 60.0 percent of odontogenic keratocysts (4,6,13). The reason for this wide variation in findings is unknown but nevertheless



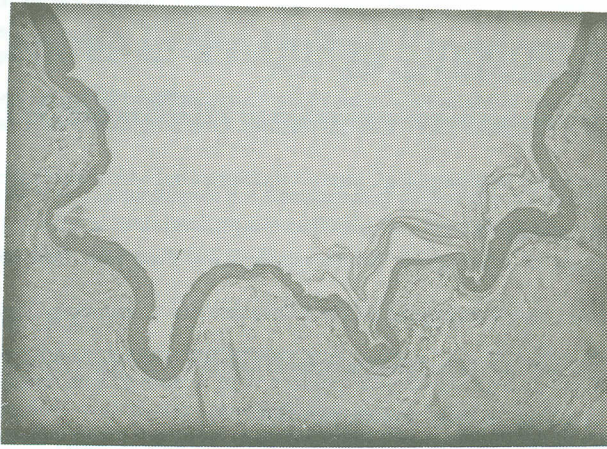


Figure 5. Epithelial infoldings. H & E X 60

both these structures are regarded as indications of the proliferative capacity of the lining epithelium and a higher frequency of these structures had been reported in odontogenic keratocysts associated with basal cell naevus syndrome (19).

Reports on the presence or absence of an inflammation in the connective tissue wall of odontogenic keratocysts remain conflicting. Some studies stated that little or no inflammation was observed in the odontogenic keratocysts analysed (17,20,21). Others reported that a high percentage of odontogenic keratocysts (72.4 percent to 98.0 percent) presented with a predominantly chronic inflammatory cell infiltration and that this was most often in a generalized pattern of distribution (4,15). Present findings accord with the latter except that a localized rather than generalized inflammatory pattern was observed. Immunocytochemical analysis disclosed that the inflammatory cell infiltrate comprised HLA-DR histiocytes and a mixture of T and B lymphocytes (22).

Cholesterol granulomas which were rather rare in this series had been reportedly found in 11.5 percent to 18.3 percent of odontogenic keratocysts (4,5,13,15,17). The source of the cholesterol is not known but local tissue damage due to severe inflammation had been cited as a possible cause.

Brannon (13) found that in 30.8 percent of OKC the lumen was full of keratin. A lower percentage was observed in this study. The variation in finding may be due to the fact that many of these cysts in the current study were removed piecemeal (probably because of their extensiveness) and an intact lumen was therefore not available for study.

It is generally known that the orthokeratinized OKC is a clinically less aggressive cyst than its parakeratinized counterpart. The cited recurrence rate for orthokeratinized OKC is 2.2 percent (14) to 4.2 percent (24) while for parakeratinized OKC it is 42.6 percent (14). In this series, as there is only one orthokeratinized OKC (with no recurrence), similar analysis along these aspects could not be made.

Recurrence of OKC has been reported as varying from five percent (5) to 62 percent (21). This wide discrepancy may partly be explained by the variation in follow-up period and treatment method (26). In the present series, recurrence rate was found to be 20 percent. This is comparable to Eversole et al's report (27) of 20 percent recurrence. The time lapse between operation and recurrence was one to five years. This concurs with reports that recurrence tends to occur within the first five years (8,26). However, follow-up in the present

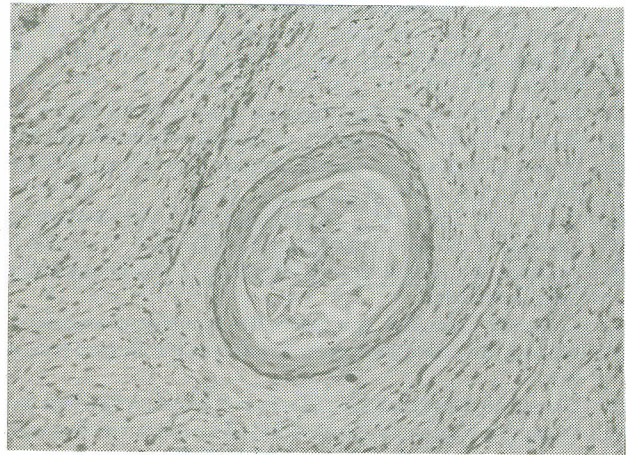


Figure 6. Satellite cyst with central keratin accumulations. H & E X 100

series was generally unsatisfactory due to the tendency for patients to seek treatment only when there were signs and symptoms rather than abiding by the recommended yearly review.

Analysis of recurrence rate of each method seems to suggest that it is highest with marsupialization (40 percent) compared with 16.7 percent to 18.2 percent in cases of enucleation. The recurrence rate does not appear to be significantly different between enucleation followed by primary closure (18.2 percent) and secondary granulation (16.7 percent). A high recurrence rate (60 percent) with marsupialization was also recorded by Forssell et al (26) while Browne (11) found no difference in recurrence rate between enucleation and marsupialization.

Extremely aggressive behaviour of the cyst was not found in the present study. Conservative treatment by thorough enucleation appears to be satisfactory. Although there was a recurrence rate of 20 percent in this series, recurrences were effectively treated by enucleation and primary closure. No second recurrence was recorded.

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