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內文：

### I. Introduction

1. KCOT renamed by WHO in 2005
2. Definition: A benign uni- or multicystic intraosseous tumor of odontogenic origin with a characteristic lining of parakeratinized stratified squamous epithelium and potentially aggressive infiltrative behavior. It may be solitary or multiple. The latter is usually one of the stigmata of the inherited nevoid basal cell carcinoma syndrome (NBCCS).
3. KOTs are associated with inactivation of PTCH, the tumor suppressive gene.
4. This study determines the radiographic and clinical features more accurately for the Indian population, which will help in further diagnosis of keratocystic odontogenic tumor.

### II. Materials and Methods

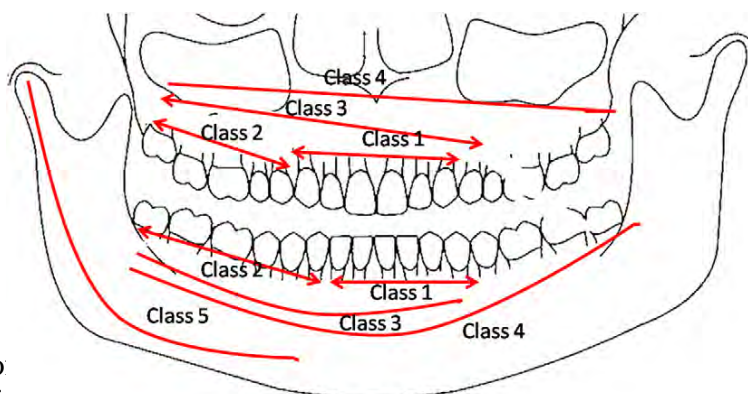
1. Searching the medical literature for the period from 1957 to March 2012.
2. Scientific databases namely Pubmed Plus, Medline (Pre-Medline and Medline), Cochrane Database of Systematic Reviews (evidence-based medicine), Dentistry and Oral Sciences Source, Access Science, Embase, Evidence-Based Medicine Reviews Multifile, Google Scholar, ISI Journal Citation Reports, and Ovid Multi-database
3. Search keywords included were keratocystic AND odontogenic AND tumor OR odontogenic AND keratocyst OR primordial.
4. Inclusion criteria: All case series, histologically confirmed as a parakeratinized variety, were included in this SR.  
(In the pre-2005 case series where both orthokeratinized and parakeratinized variety were reported, only parakeratinized reports were selected.)
5. Excluded:
  - i. Syndromic cases of NBCCS were excluded from this review, because multiple tumors in this syndrome are often not synchronous.
  - ii. Articles on the peripheral or extrasosseous variant of KOT were excluded.
6. Additional cases: Cases from Nair Hospital Dental College (Indian)
  - i. Cases diagnosed as KOT and OOC after 2005 and those diagnosed as OKC before 2005.
  - ii. Cases were reviewed for a period of 9 years, from January 2001 to

December 2009.

- iii. Final diagnosis was made on the basis of histopathology.
- iv. The various variables considered in this study were age, sex, arch and site of lesion, nature (single/multiple) of occurrence, and radiographic findings. Results compared with findings from the rest of the world.

7. Lesion site definition

	Maxillary	Mandible
Class I	tooth 13-23 distal sides	tooth 33-43 distal sides
Class II	tooth 14M-distal, 24M-distal area	tooth 34M-distal, 44M-distal area
Class III	lesions that extended into both anterior and posterior segments of the maxilla.	lesions extending into both anterior and posterior segments of mandible.
Class IV	lesions from third molar to third molar crossing the midline.	lesions from third molar to third molar crossing the midline.
Class V	X	lesions limited to posterior segment, angle of the mandible, ramus, condyle, and coronoid process.



III. Results

1. A total of 65 reports were selected for the study.
2. The results were mainly divided into 4 major groups based on origin of the genetic family: American, Caucasian, East Asian and Pacific, and African and South Asian.

American	North and south America
Caucasian	Europe
African and South Asian	Africa and Eastern, Northern, Southern Indian subcontinent
East Asian and Pacific	Southeastern Asia and the Australian continent

**Table I.** Comparison of global groupings of present and previous systematic reviews (SRs)

Present SR groups	Previous SR groups
American + Caucasian	Western + Latin American
African and South Asian	Subsaharan
East Asian and Pacific	East Asian

**Table II.** Database of cases and years of different global groups for the present and previous systematic reviews (SRs)

	American	Caucasian	American + Caucasian	Western + Latin American*	African and South Asian	Subsaharan*	East Asian and Pacific	East Asian*	Total	Total*	Additional cases
No. of reports	19	22	41	33	5	3	19	13	65	49	1
No. of KOT	2,031	1,081	3,112	2,799	554	78	2528	1,900	6194	4,777	72
No. of cases per year	5.99	4.28	5.26	5.78	8.93	0.04	7.86	6.31	6.35	5.84	8

\*Previous SR.

4. Comparison:

	African and East Asian per year ps an and Caucasian groups 1 and Subsaharan group Caucasian (74%)					
0.0789 (11)	33884 (1)	008720 (18)	810453 (12)	130508 (9)	00 (5)	
81533 (3)	1103 (1)	08388 (3)	34134 (3)	30 (1)	30 (1)	
128702 (2)	2034 (1)	338450 (9)	0383 (1)	20 (1)	20 (1)	
381 (9)	3502 (1)	3203 (13)	3022 (10)	305 (2)	1212 (5)	
213310 (0)	323515 (0)	890201 (18)	302220 (12)	233510 (4)	39 (5)	
Western + Latin	Caucasian	Caucasian + African +	African +	Eastern Asian	Sub-Saharan	

Table III. Various clinical features of cases included in the present study (no. of reports in parentheses)

	American	Caucasian	American + Caucasian	Western + Latin American*
Male:female	517:379 (9)	352:212 (9)	869:591 (18)	795:559 (15)
Mean age of presentation, y	39.1 (6)	32.93 (7)	35.93 (13)	36.75 (10)
Swelling, P:A	178:405 (5)	50:24 (1)	228:429 (6)	97:83 (4)
Pain, P:A	81:323 (2)	11:63 (1)	98:386 (3)	24:134 (1)
Mandible: maxilla	670:265 (11)	238:84 (7)	908:350 (18)	819:423 (15)

P, Present; A, absent.  
\*Previous SR.

res: NA presented, paucity of radiologic details

Table IV. Radiographic findings in the included reports

Author (year)	n	Completely radiolucent		Unilocular	Multilocular	Well defined		Buccolingual expansion		Antral involvement		Root resorption		Associated with unerupted tooth	
		Y	N			Y	N	Y	N	Y	N	Y	N	Y	N
Borello (1976) <sup>9</sup>	14	12	0	6	6	NA	NA	2	12	NA	NA	NA	NA	NA	NA
Mosadomi (1976) <sup>10</sup>	2	2	NA	NA	2	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
Chiang (1982) <sup>14</sup>	15	15	0	10	4	NA	NA	NA	NA	NA	NA	NA	NA	7	7
Nielsen (1986) <sup>20</sup>	21	NA	NA	17	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
Haring (1988) <sup>22</sup>	60	60	0	44	16	27	33	NA	NA	NA	NA	3	35	16	38
Tagesen (1990) <sup>24</sup>	38	38	0	34	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
Crowley (1992) <sup>26</sup>	387	256	0	NA	NA	NA	NA	NA	NA	NA	NA	100	109		
Santos (1999) <sup>34</sup>	40	50	0	41	9	NA	NA	NA	NA	NA	NA	NA	NA	10	50
Myoung (2001) <sup>38</sup>	256	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	70	186
Ogunsalo (2007) <sup>51</sup>	3	3	0	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
Yagyuu (2008) <sup>55</sup>	62	62	0	43	19	1	NG	NA	NA	NA	NA	NA	NA	NA	NA
MacDonald-Jankowski (2008) <sup>58</sup>	33	33	0	16	17	33	0	27	6	11	0	13	19	20	12
Ba (2010) <sup>60</sup>	274	1	NG	NA	184	62	NA	NA	NA	NA	NA	NA	NA	NA	NA
Ariji (2011) <sup>69</sup>	10	1	NG	NA	10	0	1	NG	NA	NA	NA	NA	2	8	7
Additional cases	60	60	0	38	22	58	2	32	28	NA	NA	16	35	28	32
Total	1,275	591	0	438	107	118	35	61	46	21	4	34	97	242	437

Y, Yes; N, no; NA, information not available.

- viii. Unilocular KOT: predominant in all global groups  
Except: African group (present) and Subsaharan group (previous)
- ix. Border: well defined in East Asian  
poorly defined in American group
- x. Buccolingual expansion: common in additional case
- xi. Root resorption: significantly higher in East Asian
- xii. KOT with unerupted tooth: not common in any of the groups

Table V. Comparative presentation of various radiographic features of cases included in the present and previous systematic reviews (SRs) (no. of reports in parentheses)

	American	Caucasian	American + Caucasian	Western + Latin American*	African and South Asian	Sub-Saharan*	East Asian and Pacific	East Asian*	Total	Total*	Additional cases
Uni-multilocular	91:31 (4)	51:8 (2)	142:39 (6)	142:39 (5)	0:2 (1)	0:2 (1)	320:166 (6)	69:40 (3)	462:205 (13)	211:81 (9)	38:22 (1)
Completely radiolucent, Y:N	378:0 (4)	38:0 (1)	416:0 (5)	NA	5:0 (2)	NA	110:0 (3)	NA	481:0 (10)	NA	60:0 (1)
Margin definition, good:poor	27:33 (1)	NA	27:33 (1)	27:33 (1)	NA	NA	33:0 (1)	33:0 (1)	60:33 (2)	60:33 (2)	58:2 (1)
BL expansion, Y:N	2:12 (1)	NA	2:12 (1)	2:12 (1)	NA	NA	27:6 (1)	27:6 (1)	29:18 (2)	29:18 (2)	32:28 (1)
Antral involvement, Y:N	NA	NA	NA	11:0 (1)	NA	NA	11:0 (1)	11:0 (1)	11:0 (1)	11:0 (1)	NA
Root resorption, Y:N	3:35 (1)	NA	3:35 (1)	3:35 (1)	NA	NA	15:27 (2)	13:19 (1)	16:54 (2)	16:54 (2)	16:35 (1)
Associated with unerupted tooth, Y:N	126:197 (3)	NA	126:197 (3)	126:187 (3)	NA	NA	131:261 (5)	111:250 (4)	250:455 (7)	237:437 (7)	28:32 (1)

Y, Yes; N, no; BL, buccolingual; NA, information not available.  
\*Findings of the previous SR.

**Table IX.** Statistical analysis ( $\chi^2$ ) of clinical and radiographic features

Clinical features		
Male:female, additional cases vs. Caucasian	$\chi^2 = 3.855$ (1 df); $P = .0496$	Significant
Male:female, additional cases vs. East Asian	$\chi^2 = 5.16$ (1 df); $P = .0231$	Significant
Male:female, additional cases vs. African	$\chi^2 = 8.168$ (1 df); $P = .0043$	Significant
Male:female, additional cases vs. American	$\chi^2 = 7.544$ (1 df); $P = .0043$	Significant
Mandible: maxilla: additional cases vs. African	$\chi^2 = 39.117$ (1 df); $P < .001$	Significant
Mandible: maxilla: East Asian vs. American	$\chi^2 = 108.4$ (1 df); $P < .0001$	Significant
Radiographic features		
Unilocular:multilocular, additional cases vs. Caucasian	$\chi^2 = 7.243$ (1 df); $P = .0071$	Significant
Unilocular:multilocular, east Asian vs. Caucasian	$\chi^2 = 9.344$ (1 df); $P = .0022$	Significant
Marginal definition, additional cases vs. American	$\chi^2 = 36.30$ (1 df); $P < .0001$	Significant
Root resorption, additional cases vs. American	$\chi^2 = 5.81$ (1 df); $P < .0159$	Significant

Only statistically significant results are given in this table.

- ii. Age: average age: 30.7 yrs, predominant at 3<sup>rd</sup> (n = 24) and 2<sup>nd</sup> (n = 18) decades.
- iii. Male: Female = 3:1, Mandibular-to-maxillary ratio = 2.61 : 1
- iv. Sites: most common over Class 2 area. (tooth 34M- and 44M-distal)
- v. Unilocular more common both maxillary and mandibular.

**Table VI.** Prevalence of orthokeratinized odontogenic cyst (OOC) and keratocystic odontogenic tumor (KOT) in the Indian population

Lesion	No. of cases	No. of patients in cohort	Prevalence
OOC	5	415,629	0.0012%
KOT	72	415,629	0.017%

**Table VII.** Keratocystic odontogenic tumor (KOT) distribution by sex and age

Age (y)	Male	Female	Total
0-9	0	0	0
10-19	13	5	18
20-29	18	6	24
30-39	13	0	13
40-49	6	4	10
50-59	1	1	2
60-69	2	0	2
70-79	1	2	3
Total	54	18	72

**Table VIII.** Distribution of keratocystic odontogenic tumor (KOT) in maxilla and mandible according to classification depicted in Figure 1

	Class 1	Class 2	Class 3	Class 4	Class 5	Total
Mandible	7	17	6	4	13	47
Maxilla	3	13	2	0	—	18

- 2. KOT and OOC: completely different lesions
- 3. KOT – inactivation of PTCH
  - i. Importance of genetic origin of KOT p't.
  - ii. The SR is the first compilation of KOT in Indian population
  - iii. Additional cases/ African and South Asian community
    - 1. Results might match – genetic belonging, geographic proximity
- 4. Awareness or not
  - i. American and Caucasian groups: greater number of case report and earlier diagnosed
  - ii. African and South Asian groups: higher number of cases reported per year.
  - iii. Higher number of KOTs per year in the African and East Asian groups indicates a higher incidence of KOT in these groups
- 5. Mean age
  - i. Mean age higher in American group of the present SR.
  - ii. Combined analysis of the American and Caucasian groups: didn't reflect higher mean age.
  - iii. KOT in American groups may occur much earlier.
- 6. Pain and multilocular
  - i. Rare, only 1 report presented it common in African groups.
- 7. Maxilla/Mandible predominated
  - i. Maxilla predominated in African group – because of Sri Lankan report.

8. Swelling: East Asian, Caucasian African and Latin American groups.
  - i. American groups not involved.
  - ii. Might due to increased size because of poor awareness.
9. Buccolingual expansion
  - i. Known as rare buccolingual expansion.
  - ii. Significantly higher in East Asian group.
10. Root resorption
  - i. Significantly higher in East Asian group.
11. Unerupted tooth
  - i. Significantly higher in American group.
  - ii. Might be high incidence of prophylactic removal of impacted teeth in America.
12. Additional Cases
  - i. OOC in Indian community: 6.5%, world average: 11%  
7% for Subsaharan and 8% each for the Latin American and East Asian groups
  - ii. KOT in additional cases: 93.5%,  
Subsaharan (93%), Latin American (92%), and East Asian (92%) groups but more than the Western group (89%)
  - iii. Male/ Female ratio = 3  
1.3 to 1.6 worldwide.
  - iv. Age: predominately 3<sup>rd</sup> decade, both male and female.  
1<sup>st</sup> decade of female and 3<sup>rd</sup> decade of male during previous SR.
  - v. Mandibular: Maxillary involved = 2:1
  - vi. Most common sites: class 2 > 5 > 1 > 4  
Class 4 rare might because early examination due to oral radiographic examinations..etc.
  - vii. Unilocular variant dominated: 63.3%  
American (unilocular 74.5%), and Caucasian (unilocular 86.4%), and East Asian (unilocular 54.7%) groups
  - viii. Good marginal definition  
Poorly defined borders in American group
  - ix. Root resorption: rare in India area.

**V. Conclusions**

**KOT:**

1. Painless, occasionally swelling in certain population, majorly mandible involves, Unilocular variety more common. Root resorption and buccolingual expansion mainly in East Asian group.
2. Additional cases (Indian population):
  - i. 72 KOTs and 5 OOCs.
  - ii. Male major, 3<sup>rd</sup> decade predominately.

題號	題目
1	Which one about Keratinized odontogenic tumor (KOT) and Orthokeratinized Odontogenic cyst is in correct? (A) Recurrence rate of KOT is higher than OOC. (B) KOT and OOC both predominated in posterior mandible area. (C) Histopathologic findings: KOT and OOC both has palisaded basal cell. (D) KOT and OOC are different kinds of lesion.
答案(C)	出處：Oral and Maxillofacial Pathology, edition 3

題號	題目
2	Which one about KOT was wrong? (A) Majorly infect 2rd to 3rd decade's male. (B) Root resorption and buccolingual expansion mainly happens in East Asian group cases. (C) Reclassified by WHO in 2005 and named as Keratinized Odontogenic Tumor because of it's character like high recurrence rate. (D) Associated to activation of PTCH gene and 9 <sup>th</sup> chromosome changing.
答案(D)	出處：Oral and Maxillofacial Pathology, edition 3 Keratocystic odontogenic tumor: systematic review with analysis of 72 additional cases from Mumbai, India