Keratocystic odontogenic tumor: systematic review with analysis of 72 additional cases from Mumbai, India

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Objective. The aim of this systematic review was to assess the clinical and imaging findings of keratocystic odontogenic tumor (KOT). This article also attempts to determine the prevalence of KOTs and orthokeratinized odontogenic cyst (OOC) in the western Indian population and to compare demographic and radiographic findings of KOT of our cohort with that of the rest of the world.

Study Design. Relevant publications on KOT were reviewed from the published literature from 1976 to March 2012. Data regarding the 72 additional cases from Mumbai also were analyzed.

Results. Sixty-five reports were identified for this systematic review. The findings were divided into 4 global groups for analysis. The prevalence of KOT was 0.0173% and that of OOC 0.0012%. Male sex, mandible, and the unilocular variation were predominant for the additional cases.

Conclusions. The characterization of the KOT lesions was accomplished, taking into consideration the variations to the western Indian population. (Oral Surg Oral Med Oral Pathol Oral Radiol 2013;115:128-139)

The World Health Organization (WHO) has recently renamed odontogenic keratocyst as keratocystic odontogenic tumor (KOT) and reclassified this lesion in its 2005 edition of histologic classification of odontogenic tumors.[1] KOT is now defined as "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)." Cystic jaw lesions that are lined by orthokeratinized epithelium, therefore, do not form part of spectrum of KOT,[1] which remains a subject of controversy.[2] The orthokeratinized odontogenic cyst (OOC) was first recognized by Wright in 1981[3] and was then considered to be the orthokeratinized variant of odontogenic keratocyst.[2] The first 2 editions of WHO's classification of odontogenic tumors also recognized this orthokeratinized variant.[4] The orthokeratinized variety is now recognized as an entirely different group of lesions termed OOC.[5]

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The ethnic and genetic background of the Indian subcontinent has only once been reported for odontogenic tumors in 2008.[6] However, neither KOT nor OOC was included as a part of that study. A pre-2005 systemic review (SR) on OKC and a post-2005 SR on KOT[7] were done without any input from the Indian subcontinent. This reflects the unreported data from this part of the world. KOTs are associated with inactivation of *PTCH*, the tumor suppressive gene. The involvement of the genetic component suggests possible ethnic variation. It is therefore vital that findings of KOT from this part of the world be reported.

SR is defined as a summary of the medical literature that uses explicit methods to search systematically, appraise critically, and synthesize the world literature on a specific topic.[7]

Current medical databases contain papers on the radiographic findings, treatment, and recurrence rate of OKC and KOT. It is of vital importance to appraise the published material critically and organize it according to its reliability. The rationale of the present study was to identify and critically appraise the evidence currently available in the literature. This SR also attempted to determine the radiographic features of KOT. To obtain

Statement of Clinical Relevance

This study determines the radiographic and clinical features more accurately for the Indian population, which will help in further diagnosis of keratocystic odontogenic tumor.

a better appreciation of the demographic presentation and the intriguing radiographic findings of KOT and OOC, all of the cases of these conditions reported at the Nair Hospital Dental College, Mumbai, over 9 years were analyzed retrospectively.

The aim of present SR was to critically appraise the clinical and radiographic findings of KOT from the available literature. This article also attempts to determine the prevalence of KOT and OOC in the western Indian population and to compare demographic and radiographic findings of an additional 72 KOTs in our cohort with that of the rest of the world.

MATERIALS AND METHODS

Systematic review

An SR for KOT was conducted by searching the medical literature for the period from 1957 to March 2012 using the 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. Search keywords included were keratocystic AND odontogenic AND tumor OR odontogenic AND keratocyst OR primordial. Search was also conducted with the use of MeSH terms "Odontogenic cysts" and "Odontogenic tumors."

Inclusion criteria. All case series, histologically confirmed as a parakeratinized variety, were included in this SR. In the pre-2005 literature, only those case series with histopathologically confirmed parakeratinized variety were included. In the pre-2005 case series where both orthokeratinized and parakeratinized variety were reported, only parakeratinized reports were selected. In case series where both parakeratinized and syndromic KOTs were reported, only the parakeratinised KOTs were retained for analysis. Studies reporting odontogenic tumors including histopathologically diagnosed KOT with details of clinical and radiographic features of KOT were also selected. Studies dealing with the management of KOT and providing details of the clinical and radiographic features were included in this SR. Studies on recurrent KOTs in which clinical and radiographic features of primary KOT was described were also included in this SR.

Exclusion criteria. All case series without histopathologic confirmation of the lesion being a parakeratinized variety were excluded from this review. Studies reporting exclusively mixed orthkeratinized and parakeratinized varieties were excluded from this review. In mixed case series where both parakeratinized and syndromic KOTs were reported, the syndromic cases were excluded. Studies adhering to the pre-2005 nomenclature of KOT and not differentiating the parakeratinized and orthokerationized varieties were excluded from this review. Syndromic cases of NBCCS were excluded from this review, because multiple tumors in this syndrome are often not synchronous. Articles reporting both ortho- and parakeratinized variety, with ambiguity in the description of clinical and radiographic features of the parakeratinized variety, were excluded. KOT lesions associated with any other lesions were excluded. Articles on the peripheral or extraosseous variant of KOT were excluded.

Additional cases

The radiographic and pathologic records of cases diagnosed as KOT and OOC after 2005 and those diagnosed as OKC before 2005 were retrieved from the Oral and Maxillofacial Radiology and Oral Pathology archives of the Nair Hospital Dental College. Cases were reviewed retrospectively by oral radiologists (K.S. and F.K.) for radiographic findings and oral pathologists (S.B. and R.D.) for histopathologic findings. The reviewers all had >5 years of experience in evaluating radiographs or histopathologic slides. Cases were reviewed for a period of 9 years, from January 2001 to December 2009. Patients with adequate histories and relevant radiographic and histopathologic data were included in this study. The radiographical and histopathologic data were independently reviewed retrospectively to eliminate the subjective expectation bias inherent to these kinds of studies. The histopathologic reports and slides were revisited, and the parakeratinized variants were considered to be KOT and the orthokeratinized variety OOC. Lesions reported as mixed (both parakeratinized and orthokeratinized) variety were excluded from this study. In case of disagreement between reviewers, a mutual consensus was obtained by a joint discussion.

In each case, age, sex, and radiographic data were obtained from records, and final diagnosis was made on the basis of histopathology. The various variables considered in this study were age, sex, arch and site of lesion, nature (single/multiple) of occurrence, and radiographic findings. The results obtained from these data were compared with findings from the rest of the world.

The maxillary lesions were grouped into 4 (proposed) categories based on the radiographic extent (Figure 1):

Class 1: lesions limited to the anterior segment of maxilla (distal aspect of right canine to distal aspect of left canine).

Class 2: lesions limited to the posterior segment of maxilla (from mesial aspect of first premolar distally).

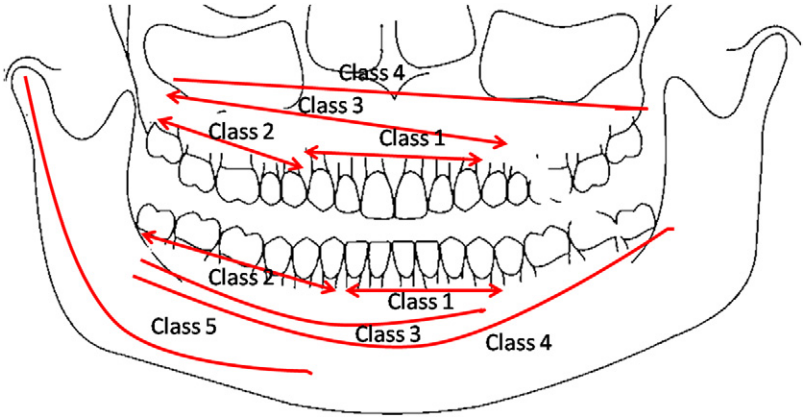

Fig 1. Class categories for maxilla and mandible.

Class 3: lesions that extended into both anterior and posterior segments of the maxilla.

Class 4: lesions from third molar to third molar crossing the midline.

Similarly, the mandibular lesions were grouped into 5 (proposed) categories (Figure 1):

Class 1: lesions limited to the anterior segment of mandible (distal aspect of right canine to distal aspect of left canine).

Class 2: lesions limited to the posterior segment of mandible (mesial aspect of first premolar to distal aspect of third molar).

Class 3: lesions extending into both anterior and posterior segments of mandible.

Class 4: lesions from third molar to third molar crossing the midline.

Class 5: lesions limited to posterior segment, angle of the mandible, ramus, condyle, and coronoid process.

RESULTS

Systematic review

A total of 17,449 articles were found in the various scientific databases with the search expressions relevant to our study. Of these, 134 articles were searched for compatibility with the inclusion and exclusion criteria of the SR. A total of 65 articles[4,5,8-71] were finally selected for the SR. Some of the reports from the post-2005 period did not differentiate the orthokeratinized and the parakeratinized forms of OKC so needed to be rejected. The age range, sex distribution, jaw distribution, clinical features, and radiologic details were extracted from each of the series.

For the convenience of interpretation of the results of SR, the results were mainly divided into 4 major groups based on origin of the genetic family, namely, American, Caucasian, East Asian and Pacific, and African and South Asian. Each group represents a unique genetic family shaped by shared history and geography. These groups are characterized by distinctive patterns of allele frequencies across the short tandem repeat loci.[72] Although all humans are connected by ancient common origins, each of these genetic groupings shares a unique relationship due to more intense and persistent contacts within a geographic area. These groups are therefore not based on racial or ethnic considerations.

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

The American group includes North and South America, the Caucasian group includes Europe, the African and South Asian group includes Africa and the eastern, northern, and southern part of the Indian subcontinent, and the East Asia and Pacific group includes southeastern Asia and the Australian continent. For comparative analysis, the groups of the present SR were compared with the groupings of the previous SR (Table I). The combined American and Caucasian group of the present SR represented the combined Western and Latin American group of the previous SR. The other 2 groups mainly remained the same for both the present and the previous SR. For the ease of comparison, the African and South Asian group and the East Asian and Pacific group of the present SR will be hitherto referred to as the African group and the East Asian group, respectively.

The African group was represented by only 5 reports: 3 from Africa[10,12,51] and 2 from the Indian subcontinent.[63,68] The East Asian group was mainly represented by reports from the East Asian region. There were no reports from the Australian continent. The American and Caucasian groups were fairly well distributed geographically for the reports.

The distribution of reports for the 4 global groups were: 19 reports for the American group, 22 reports for the Caucasian group, 5 reports for the African group, and 19 reports for the East Asian group (Table II). Table II also demonstrates the increased number of reports, the total number of KOTs, and the increased number of cases per year for each group compared with the corresponding group from the previous SR.

The American, Caucasian, and East Asian groups were fairly well represented, and the African group was underrepresented in the number of reports. Despite the highest number of reports coming from the Caucasian group, the numbers of KOTs per year were much higher for the African and East Asian groups. The number of cases seen per year was highest for the African region (8.93 per year) in a period of 62.3 years. Globally, 6.35 new cases of KOT were seen annually (Table II).

Male patients predominated in all of the global groups, including the additional cases and the comparative groups of both the SRs. The mean age of presentation was higher for the American group of the present SR and the African and Subsaharan group from both the present and the previous SR (Table III). Swelling at first presentation was significantly more frequent in the East Asian and Caucasian groups, whereas KOT without swelling was more frequent in the American reports from the present SR (Table III). The American group influenced the figures of the global group as well, with KOT presenting without swelling globally. Pain at first presentation was not a significant finding in any of the global groups except the African group of the present SR and the Subsaharan group of the previous SR (Table III). This however needs to be interpreted with caution, because there was only 1 report representing this feature in both SRs. Mandible was the most common site for the groups, more significantly in the American group (71%) and the Caucasian group (74%), except for the African group of the present SR. Mandible was also commonly affected in the additional cases (Table III). Maxilla was predominant for the African group of the present SR, in contrast to the finding of the previous SR.

The frequent use of NA (information not available) in Table IV demonstrates the paucity of radiologic details noted in the earlier studies. Radiologic details were noted in one East Asian report and in the present study of additional cases.

Unilocular KOT was significantly more predominant than the multilocular counterpart in all of the global groups, including the additional cases, except for the African group in the present SR and the Subsaharan group in the previous SR (Table V).

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.

Table III. Various clinical features of cases included in the present and previous systematic reviews (SRs) in relation to global groups (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*
Male:female	517:379 (9)	352:212 (9)	869:591 (18)	795:559 (15)	272:210 (4)	3:3 (2)	1,104:706 (13)	1,068:681 (9)	2,245:1,507 (35)	1,866:1,243 (26)	54:18 (1)
Mean age of presentation, y	39.1 (6)	32.93 (7)	35.93 (13)	36.75 (10)	39.2 (3)	43.15 (2)	34.25 (12)	35.62 (8)	36.58 (28)	37.81 (20)	30.70 (1)
Swelling, P:A	178:405 (5)	50:24 (1)	228:429 (6)	97:83 (4)	3:0 (1)	3:0 (1)	216:144 (3)	216:143 (3)	447:568 (10)	316:226 (8)	ING
Pain, P:A	81:323 (2)	11:63 (1)	98:386 (3)	24:134 (3)	3:0 (1)	3:0 (1)	132:227 (3)	119:170 (2)	227:613 (7)	146:304 (6)	ING
Mandible:maxilla	670:265 (11)	238:84 (7)	908:350 (18)	819:423 (15)	136:298 (4)	6:0 (2)	1,286:374 (10)	1,234:374 (8)	1,286:374 (31)	2,059:797 (25)	47:18 (1)

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

The borders of KOT were significantly well defined in the East Asian group of the present and previous SRs and the additional cases. Margins were poorly defined in the American group of the present SR (Table V). Buccolingual expansion was noted and present in the East Asian group of both the present and the previous SRs. This, however, was represented by only 1 report for both SRs. Buccolingual expansion was also common for the additional cases (Table V). Root resorption was noted and not a dominant feature for all of the groups, but significantly higher in the East Asian group of both SRs (Table V). This, again, was represented by only 1 report in the previous SR and 2 reports in the present SR. Association of KOT with unerupted tooth was noted but not a common feature for any of the groups (Table V).

Additional cases

Prevalence. During the 9-year period of study, a total of 77 cases, including 72 KOTs and 5 OOCs, were identified from a pool of 415,629 patients. This amounted to an average of 8 new KOTs for each year of the study. The prevalence of OOC was 0.0012% and of KOT 0.0173% (Table VI). From here on, only the results of KOT will be discussed.

Demographic data. The cases were in the range of 10-70 years at presentation with an average age of 30.7 years. An increased prevalence of KOT was seen in the 3rd (n = 24) and 2nd (n = 18) decades of life.

Male patients were predominantly affected, with 54 cases; female patients accounted for 18 cases; male-to-female ratio of incidence was 3:1 (Table VII).

Anatomic location. Mandible was more commonly involved, with 47 tumors in mandible and 18 in maxilla (Table VIII). The overall mandibular-to-maxillary ratio of KOT occurrence was 2.61:1. The remaining 7 KOTs were multiple and not considered in the ratio. In the multiple KOTs, mandible and maxilla both were involved in 4 cases and mandible only was involved in 3 cases.

Site involved. According to the classification described above, the KOT results were divided into the following classes. In mandible, 7, 17, 6, 4, and 13 KOTs were class 1, 2, 3, 4, and 5, respectively, with class 2 being most common. In maxilla 3, 13, 2, and 0 KOTs were class 1, 2, 3, and 4, respectively, with class 2 again being most common (Table VIII). The 7 multiple cases were excluded from this classification to avoid overlap of cases.

Radiographic findings. KOTs appeared radiographically as well defined unilocular or multilocular radiolucencies. In 5 cases, radiographic data could not be retrieved, so they were excluded when analyzing radiographic findings. The 7 multiple cases also were ex-

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)[9]	14	12	0	6	6	NA	NA	2	12	NA	NA	NA	NA	NA	NA
Mosadomi (1976)[10]	2	2	NA	NA	2	NA	NA	NA	NA	NA	NA	NA	NA	NA	
Chiang (1982)[14]	15	15	0	10	4	NA	NA	NA	NA	NA	NA	NA	NA	7	7
Nielsen (1986)[20]	21	NA	NA	17	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
Haring (1988)[22]	60	60	0	44	16	27	33	NA	NA	NA	NA	3	35	16	38
Tagesen (1990)[24]	38	38	0	34	4	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
Crowley (1992)[26]	387	256	0	NA	NA	NA	NA	NA	NA	NA	NA	100	109		
Santos (1999)[34]	40	50	0	41	9	NA	NA	NA	NA	NA	NA	NA	NA	10	50
Myoung (2001)[38]	256	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	70	186
Ogunsalo (2007)[51]	3	3	0	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
Yagyuu (2008)[55]	62	62	0	43	19	I NG	NA	NA	NA	NA	NA	NA	NA	NA	NA
MacDonald-Jankowski (2008)[58]	33	33	0	16	17	33	0	27	6	11	0	13	19	20	12
Ba (2010)[60]	274	I NG	NA	184	62	NA	NA	NA	NA	NA	NA	NA	NA	NA	NA
Ariji (2011)[69]	10	I NG	NA	10	0	I NG	NA	NA	NA	NA	NA	2	8	7	3
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.

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 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

cluded from this variable, because our aim was not to project the number of KOT lesions but the number of cases of KOTs. Sixty cases of KOTs were finally evaluated for radiographic findings.

In mandible, 28 KOTs were unilocular and 18 were multilocular; in the maxilla, 10 KOTs were unilocular and 4 were multilocular (Tables III and V). Unilocular variant was thus more common in both mandible and maxilla.

Association with NBCCS. Two KOTs were found to be associated with NBCCS; both were multiple KOT. In one case multiple KOT occurred in the mandible, and in the other case both mandible and maxilla were involved. In our cohort we found 1 case of multiple KOT associated with Marfan syndrome. However, because Marfan syndrome is not known to be associated with KOT, this was considered to be an incidental finding and included in the multiple KOT category. Out of 7 cases of multiple KOTs (all male patients), only 2 were associated with NBCCS.

DISCUSSION

Systematic review

In 1984, Ahlfors et al.[18] suggested that if the OKC was recognized as a true benign cystic epithelial neoplasia,

the question of modified treatment schedules would be raised. Finally in 2005, published reports influenced the WHO to reclassify the lesion as a tumor, mainly because of its parakeratinized variety and tendency to recur.[5] The parakeratinized variety is now identified as KOT, a tumor, and the orthokeratinized variety as an OOC, a cyst. It is well known that the inactivation of *PTCH*, the tumor suppressor gene, is associated with KOT.[54] The presence of a genetic component suggests that the patient's genetic makeup, which is nothing but "family history," may have a significant role to play. Moreover, the significant differences observed between the global groups in earlier studies[9,10,24,59] shows the importance of genetic origin of the KOT patient. Also, the presence of genetic component in KOT cases makes it important to compare data between different groups. It was therefore decided to compare KOT findings of different global groups and the additional 72 cases of our study. The "world data" were divided into 4 groups. These groups are based on genetic divisions, in contrast to the ethnic division in the previous SR.[7] It was believed that these genetic divisions would add a different perspective to the understanding of KOT and aid in promoting future research.

In the previous SR,[7] the Indian subcontinent was not reported, either in the reports included in the SR or in the reports excluded from the SR. This, coupled with the lack of data from the Indian subcontinent on KOT and OOC, initiated this study. To the best of our knowledge, this is the first such compilation of KOT on an Indian population, demonstrating the analysis of various presentations of KOT. There are certain variations and concordance in the present study compared with the earlier reports and studies on KOT. This article attempts to compare the results from the present study with that of the 4 global groups of the present SR.

The figures representing the African group.[10,51,63,68] should be interpreted with caution owing to the paucity of reports from this region. It was expected that the findings of the additional cases would match those from the African and South Asian community because of the genetic belonging and geographic proximity.

The exclusion of OOC and syndromic cases was necessary because they are completely different lesions. The additional exclusion of the mixed cases was desirable because they appear to be intermediate in behavior between KOT and OOC. Isolated case reports were excluded because they would not add much to the clinical or radiographic features of that particular group.

It was decided to analyze articles published both before and from 2005 onward that comply with the current microscopic definition of KOT and strictly distinguish the orthokeratinizing from the parakeratinizing type. Although a number of the pre 2005 articles met this requirement, it was sometimes impossible to extract full clinical and radiographic data on patients with the parakeratinizing type. It was also surprising that even after the introduction of the 2005 KOT concept, some workers worked with the old 1992 terminology.

The significantly greater number of reports for the East Asian,[14,16,19,28,32,35,37,38,43,49,52,53,55,58,69-71] American,[4,9,11,14,21,22,26,27,31,33,42,45,47,50,64,66] and Caucasian[8,13,15,17,18,20,23-25,29,30,36,39,40,41,44,52-54,56,57] groups may reflect their earlier commencement of record keeping, regular dental and radiographic examinations, and increased awareness. The paucity of reports from the African and South Asian[10,12,51,63,68] groups depicts poor awareness and record keeping. The East Asian and African and South Asian groups presented with significantly higher number of cases reported per year. There was a significant difference between the number of cases reported in the African group of the present SR and the Subsaharan group of the previous SR. This could be attributed to the addition of a few of reports from the Indian subcontinent to this group, with a large number of cases added by a Sri Lankan report.[68] The higher number of KOTs per year in the African and East Asian groups indicates a higher incidence of KOT in these groups, indicating the need to increase awareness.

Although the mean age of presentation was higher for the American group of the present SR, a combined analysis of the American and Caucasian groups of the present SR did not reflect a higher mean age of presentation. This was also in agreement with the finding of the combined Western and Latin American group of the previous SR. This brings forth the point that despite increased awareness for the American group, the mean age of presentation for this group is higher. This was in contrast to the poor awareness for the other groups and lower mean age of presentation. It could therefore be inferred that KOT for the groups other than the American may be occurring at a much earlier age than what is reflected in the Table III.

The incidence of pain and the multilocular nature of the KOT were significantly more common for the African group of the present SR and the Subsaharan group of the previous SR. This, however, needs to be interpreted with caution, because there was only 1 report representing these features in both SRs.

Maxilla predominated over mandible in the African group of the present SR. This was because of the addition of the reports from the Indian subcontinent. Though mandible predominated in the Indian report, maxilla dominated the Sri Lankan report, adding a large number of cases to that group.

Table IX. Statistical analysis (χ^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.

The East Asian,[32,38,58] Caucasian,[48] and African[10] groups presented with significantly more cases of swelling. The American[9,22,26,42,45] KOT did not commonly present with a swelling. This may be because East Asians and the Africans are probably more likely to present with a swelling due to the increased size of the lesion because of poor awareness. The American KOT was probably diagnosed earlier before it increased in size. It needs to be emphasized that size of the KOT was not a feature for the articles included for these groups.

Although swelling was common in the Caucasian group, the combined American and Caucasian group did not dominate with swelling. This could be attributed to a large number of nonswelling cases added by the American group. The corresponding Western and Latin American group of the previous SR demonstrated a marginally increased number of KOTs with swelling.

Buccolingual expansion was also found to be significantly higher in the East Asian reports[14,38,55,58] of both SRs. The combined American and Caucasian group of the present SR and the combined Western and Latin American group of the previous SR did not present with buccolingual expansion. There was no information available regarding this feature for the other groups. Theoretically, KOTs are known to demonstrate minimal buccolingual expansion except in a large-sized KOT.[73] It could therefore be inferred that some communities are prone to buccolingual expansion in KOT.

Philipsen recently stated that root resorption was a rarity in KOT.[6] The East Asian[51] and American[22] groups of the present SR and the additional cases from the present study reported root resorption in 40%, 8%, and 31% of cases, respectively. A significantly higher number of KOTs with root resorption were present in the East Asian group of both the present and the previous SR. Therefore, although root resorption is not common, it may not be rare in certain populations.

Both buccolingual expansion and root resorption were significantly more common in the East Asian group. The reader needs to be cautioned, however, because buccolingual expansion was represented by 1 report and root resorption by 2 reports.

The significant association of the American group[22] and additional cases with unerupted tooth was surprising, particularly because of the high incidence of prophylactic removal of impacted teeth in the American group and the low incidence of prophylactic removal of impacted teeth in the additional cases cohort.

Findings of border definition for the combined American and Caucasian group were consistent with the Western and Latin American group of the previous SR.

The frequency of NA in Table IV indicates that the SR-included case series reported few radiologic features beyond the fact that KOT is a radiolucency that presents with a unilocularity or multilocularity.

Additional cases

The percentage of OOC in the Indian community was found to be 6.5%. This was lower than 11% for the Western group but in concurrence with 7% for Subsaharan and 8% each for the Latin American and East Asian groups of the previous SR.[7]

The percentage of KOT in the additional cases was 93.5%. This again was in concurrence with the Subsaharan (93%), Latin American (92%), and East Asian (92%) groups but more than the Western group (89%) from the previous SR.[7]

Male patients were predominantly affected by KOT compared with female patients in the present study (Table III). The male-to-female ratio of the additional cases showed statistically significant difference from the other global groups of this SR[9,10,14,17,21-22,25,26,37,38,42,45,47-53,62-65,68-71] (Table IX). The Indian male patient is at threefold risk to be affected by KOT compared with the Indian female patient. For the rest of the world groups, the male risk element ranges from 1.3 to 1.6 times that of the female. It could

therefore be inferred that the Indian male is at a higher risk for KOT than his global counterparts.

The 3rd decade was predominant for both male and female patients in the additional cases (Table VII). The percentage of cases presenting in the 3rd decade was 33% in our study group. This was in contrast to the female predominance in the 1st decade and the male predominance in the 3rd decade for the earlier studies.[9,10,14,20,25,32,38,45,47,53,58]

Mandible was involved twice as many times as maxilla in the additional cases. Seven multiple lesions were exempted from this count to avoid case overlap and misleading to more number of KOTs than the number of patients. However among multiple KOTs, mandible slightly predominated over maxilla. This data was not available for the rest of the world hence comparative analysis could not be established.

Mandibular predominance was more or less in agreement with that of the rest of the world data except the African group[10,51,63,68] (Table III). The mandible-to-maxilla ratio of the additional cases showed statistically significant difference from the African group of the present SR (Table IX). This could be attributed to the limited number of reports for this parameter and a report from Sri Lanka with maxillary predominance adding 285 cases to the group.[68]

To establish the common site affected in either jaws, it was attempted to divide the mandible into 5 classes and maxilla into 4 classes (Figure 1). The most common class involved for both maxilla and mandible was class 2, which involved area extending from the distal surface of canine to the distal surface of the second/third molar, followed by class 5 (combination of posterior segment, angle, ramus, and condyle) and class 1 (anterior segment; Table VIII), with class 4 (extending from right third molar to left third molar) being a rare site. This could be because the lesion could have been incidentally picked up early by either the clinician or the oral radiologist during routine examinations. The other possibility could be that the patient may be reporting early to the clinician before it extends enough to the posterior and anterior segment bilaterally.

An attempt was also made to retrieve the unilocular or multilocular nature of the lesion. Excluding 7 multiple cases and 5 cases in which radiographic data could not be retrieved, the radiographic nature could be analyzed in 60 cases. Unilocular KOT was found to be more common in maxilla as well as mandible. The unilocular-to-multilocular ratio of the additional cases showed statistically significant difference in the Caucasian and African groups of the present SR[10,51,20,24] (Table IX). This was because of the paucity of reports from the African group[10,51] and unilocular variant being >6 times more common in the Caucasian group[20,24] compared with 1.2-2.9 times for the other groups, including the additional cases.

In our population, unilocular variant (63.3%) was more dominant than multilocular variant, which is in concurrence with the American[9,22,34,26] (unilocular 74.5%), and Caucasian[20,24] (unilocular 86.4%), and East Asian[14,38,55,58] (unilocular 54.7%) groups, but in contrast to the African group[10] (unilocular 0).

The East Asian group[58] and the additional cases had a good marginal definition for the KOT in contrast to the poorly defined borders of the American KOT.[22] Information for the other groups was not available for this parameter (Table IV). The well-to-poorly defined KOT ratio of the additional cases therefore showed statistically significant difference in the American group[22] of the present SR (Table IX).

Information on root resorption for the African[51] and Caucasian[20,24] groups was not available (Table V). Root resorption, though not a prominent feature for the other groups, was statistically more common in the American group than in the additional cases (Table IX).

CONCLUSION

The present SR concludes that KOT presents as a painless lesion. Swelling could be an occasional finding in certain populations. Mandible remains the most common site for KOT. Buccolingual expansion could be expected in the East Asian community. The unilocular variety is more predominant. Root resorption may be an occasional feature in the East Asian community. It could also be concluded that radiographic features were the most neglected part of KOT in earlier studies.

This retrospective study of 72 KOTs and 5 OOCs attempts to reveal findings of KOT as seen in the Indian diaspora. The Indian male was at a higher risk for KOT. The 3rd decade predominance in the additional cases was in concurrence with earlier findings. For the additional cases, both male and female patients predominated in the 3rd decade as against female predominance in the 1st decade and male in the 3rd decade in earlier studies. Mandibular and unilocular predominance was in compliance with the earlier studies.

A comparative analysis of our findings with the rest of the world groups (Tables III and V) reveals that the overall KOT features for the additional cases are in concurrence with their global counterparts.

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