SYSTEMATIC REVIEW

Orthokeratinized odontogenic cyst: a systematic review

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Objectives: The aims of the review were to evaluate the principal clinical and conventional radiographic features of orthokeratinized odontogenic cyst (OOC) by systematic review (SR), and to compare the frequency of OOC between four global groups.

Methods: The databases searched were the PubMed interface of MEDLINE and LILACS. Only those reports of OOCs that occurred in a consecutive series of OOCs in the reporting authors' caseload were considered.

Results: 37 reports on 36 case series were included in the SR. OOC affected males twice as frequently and the mandible almost 2.5 times as frequently. Although the mean age at first presentation was 35 years, the largest proportion of cases first presented in the third decade for the Western, East Asian and Latin American global groups. Seven reports included details of at least one clinical finding. 11 reported case series included at least 1 radiological feature. All OOCs were radiolucent, 93% were unilocular and 68% were associated with unerupted teeth. 28% of the reported case series included follow up. 4% of OOCs recurred and all of these were in the Western global group.

Conclusions: Although one feature of OOCs is that they are unlikely to recur, some do. Not only is there a lack of long-term follow up of large series with long-term outcomes of OOC, but there is a paucity of clinical and radiological details of OOC at initial presentation.

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Introduction

The orthokeratinized odontogenic cyst (OOC)¹ was first clearly identified as an orthokeratinized variant of the odontogenic keratocyst by Wright in 1981² owing to its different histopathology and reduced likelihood to recur. Although both the first two editions of the World Health Organization's (WHO) histological classification of odontogenic tumours recognized that "cases with orthokeratosis are seen"³ the WHO's 2005 edition⁴ expressly excluded it from its definition of a KCOT. The 2005 edition reclassified the parakeratotic type as a keratocystic odontogenic tumour (KCOT) and stated "Cystic jaw lesions that are lined by orthokeratinizing epithelium do not form part of the spectrum of a ...KCOT."⁴ To fully define the OOC's representation in the literature a systematic review (SR) is necessary. Although Blanas et al⁵ have performed an SR on the

odontogenic keratocyst, all their SR-included reports did not distinguish between KCOT and OOC.

Sackett et al⁶ defined an SR as a summary of the medical literature that uses explicit methods to search systematically, appraise critically and synthesize the world literature on a specific issue. This means that unlike a traditional review the SR, like any other form of primary research, will have a Materials and Methods, and a Results section.⁷

The SR has generally been applied to treatment and drug trials (most recently to osseointegrated implants),⁸ but has also become a powerful tool when adapted to clinical and radiological presentations of important oral and maxillofacial lesions, such as ameloblastomas,^{9,10} KCOT,¹¹ myxomas,¹² dentigerous cysts,¹³ fibrous dysplasia,¹⁴ ossifying fibroma,¹⁵ idiopathic osteosclerosis (IOS, dense bone island),¹⁶ florid osseous dysplasia (FOD),¹⁷ focal osseous dysplasia (FocOD),¹⁸ giant cell granuloma,¹⁹ cleidocranial dysostosis²⁰ and temporomandibular joint disorders.^{21,22} Such SRs recently contributed to a diagnostic algorithm (refer to MacDonald-Jankowski²³ Figure 12).

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Aims and research question

The principal aims were to include as many reports or pertinent parts of the reports as possible and to evaluate the principal clinical and conventional radiographic features of OOC by SR.

The primary research question for this SR was "Do clinical and radiological features improve diagnosis of OOC compared with an orthokeratotic histopathology?" This follows the four-part format required for the SR's research question as set out by MacDonald-Jankowski and Dozier.⁷ To include as many reports as possible a wide search of the literature was made, including non-English reports. In addition to a medical subject heading (MeSH) search, the literature search included free-text searches by hand-searching of journals, which are the natural destinations for reports on oral and maxillofacial lesions, and by reference-harvesting of reports identified by database searches and hand-searching.

SR-included reports were divided into four global groups: Western, sub-Saharan African, East Asian and Latin American, broadly reflecting ethnic origin.

Materials and methods*Systematic review*

The approach follows the SR procedure set out in earlier SRs for other oral and maxillofacial lesions.⁹⁻²⁰ The research question is described above; the search strategy, strategy for sifting the literature and interpretation of the data retrieved are set out below.

The search strategy

The search is based on the research question. The databases searched were the PubMed interface of MEDLINE (National Library of Medicine) and LILACS (Literatura Latino Americana e do Caribe em Ciências da Saúde) by BIREME (Latin American and Caribbean Center on Health Sciences Information). LILACS provides access to important Latin American publications largely not indexed by MEDLINE and has been discussed in the SR on FocOD.¹⁸

The sole MeSH is "odontogenic cyst". Its entry terms are "keratocyst" and "keratocysts". The free-text terms used were "orthokeratinised odontogenic cyst" or "orthokeratinized odontogenic cyst". The terms used to search LILACS were "keratocyst", "orthokeratinised odontogenic cyst" and "orthokeratinized odontogenic cyst". To include as many reports as possible the emphasis was placed on recall rather than precision. Bearing in mind that both the MeSH for dentistry and radiology are generally inadequate and that free-text searching may not identify relevant articles if the term sought had not been included in the title or abstract. This strategy was further augmented by reference to the bibliographies (or citation lists) of all reports identified by the databases (reference-harvesting) or

hand-searching of journals listed in Table 1 of the SR on FocOD.¹⁸ Both database searches and hand-searches were last conducted on 2 September 2009.

The decision to include a report was generally made by reading the title and the abstract. As abstracts were infrequently used before 1979 it was anticipated that a call for the full paper, to determine whether it should be considered by the selection criteria, would be made more frequently for reports published before that year.

Strategy for sifting the literature

Selection criteria: There were two inclusion (criteria 1 and 2) and three exclusion (criteria A to C) criteria for the SR. Each report passed through these criteria in strict sequence. Although a report may be excluded by more than one criterion, only the first criterion to exclude a particular report is tabulated in the Appendix. For the sake of brevity only those reports that cannot be readily included by reference to their title or abstract will be discussed and cited.

Information included in the SR was generally reduced to numbers and tabulated. This took into account the number of cases that passed the selection criteria for inclusion and therefore may differ from the numbers available in the original report. Information not given (ING) was used whenever information on a particular feature could not be determined (either expressed or implied) from the original text. Inadequate information given (IIG) was applied to features in a partially included report, which had been compromised and could not be included in the SR. IIG was also entered against features that the reporters had not adequately quantified.

As the aim was to not only include as many reports as possible, but also as many features of those reports as possible and to include as many cases of each report as the selection criteria would allow, this required a separate strategy. Although the terms "exclusion", "deletion" and "deduction" are normally synonyms, for the purposes of this SR they are specifically defined as follows: "Exclusion" refers to the non-inclusion of a report in the SR and has been used regardless of whether it is in relation to either inclusion or exclusion selection criteria. "Deletion" refers to the removal of cases within a report which are not consistent with one or more selection criteria and have been reported in sufficient detail to permit their identification and removal from the report, allowing the rest of the report to be included in the SR. "Deduction" is applied to those reports where "deletion" is not possible and the number of non-deletable cases exceeds 10% of the report and therefore exceeds the less than 10% allowance for such non-conforming and non-deletable cases. For "deduction" to be permitted the report must include, for at least one feature, wholly inclusive words such as "all" or "every" qualifying "patient" or "case". The use of "almost all", "every" or "the overwhelming majority" (for example, "...of the reported lesions were

Table 1 Orthokeratinized odontogenic cyst: systematic review - analysis of the included reports

First author (year) [Language of publication] [database]	Nationality and/or ethnic origin	Period covered	N ^o of KCOT (N ^o per year)	Gender		Age: mean (range) in years	Presenting signs and symptoms			Site				Comments and percentage of OOC in a series of keratocysts
				Males	Females	Pre-presenting duration mean (range) in years	Swelling	Pain	Other	Mandible		Maxilla		Follow-up and recurrence
										Ant.	Post.	Ant.	Post.	
Borello ((1976) ²⁵ [Spanish] [MEDLINE: Me1, T ₁]	Argentinian	1957–1976 20 years	3 (0.1)	3	0	37.2 SD 4.9 ING	3	0	1 infection (trismus)	4*		0		*2 cysts in 1 patient; 17.6% FU mean 9.3 (2–13) years; 0 recur 11.8% FU IIG; 0 recur 7.7% FU ING; recur ING 18.8% FU 6 years; 1 recur
Brannon (1977) ^{26,27} [English] [MEDLINE: Me1, T ₁]	American	1950–1972 22 years	30 (1.4) ²⁻⁶ 0 (0) ²⁷	ING		ING ING	ING	ING	ING	ING		ING		*2 cysts in patient; 13.3% FU mean 9.3 (2–13) years; 0 recur 11.8% recur IIG 4.5% FU ING; recur ING 22.1% FU IIG; recur IIG 4.5% FU ING; 0 recur 1.5% FU ING; recur ING 14.3% FU ING; IIG recur
Cohen 1980) ²⁸ [English] [MEDLINE: Me1, T ₁]	South African	ING	6	ING		ING ING	ING	ING	ING	ING		ING		
Anniko (1981) ²⁹ [English] [MEDLINE: Me1, T ₁]	Swedish	ING	3	IIG		IIG ING	IIG	IIG	IIG	ING		ING		
Wright (1981) ² [English] [MEDLINE: Me1, T ₁ ,T3]	American	1950–1980 30 years	60*(4.0) [ING, E]	45	14	35.4 (15–73) ING	11	13	25 incid 5 infect 13 0	4 (38) [†]	30	10	10	
Chiang (1982) ³⁰ [English] [MEDLINE: Me1, T ₁]	Taiwanese	1959–1979 20 years	2 (0.1)	1	1	44.5 SD 9.2 ING	ING	ING	ING	0	1	1	0	
Balercia (1983) ³¹ [Italian] [MEDLINE: Me1, T ₁]	Italian	1977–1983 7 years	1 (0.1)	ING		ING ING	ING	ING	ING	ING		ING		
Geng (1983) ³² [Chinese] [MEDLINE: Me1, T ₁]	Chinese	1962–1980 19 years	25 (1.3)	IIG		IIG ING	IIG	IIG	IIG	IIG		IIG		
Ahlfors (1984) ³³ [English] [MEDLINE: Me1, T ₁]	Swedish	1972–1982 11 years	11 (1.0)	IIG		IIG ING	ING	ING	ING	IIG		IIG		
Blondeau (1986) ³⁴ [French] [MEDLINE: Me1, T ₁]	Canadian	9 years	1 (0.1)	IIG		IIG ING	ING	ING	ING	IIG		IIG		
Chen (1986) ³⁵ [English] [MEDLINE: Me1, T ₁]	Taiwanese	1979–1983 5 years	2 (0.4)	IIG		IIG ING	ING	ING	ING	IIG		IIG		
Siar (1988) ³⁶ [English] [MEDLINE: Me1, T ₁]	Malaysian (5C;3M;1I)	1967–1986 20 years	9 (0.4)	7	2	37.2 SD 18.0 1.3 SD 2.1(7)	8	2	2 infect 1 0	0 (5) [†]	3	1 (4)	0	

Table 1 Continued

First author (year) [Language of publication] [database]	Nationality and/or ethnic origin	Period covered	N ^o of KCOT (N ^o per year)	Gender		Age: mean (range) in years	Presenting signs and symptoms			Site				Comments and percentage of OOC in a series of keratocysts
				Males	Females	Pre-presenting duration mean (range) in years	Swelling	Pain	Other	Mandible	Maxilla	Follow-up and recurrence		
										Ant.	Post.		Ant.	Post.
Kakarantza-Angelopoulou (1990) ³⁷ [English] [MEDLINE: Me1, T ₁]	Greek	1974–1986 13 years	2 (0.2)	IIG		IIG IIG	ING	ING	ING	IIG		IIG		3.4% FU ING; recur ING
Brøndum (1991) ³⁸ [English] [MEDLINE: Me1, T ₁]	Danish	1971–1983 13 years	5 (0.4)	3	2	IIG ING	ING	ING	ING	IIG		IIG		15.6% FU ING; 0 recur
Crowley (1992) ³⁹ [English] [MEDLINE: Me1, T ₁ ,T3]	American (44W; 6B)	1970–1989 20 years	55 (2.8)	30	23	33.8 ING	IIG	IIG	IIG	IIG		IIG		12.4% 45 FU ING; 1 recur
Vuhahula (1992) ⁴⁰ [English] [MEDLINE: Me1, T ₁ , T3]	Japanese	ING	12	7	5	25(5–41) ING	1	2	9 incid	8		4		12 FU >4 year; 0 recur
Berrone (1994) ⁴¹ [Italian] [MEDLINE:Me1, T ₁]	Italian	1980–1991 12 years	9 (0.8)	IIG		ING ING	ING	ING	ING	IIG		IIG		26.4% FU ING; recur ING
Anand (1995) ⁴² [English] [MEDLINE: Me1, T ₁ T3]	American	1977–1993 17 years	21 (1.2)	ING		ING ING	ING	ING	ING	ING		ING		36.2% FU 14; 5 recur
Ong (1995) ⁴³ [English] [Reference-harvesting]	Malaysian	1976–1992 17 years	3 (0.2)	IIG		IIG IIG	IIG	IIG	IIG	IIG		IIG		9.2% FU ING; recur ING
El-Hajj (1996) ⁴⁴ [English] [MEDLINE: Me1, T ₁]	Swedish	1974–1993 20 years	3 (0.2)	IIG		IIG ING	ING	ING	ING	IIG		IIG		6.8% FU IIG; recur IIG
Filho (1997) ⁴⁵ [Portuguese] [LILACS]	Brazilian	1960–1994 35 years	15 (0.4)	IIG		IIG ING	ING	ING	ING	IIG		IIG		24.1% FU IIG; 1 recur
Chow (1998) ⁴⁶ [English] [MEDLINE: Me1, T ₁]	Singaporean (56C; 6M; 6I;2other)	1981–1996 16 years	6 (3.7)	ING		IIG ING	ING	ING	ING	ING		ING		7.9% FU ING; recur ING
Li (1998) ⁴⁷ [English] [MEDLINE]	Chinese	1985–1995 10 years	6 (0.6)	6	0	29.8 SD 12.1 ING	10 5 5 incid (Combined enteries for both Chinese and Japanese)			0	6	0	0	5 FU 7.0 SD 2.8 years; 0 recur
Li (1998) ⁴⁷ [English] [MEDLINE]	Japanese	1980–1997 17 years	9 (0.5)	4	5	32.7 SD 10.0 ING		1	7	0	1	9 FU 8.2 SD 2.5 years; 0 recur		
Santos (1999) ⁴⁸ [Portuguese] [LILACS]	Brazilian	1977–1998 22 years	8 (0.4)	IIG		IIG ING	ING	ING	ING	IIG(3) [†]	IIG	IIG(5)	IIG	14.5% FU ING; recur ING

Table 1 Continued

First author (year) [Language of publication] [database]	Nationality and/or ethnic origin	Period covered	N ^o of KCOT (N ^o per year)	Gender		Age: mean (range) in years	Presenting signs and symptoms			Site				Comments and percentage of OOC in a series of keratocysts
				Males	Females	Pre-presenting duration mean (range) in years	Swelling	Pain	Other	Mandible		Maxilla		
										Ant.	Post.	Ant.	Post.	
Bolbaran (2000) ^{49,50} [English] [MEDLINE]	Chilean	1975–1996 22 years	12* (0.5)	IIG		IIG IIG	ING	ING	ING	IIG		IIG		*1 associated with GS FU IIG; 1 recur 2.4% FU ING; recur ING
Myoung (2001) ⁵¹ [English] [MEDLINE: Me1, T ₁]	Korean	1980–1998 19 years	5 (0.3)	ING		IIG ING	ING	ING	ING	ING		ING		*from abstract only as full-paper was unavailable 3.6% FU IIG; IIG recur
da Silva (2001) ⁵² [Portuguese] [LILACS]	Brazilian*	1995–1999 5 years	6 (0.8)	IIG*		IIG* IIG*	IIG*	IIG*	IIG*	IIG*		IIG*		7.9% FU ING; 1 recur
Stoelinga (2001) ⁵³ [English] [MEDLINE: Me1, T ₁]	Dutch	1973–1998 26 years	6* (0.2)	IIG		IIG ING	ING	IIG	ING	0	6	0	0	20.0% FU IIG; 0 recur
Sortino (2002) ⁵⁴ [English] [MEDLINE: Me1, T ₁]	Italian	1998–2000 3 years	3 (1.0)	IIG		IIG ING	ING	ING	ING	IIG		IIG		15 FU (0.5–15); 0 recur
Li (2003) ⁵⁵ [Chinese] [MEDLINE: Me1, T ₁ , T3]	Chinese	1985–2001 17 years	20 (1.2)	14	6	39.1(13–68) ING	IIG	2	ING	18		2		7.8% Irreg FU; 1 recur
Chirapathomsakul (2006) ⁵⁶ [English] [MEDLINE: Me1, T ₁]	Thai	1988–2003 16 years	4 (0.2)	IIG		IIG ING	IIG	IIG	IIG	IIG		IIG		15.5% FU; IIG; recur ING
Meningaud (2006) ⁵⁷ [English] [MEDLINE: Me1, T ₁]	French	1995–2005 10 years	16 (1.6)	IIG		IIG ING	IIG	IIG	IIG	IIG		IIG		* 1 jaw not given; 4.1% 82% FU 0.7 SD 1.2(0–120) years; recur ING
Grossmann (2007) ⁵⁸ [English] [MEDLINE: Me1, T ₁]	Brazilian	1953–2003 51 years	9 (0.2)	5	4	>20 to <60 ING	ING	ING	ING	3*		5*		6.4% FU ING; 0 recur
González-Alva (2008) ⁵⁹ [English] [MEDLINE: Me1, T ₁]	Japanese	1978–2006 28 years	11 (0.4)	ING		ING ING	ING	ING	ING	ING		ING		13.2% 4 FU; 0 recur
MacDonald-Jankowski ⁶⁰ (2009) [English]	Hong Kong Chinese	1989–2005 16 years	5 (0.3)	2	3	34.2 SD 14.5 0.11 SD 0.09	4	2	2 infect	0	4	0	1	

E, exclude; FU, follow-up; I, include; IIG, inadequate information given; Infect, infection; ING, information not given; incid, incidental finding; KCOT, keratocystic odontogenic tumour
* in table refers to comments column; values in parentheses refer to the total number for each jaw which is greater than the sum of anterior and posterior sextant for each jaw. †the number in
brackets represents the total number of cases presenting in that jaw for that report, which exceed the sum of reports affecting the anterior and posterior segments alone

well-defined") were construed to be over 90% (or less than the 10% allowance) and thus also considered to approximate to 100% and were considered to be wholly inclusive. On the other hand studies reporting that "most" or "majority" of the cases were well-defined would not be used, because these and similar phrases would be construed to represent 51% to 89% and, therefore, well outside the 10% allowance. The strategy for minimizing the impact of non-conforming reports on the SR is exclusion, deletion and deduction (including the less than 10% allowance rule).

Inclusion criteria: Criterion 1: Consistency with the WHO classification. The lesions had to be consistent with the histopathology established by either of the first two editions of the WHO classification of odontogenic tumours, which recognized at least the orthokeratotic type of odontogenic keratocyst³. To include only data pertaining to OOC, reports were excluded if:

- (a) their parakeratotic (or KCOT) cases could not be identified and excluded
- (b) their unidentified parakeratotic (or KCOT) cases exceeded the less than 10% allowance. The whole report was excluded unless there remained at least one feature that clearly refers to only cases of OOC arising within that community (deduction). In such cases the report was included only with regard to that particular feature.

Criterion 2: A complete collection of OOC cases. The study should represent a complete collection of cases of OOC arising within a particular community and occurring in the reporter's caseload. Reports that were merely a selection of cases, such as case reports and those studies which were primarily concerned with specific investigations or a discrete age group, for example, children or a particular jaw, were excluded.

Exclusion criteria: Criterion A: Excludes reports already reported and included in the SR. Prevents double reporting of the same clinical cases, by excluding those reports whose data have already been reported and included in the SR, either by the same or different authors, unless the degree of overlap did not exceed 50% and there was at least one statistically different feature between them.

Criterion B: Excludes cases that recurred after primary treatment performed elsewhere and/or earlier than the range in years of the study. It excluded reports if:

- (a) their details of the recurrent (recidivist) cases could not be identified and deleted
- (b) their unidentified recurrent cases exceed the less than 10% allowance. The whole report was excluded unless there remained at least one feature that clearly refers only to those primary (have not yet been treated) lesions which are presenting for the first time

(deduction). The report was included only with regards to that particular feature.

Criterion C: Excludes referred cases. It reinforces criterion 2 by minimizing dilution of the data arising primarily within a specific community. It therefore excludes reports that include referred cases from outside that community, because they may possess unusual features that may skew the profile of OOC within that community and would, in turn, skew the SR. In order to include only data pertaining to the jaws, it excluded reports if:

- (a) their referred cases could not be identified and excluded
- (b) their unidentified referred cases exceeded the less than the 10% allowance. The whole report was excluded unless there remained at least one feature that clearly refers only to those cases arising within that community (deduction). The report was included only with regards to that particular feature.

Interpretation of the literature retrieved

Definition of parameters: Definitions of the parameter, such as the number of years a report covered, the number of OOCs per year, the division of each jaw into sextants and the radiologically apparent boundaries between the basal and alveolar processes for each jaw, are the same as those in the recent SR on FocOD,¹⁸ including the parameter for a well-defined margin established by Slootweg and Muller.²⁴

The term radiolucency could be implied from reference to the radiological shape of the lesions as unilocular or multilocular. The global groups have already been defined and discussed in the SR on KCOT.¹¹

Statistical analysis

Significant differences in frequencies were tested by the χ^2 test with $P < 0.05$. Significant differences in age were tested by a Student's *t*-test with $P < 0.05$.

Results

Systematic review

Many of the reports were automatically rejected because they were single case reports or review articles. Figure 1 outlines the process and disposal of reports considered for a call of the full paper.

Selection criteria: The 36 SR-included reports^{2,25-59} are set out in Tables 1 - 7, 97 reports (two of the excluded reports included two reports counted as one) were excluded under specific exclusion. The report by MacDonald-Jankowski and Li was already known to the present author/reviewer who was one of its co-authors and therefore excluded from Figure 1 and Table 2. The SR-excluded reports of the present SR are almost identical to those excluded from the SR on

The MEDLINE search was as follows -

- #1 Search "odontogenic cyst" [MeSH-Human] [Me1] 4146
- #2 Search "odontogenic keratocyst" [freetext-Human] [T1] 4372
- #3 Search "orthokeratinised odontogenic cyst" [Text-Word][T2] 6
- #4 Search "orthokeratinized odontogenic cyst" [Text-Word][T3] 23

#5 Search (#1) or (#2) or (#3) or (#4) → 4373 titles → 155 full papers → 120 called-up & reviewed 1

The LILACS search was as follows -

- "odontogenic cyst" 113
- "odontogenic cysts" 169
- "orthokeratinised odontogenic cyst" 2
- "orthokeratinized odontogenic cyst" 23
- Total → full paper called-up & reviewed → 8

HANDSEARCHING of Journals (see Table 1 of Reference 12) → 0

1 → 128 → REFERENCE-133 → SELECTION → 36* SR-included reports of 35* case series
HVESTING CRITERIA

SOURCES of	SR-included reports	SR-excluded reports
MEDLINE	32#	89
LILACS	3	5
Handsearching	0	0
Reference Harvesting	1	3

* does not include Reference 60, which the present author was a co-author
number includes 2 sets of reports each reporting the same case series and 1 report reported 2 case series

Figure 1 Orthokeratinized odontogenic cyst: systematic review - search strategy and results

KCOTs and are listed in its Appendix.¹¹ The only difference is the present SR does not include the KCOT SR's references 70 - 72, 89 and 118¹¹ and includes the da Silva et al report⁶¹ excluded under criterion 2.

82% of reports were excluded under criterion 1. There was no significant difference regarding this selection criteria of the reports excluded, published prior to 1990 and those published on or after 1990 ($\chi^2 = 0.92$; 1df: $P > 0.05$).

Analysis of the SR

37 reports^{2,25-60} of 36 communities were included in the SR (Table 1). Li et al⁴⁷ reported on both a Japanese and a Chinese community. Brannon^{26,27} and Bolbaran et al^{49,50} both published two reports reporting one community. One of the reports⁶⁰ was already known to the present author, as a co-author. As it is now apparent that the terms "report" and "case series" do not mean the same thing in all circumstances then when a "case series" rather than a specific "report" is intended then "case series" will be used. The clinical features, decades, age distribution in decades at first presentation and radiological details extracted from each of the series are presented in Tables 1, 3, 4, 5 and 6, respectively. The statistical analyses of the information in Table 1 are displayed in Tables 3 and 4, and that of Table 6 in Table 7.

All four global groups were represented, but only one report represented the sub-Saharan African group (Table 2). Fewer than half of the reports were from the Western group. Reports from the East Asian group included a wide range of nations.

The number of OOCs per year was not possible to determine for three reports. The number of OOCs per year fell from 0.89 (SD 1.20) OOCs per year in the 10 case series reported prior to 1990, to 0.79 (SD 0.90) OOCs per year in the 23 case series reported after and including 1990; this was not significant ($t = 0.24$; 31 df: $P > 0.05$).

Although the mean number of cases ranged widely between the global groups, as shown in Table 3, this was not significant.

There was a higher proportion of males in the three global groups, Western, East Asian and Latin American (Table 4), but there was no significant difference between them.

The mean age at first presentation was slightly lower for the East Asian global group than for Western and Latin American global groups; this was not significant (Table 3). Table 5 is a comparison between the nine reported case series of the distribution according to age in decades. Most of the cases first presented in the third decade. Males predominated in the first decade and every decade from the third onwards, whereas the

Table 2 Orthokeratinized odontogenic cyst: systematic review - comparison between SR-included and SR-excluded reports

Source of reports	MEDLINE	LILACS	Reference-harvesting	Handsearching
SR-included reports/SR-excluded reports	32 of 36 / 89 of 97	3 of 36 / 5 of 97	1 of 36 / 3 of 97	0 of 36 / 0 of 97
Language of publication	English	Other European	East Asian	
SR-included reports/SR-excluded reports	27 of 36 / 71 of 97	7 of 36 / 22 of 97	2 of 36 / 4 of 97	
Global groups	East Asian	Sub-Saharan	Latin American	Western
SR-included reports/SR-excluded reports	12 of 36 / 18 of 97	1 of 36 / 2 of 97	7 of 36 / 10 of 97	16 of 36 / 67 of 97

Statistical Analysis χ^2 : Significant difference for global group: Western ($\chi^2 = 6.87$; 1df: $0.01 > P < 0.001$) SR-included and excluded reports: East Asian (12 : 18) vs Western (16 : 76): $\chi^2 = 7.51$; 1df: $0.01 > P > 0.001$

Table 3 Orthokeratinized odontogenic cyst (OOC): systematic review - mean number of features per SR-included OOC reports in relation to global groups

Features	Global groups				
	All (number of reports)	East Asian (number of reports)	Sub-Saharan African (number of reports)	Latin American (number of reports)	Western (number of reports)
Number of OOCs	11.22 SD 13.29 (36)	8.57 SD 6.73 (14)	6 (1)	8.83 SD 4.26 (6)	15.07 SD 19.11 (15)
Number of years	17.82 SD 9.35 (33)	16.92 SD 5.34 (13)	ING	25.83 SD 13.17 (6)	15.21 SD 7.60 (14)
Number of OOCs/year	0.82 SD 0.97 (33)	0.75 SD 0.96 (13)	ING	0.40 SD 0.24 (6)	1.07 SD 1.13 (14)
Age at first presentation	34.89 SD 5.28 (10)	34.64 SD 6.38 (7)	ING	37.20 (1)	34.60 SD 1.13 (2)

Statistical analysis χ^2 : All not significant

females slightly predominated in the second decade overall, and for the Western global group. There was no significant gender difference between the East Asian and Western global groups ($\chi^2 = 0.20$; 1df: $P > 0.05$).

Only two reports,^{36,60} both East Asian, indicated the duration of the patients' pre-existing awareness of their lesion prior to first presenting for treatment. The period between first awareness of the lesion and first presentation for its treatment, for the Hong Kong Chinese⁶⁰ case series, was significantly shorter than the Malaysian³⁶ case series published two decades earlier ($t = 2.64$; 11df: $0.05 > P > 0.01$).

A significantly higher proportion of East Asian OOCs first presented with swelling compared with Western OOCs (Table 4). Location of the lesions, by quadrant or sextant, was specified in only 7 case series for the mandible and in 5 for the maxilla, whereas location by jaw was specified in 12 (Table 4). The mandible was affected more than twice as frequently as the maxilla. There were no difference between the groups for either the mandibular or maxillary sextants; all four global groups displayed a predilection for the posterior sextants of the mandible, but only the East Asian and Western global groups reported an equal distribution between maxillary sextants.

Table 6 contains 11 SR-included case series covering 3 global groups; the sub-Saharan African group was not represented. The general paucity of radiological detail is illustrated by the frequent use of ING in Table 6. All OOCs were well defined. Details concerning cortication, expansion and tooth displacement were largely provided by one East Asian report.⁶⁰ 68% of OOCs were associated

with unerupted teeth (Table 6). There was no significant difference between the global groups (Table 7).

Although only 15 case series did not report whether their cases recurred after treatment, only 10 considered recurrence in addition to the period of follow-up (4 East Asian, 2 Latin American and 4 Western reports; this is not significant). The overall recurrence rate was 4%. Recurrence after a period of follow-up was reported only for the Western global group; this was not significant (Table 3). Two early reports^{2,25} reported a patient with two OOCs.

Discussion

Although there were Western case series included in the SR, significantly more were excluded in comparison with the higher proportion of East Asian case series included in the SR. This may simply reflect an increased awareness of OOC by East Asian reporters. The four SR-included reports on five communities,^{35,39,46,53} which focused solely on the OOC, were from the East Asian global group. This degree of interest in the OOC was in marked contrast to Western case series; however, they recorded, proportionally, significantly more OOCs within their keratocyst case series (refer to the SR on KCOTs¹¹). The four East Asian reports^{36,40,47,55} focused on the OOC were intrigued by its different character from the majority of keratocysts.

The marked absence of SR-included reports on OOCs from the sub-Saharan global group was consistent with

Table 4 Orthokeratinized odontogenic cyst (OOC) systematic review - number of features per SR-included OOC reports in relation to global groups

Features	Global Groups				
	All (No. of reports)	East Asian (No. of reports)	Sub-Saharan African (No. of reports)	Latin American (No. of reports)	Western (No. of reports)
Male: female	127:65 (12)	41:22 (7)	ING	8:4 (2)	78:39 (3)
Number of cases: swelling Y:N	37:53 (6)	23:17 (4)	ING	3:0 (1)	11:36 (1)
Number of cases: pain Y:N	26:82 (7)	13:45 (5)	ING	0:3 (1)	13:34 (1)
Number of cases: incidental findings Y:N	39:43 (4)	14:21 (3)	ING	ING	25:22 (1)
Number of cases: infected Y:N	10:53 (4)	4:9 (2)	ING	1:2 (1)	5:42 (1)
Number of jaws: mandible:maxilla	104:43 (12)	50:13 (7)	ING	10:10 (3)	44:20 (2)
Number of sextants: mandible, ant: post	5:57 (7)	1:21 (5)	ING	ING	4:36 (2)
Number of sextants: maxilla, ant: post	12:12 (5)	2:2 (4)	ING	ING	10:10 (1)
Number of recurrent lesions: Y:N	7:158 (9)	0:33 (4)	ING	0:3 (1)	7:122 (4)

Ant:Post, Anterior: Posterior. ING, information not given; Y:N, Yes: No

Statistical analysis χ^2 : Number of swelling: East Asian vs Western ($\chi^2 = 10.64$; 1df: $P < 0.001$)

Table 5 Orthokeratinized odontogenic cyst: systematic review - distribution of cases according to age in decades

Report decade	Borello ²⁵ (L)	Chiang ³⁰ (E)	Sitar ³⁶ (E)	Vuhahula ⁴⁰ (E)	El-Hajj ⁴⁴ (W)	Li Chinese report ⁴⁷ (E)	Li Japanese report ⁴⁷ (E)	Grossman ⁵⁸ (L)	MacDonald ⁶⁰ (E)	Total for each decade	Decade % of males/total			Western (1 report)			East Asian (6 reports)			Latin American (2 reports)		
											each decade	% of males/total	% of males/total	Total for decade	% of males/total	Total for decade	% of males/total	Total for decade	% of males/total	Total for decade	% of males/total	
0-9	0	0	0	1(1:0)	0	0	0	0	0	1(1:0)	0.9	100.0	0	0	0	0	1(1:0)	2.3	100	0	0	
10-19	0	0	3(2:1)	1(0:1)	9(3:6)	1	0	3	1(1:0)	18(6:8)	16.7	42.8	9(3:6)	17.3	33	33	6(3:2)	14.0	60	3	25	
20-29	1(1:0)	0	2(1:1)	7(4:3)	15(11:4)	1	4	3	1(1:0)	34(18:8)	31.5	69.2	15(11:4)	28.8	73	75	15(6:4)	39.9	60	4(1:0)	33.3	
30-39	1(1:0)	1(0:1)	1(1:0)	2(1:1)	4(3:1)	3	4	2	1(0:1)	19(6:4)	18.5	63.6	4(3:1)	7.7	75	75	12(2:3)	27.9	40	3(1:0)	25	
40-49	1(1:0)	1(1:0)	1(1:0)	1(1:0)	9(6:3)	1	0	1	1(0:1)	15(9:4)	13.9	62.2	9(6:3)	17.3	67	67	4(2:1)	9.3	67	2(1:0)	17	
50-59	0	1(1:0)	1(1:0)	0	5(3:2)	0	1	0	1(0:1)	9(5:3)	8.3	62.5	5(3:2)	9.6	60	60	4(2:1)	9.3	67	0	0	
60-69	0	0	0	0	10(6:4)	0	0	0	0	10(6:4)	9.2	60.0	10(6:4)	19.2	60	60	1(1:0)	2.3	100	0	0	
70-79	0	0	1(1:0)	0	0	0	0	0	0	1(1:0)	0.9	100.0	0	0	0	0	0	0	0	0	0	
80-89	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	
90-100	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	0	
Total	3(3:0)	2(1:1)	9 (7:2)	12 (7:5)	52 (32:20)	6	9	9	5(2:3)	107 (52:31)	100	63.1	52 (32:20)	100	62	62	43 (17:14)	100	61	12 (3:0)	100	

The number of males and females are in parentheses
E, East Asian; L, Latin American; W, Western

the SR on KCOT. These findings are a marked contrast to the SR of another odontogenic lesion, the ameloblastoma.^{9,10} This has already been discussed in the SR on KCOTs.¹¹

There was no significant difference between the reports excluded from the SR on the basis of, language of publication or source.

Although Wright² stated that OOCs did not occur in naevoid basal cell carcinoma syndrome, Bolbaran and et al^{49,50} reported one case that did.

In Table 5 only the second decade exhibited a preponderance of females in the overall synthesis. As this decade coincides with the menarche this could suggest that there may be a hormonal element to the occurrence of this lesion in females at this age. On closer examination it can be observed that this phenomenon is largely owing to el Hajj et al's Western report.⁴⁴

The significantly shorter period between first awareness of the lesion and first presentation for treatment among the Hong Kong Chinese⁶⁰ compared with an earlier Malaysian report³⁶ may suggest a greater awareness of the need to treat oral and maxillary lesions in the former community. On the other hand, a recent report indicated that although the Hong Kong Chinese reported a memory of a painful episode affecting the mouth, few actually took action.⁶² The significantly shorter period between first awareness of the lesion and first presentation for its treatment was also noted with regards to KCOT affecting the same community.⁶⁰ The reason for this unexpected phenomenon has already been discussed.⁶⁰

48% of OOC were discovered as incidental findings, 41% first presented with swellings and 24% first presented with pain. These differ not only between global groups, but also within a clearly defined global group, such as the East Asian group. All 5 cases in the Hong Kong Chinese report⁶⁰ presented with symptoms, whereas 75% of the 12 OOC cases in the Vuhahula et al Japanese report⁴⁰ were discovered as incidental findings. This suggests that the Japanese patients presented for reasons other than OOC. Nevertheless, the East Asian global group was significantly associated with swelling, as the primary presenting symptom, even when the Vuhahula et al report⁴⁰ was included. Although this suggests that OOCs affecting the East Asian global group might be more aggressive, it was only the Western group that reported recurrences.

All global groups displayed a predilection for the mandible, particularly the posterior sextants; however, the 68% association with unerupted third molars was not expected, particularly as the largest of such reports was an American report.³⁹ A possible explanation for this is that this report may not have been affected by the routine prophylactic removal of third molars. Vuhahula et al⁴⁰ described most of the OOCs as dentigerous cysts with orthokeratinization. The association with unerupted teeth suggests that many OOCs may have first developed during adolescence, when the third molars were developing, and were only noticed later either owing to the development of symptoms or as an incidental discovery during investigation of another dental problem.

Table 6 Orthokeratinized odontogenic cyst: systematic review - analysis of the radiology in the included reports

Author (year)	No. of cases	Radiological features						Expansion													
		Completely radiolucent?		Shape		Degree of marginal definition		Cortication or sclerosis of the periphery		Buccolingual		Lower border of the mandible displaced and/or eroded		Antral involvement		Tooth displacement		Root resorption		Cyst associated with unerupted tooth?	
		Yes	No	Uni-locular	Multi-locular	Well-defined	Poorly-defined	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No	Yes	No
Borello (1976) ²⁵	4	IIG	IIG	IIG	IIG	IIG	IIG	ING	ING	ING	ING	ING	ING	N/A	N/A	1	1	ING	ING	ING	ING
Wright (1981) ²	60	60	0	59	1	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	IIG	IIG
Chiang (1982) ³⁰	2	2	0	2	0	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	IIG	IIG	1	1
Crowley (1992) ³⁹	55	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	28	9
Vuhahula (1992) ⁴⁰	12	12	0	11	1	12	0	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	9	2
Li (1998) Chinese report ⁴⁷	6	6	0	6	0	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	4	5
Li (1998) Japanese report ⁴⁷	9	9	0	8	1	IIG	IIG	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING		
Santos (1999) ⁴⁸	8	8	0	8	0	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	5	3
Stoelinga (2001) ⁵³	6	6	0	4	2	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	IIG	IIG
Li (2003) ⁵⁵	20	20	0	18	2	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING	ING
MacDonald-Jankowski (2005) ⁶⁰	5	5	0	3	2	5	0	5	0	5	0	1	3	1	0	4	1	0	5	3	2
Total	187	128	0	119	9	17	0	5	0	5	0	1	3	1	0	5	2	0	5	50	22

Table 7 Orthokeratinized odontogenic cyst (OOC): systematic review - number of radiological features per SR-included OOC reports in relation to global groups

Features	All (No. of reports)	Global groups			
		East Asian (No. of reports)	Sub-Saharan African (No. of reports)	Latin American (No. of reports)	Western (No. of reports)
Shape, unilocular:multilocular Y:N	119:9 (9)	48:6 (6)	ING	8:0 (1)	63:3 (2)
Associated unerupted tooth? Y:N	50:23 (6)	17:11 (4)	ING	5:3 (1)	28:9 (1)

Statistical analysis χ^2 : all not significant

Y:N, yes:no; Ant:Post, anterior:posterior; ING, information not given

Table 8 shows the important statistical differences between the present SR and that of the KCOT.¹¹ The KCOT's incidence is more than eight times that of the OOC, reflecting the 10% proportion of OOCs of the formerly known odontogenic keratocyst (refer to the SR on KCOTs¹¹). The lack of difference between the two SRs, regarding the range of years of the reports they included, is not surprising as the majority of SR-included reports for both SRs were the same. The mean age at first presentation of KCOTs was 3 years older than that of OOC; this difference was not significant. Although the OOC is more likely to present in males, this is not significant. 28% of KCOTs recur in contrast to the 4% recurrence rate of OOC. The margins of all cases of the two small case series of OOCs, reported so far, were well-defined, whereas over a third of KCOTs first presented were poorly defined. KCOT first presented with swelling significantly more frequently than OOCs,

but OOCs were significantly more associated with unerupted teeth.

Conclusions

1. Reports derived from East Asian communities were included significantly more frequently. The language of publication and source had no significant effect on the inclusion or exclusion of a report.
2. Many recent reports have not recognized that the formerly orthokeratotic type of the odontogenic keratocyst is a completely separate lesion.
3. Three of the four global groups were well represented. The exception was the sub-Saharan African global group. This is likely to reflect the low incidence of this lesion in this global group
4. Although males predominated in all global groups and for most decades, females predominated in the second decade; this may be associated with the menarche.

Table 8 Comparison between keratocystic odontogenic tumour (KCOT) and orthokeratinized odontogenic cyst (OOC): systematic review - number of radiological features per SR-included reports in relation to global groups

Statistical analysis: student t-test			
	KCOT	OOC	Statistics
Number of lesions	98.86 SD 131.47 (49)	11.22 SD 13.29 (36)	$t = 4.53$; 83 df: $P < 0.001$
Number of years	16.64 SD 11.12 (45)	17.82 SD 9.35 (33)	$t = 5.90$; 76 df: $P > 0.05$
Number of lesions/year	5.84 SD 5.57 (45)	0.82 SD 0.98 (33)	$t = 5.90$; 78 df: $P < 0.001$
Age at first presentation	37.81 SD 6.33 (20)	34.82 SD 5.30 (10)	$t = 1.36$; 53 df: $P > 0.05$
Statistical analysis: χ^2			
	KCOT	OOC	Statistics
Male:female	1866:1243 (26)	127:65 (12)	$\chi^2 = 2.79$; 1df: $P > 0.05$
Number of cases: swelling Y:N	316:226 (8)	37:53 (6)	$\chi^2 = 9.30$; 1df: $0.01 > P > 0.001$
Number of cases: pain Y:N	146: 304 (8)	26:82 (7)	$\chi^2 = 2.86$; 1df: $P > 0.05$
Number of cases: incidental findings Y:N	129: 486 (9)	39:43 (4)	$\chi^2 = 27.83$; 1df: $P < 0.001$
Number of cases: infection Y:N	55: 395 (6)	10:53 (4)	$\chi^2 = 0.65$; 1df: $P > 0.05$
Number of jaws: mandible:maxilla	2059: 797 (25)	104:43 (12)	$\chi^2 = 0.12$; 1df: $P > 0.05$
Number of sextants: mandible Ant:Post	166: 1214 (16)	5:57 (7)	$\chi^2 = 0.92$; 1df: $P > 0.05$
Number of sextants: maxilla Ant:Post	126: 275 (12)	12:12 (5)	$\chi^2 = 3.55$; 1df: $P > 0.05$
Number of recurrent lesions Y:N	276: 705 (15)	7:158 (9)	$\chi^2 = 18.77$; 1df: $P < 0.001$
Number of syndromic:non-syndromic cases	199: 2925 (27)	1:11 (1)	$\chi^2 = 0.05$; 1df: $P > 0.05$
Shape: Unilocular: Multilocular	211:81 (9)	119:9 (9)	$\chi^2 = 0.49$; 1df: $P > 0.05$
Degree of margin definition: Y:N	60:33 (2)	17: 0 (2)	$\chi^2 = 42.61$; 1df: $P < 0.001$
Cortication: Y:N	36:24 (2)	5:0 (1)	$\chi^2 = 3.03$; 1df: $P > 0.05$
Buccolingual expansion: Y:N	29:18 (2)	5:0 (1)	$\chi^2 = 2.73$; 1df: $P > 0.05$
Tooth displacement: Y:N	22:10 (1)	4:3 (2)	$\chi^2 = 0.46$; 1df: $P > 0.05$
Root resorption: Y:N	16.54 (7)	0:5 (1)	$\chi^2 = 1.51$; 1df: $P > 0.05$
Associated unerupted tooth: Y:N	239:435 (7)	50:22 (6)	$\chi^2 = 42.72$; 1df: $P < 0.001$

Ant:Post, anterior:posterior; Y:N, Yes:No

Figures in parentheses indicate the number of reports

5. Swelling was a significant presenting symptom in the East Asian global group.
6. OOCs displayed few differences between the global groups this may simply be due to the paucity of details as indicated by the INGs and IIGs in the tables.
7. It is clear from the frequency of ING, particularly in Table 6, that the 11 case series out of the 36 SR-included case series reported little radiology beyond the fact that the OOC was a radiolucency and presented with a unilocular or multilocular shape.

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