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REVIEW ARTICLE

The presentation of odontogenic keratocysts in the jaws with an emphasis on the tooth-bearing area: a systematic review and meta-analysis

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Abstract

Purpose This study was conducted in order to gain insight in the actual ratio of odontogenic keratocysts occurring in the tooth-bearing area as compared to the posterior region of the jaws in order to come up with reliable data to base upon a rational treatment policy.

Methods We searched MEDLINE, Web of Science, Scopus, and Cochrane databases for studies reporting on the location of mandibular and maxillary odontogenic keratocysts. All records were independently assessed and a meta-analysis was performed. Risk difference with a confidence interval of 95% of having the lesion in the posterior region versus the tooth-bearing area was the effect measure. P value for the summary effect of < 0.05 was considered statistically significant.

Results The 2615 records retrieved were reduced to 34 studies to be qualitatively/quantitatively assessed. The pooled values showed that the difference in the clinical risk of having keratocysts in the posterior region of the mandible and in the tooth-bearing area of the maxilla is 21 and 43%, respectively ($P < 0.02$ and $P < 0.00001$).

Conclusions A substantial amount of keratocysts occur in the tooth-bearing area of the jaws, requiring attention.

Keywords Odontogenic keratocyst · Keratocystic odontogenic tumor · Prevalence · Systematic review · Meta-analysis

Introduction

It is well known that odontogenic keratocysts (OKCs) are often occurring in the posterior mandible/ascending ramus, and also in the maxilla, they tend to be located in the molar tuberosity area. In fact, the vast majority of cysts in the posterior mandible are OKCs, while sporadically one may encounter a cystic ameloblastoma or a regular odontogenic (dentigerous) cyst in that area. This has consequences for the surgical approach as outlined by Chapelle et al. [1]. They suggested an algorithm for the treatment of cystic lesions of the jaws, which entailed that all cysts in the posterior mandibular area should be treated as if they were OKCs or cystic ameloblastomas. In a recent systematic review and meta-analysis, Al-Moraissi et al. [2] have proposed to treat OKCs according to a defined protocol as suggested by Stoelinga [3]. This includes excision of the overlying attached mucosa, enucleation of the cyst, and treatment of the bony defect with Carnoy's solution. This would be entirely possible for cysts in the posterior mandible (third molar area and extending into the ascending ramus) and even posterior maxilla, but it is different with cysts in the tooth-bearing area because they are usually not easily diagnosed as such. They often present as lateral periodontal or (lateral) dentigerous cysts not arising any suspicion. The diagnosis is usually made after enucleation not followed by any additional treatment. In a prospective study of Stoelinga [3], 24 of 75 OKCs were located in the tooth-bearing area of which five recurred between 1 and 10 years, whereas only four recurrences (less than 10%) were seen of the 51 OKCs located in the posterior mandible or maxilla.

For reasons mentioned above, it would be advisable to have some insight in the actual ratio of OKCs occurring in the tooth-bearing area as compared to the posterior area of the

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jaws. Some solid data on the presentation of OKCs in both jaws would be helpful to formulate a treatment policy for those OKCs occurring in the tooth-bearing areas. It is the intention of this systematic review and meta-analysis to come up with reliable data to base upon a rational treatment policy.

Material and methods

Protocol and registration

The present review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses—PRISMA protocol [4] and was registered in the International Prospective Register of Systematic Reviews—PROSPERO (CRD42018092088).

Focused question

We intended to answer the following focused question: "What's the overall ratio of OKCs that occur in the posterior region versus the dentate area of the mandible and maxilla?"

Eligibility criteria

Original randomized/nonrandomized clinical trials and observational studies written in English were considered in the present systematic review (https://prsinfo.clinicaltrials.gov/definitions.html [Accessibility verified June 11, 2018]). Records that fulfilled the following items were included: (a) human patients with primary OKC histologically diagnosed [5] and (b) location of the OKCs in the posterior region and/or in the dentate area of the mandible and/or maxilla. According to our protocol, the posterior region was considered to be the mandibular ramus/angle and maxillary tuberosity, including those OKCs in the third molar area. On the other hand, the dentate area was considered to be the tooth-bearing area/alveolar process, including those OKCs intimately related with teeth (i.e., incisors, canines, premolars and first/second molars).

Records presenting data from (a) recurrent OKCs; (b) OKCs associated with the nevoid basal cell carcinoma syndrome; (c) cystic lesions that are lined only by orthokeratotic epithelium; (d) histological and molecular studies; and (e) in vitro and animal experiments, literature reviews, systematic reviews, case reports, and studies with less than ten participants were not considered.

Studies that met the inclusion criteria or those with doubtful information either in the title or abstract were selected for full-text assessment in a second round of this review. Reasons for rejection of studies were recorded for each report.

Search strategy

The first hit was conducted online by two independent reviewers (YSS and MN), in MEDLINE (via PubMed), Web of Science, Scopus, Cochrane Library, and Google Scholar (other source), from the inception until January 31st of 2018. Publications were searched using the following strategy: (((((((odontogenic keratocyst) OR keratocystic odontogenic tumor) OR keratocystic odontogenic tumour) OR primordial cyst)) AND (((odontogenic cysts) OR jaw cysts) OR developmental odontogenic cysts)) AND ((((diagnosis) OR incidence) OR prevalence) OR demographics)) NOT case report. Duplicate records were subsequently removed.

Study selection

Records that remained from the first hit were independently selected by reading their title and abstract (first round). Disagreements in this selection were resolved by mutual discussion. Afterwards, all records screened from the first round had their full text independently assessed for eligibility by the same reviewers.

Data collection process

The reviewers YSS and MN separately submitted all eligible studies to a qualitative synthesis using an extraction data form including mainly country and total period of the study, clinical characteristics of the patients, and number/location of the primary OKCs. The mandible and maxilla were considered separately. Whenever the article mentioned only "posterior or anterior region," without a precise description of the cysts location, we considered these areas as being the same as of our protocol, as stated in the "Eligibility criteria" section.

Subsequently, all extraction data forms with the results of each included study were verified together in order to calibrate validity and reliability of this process.

Risk of bias in individual studies

To assess the quality of the studies, we adapted a few checklists from the Joanna Briggs Institute (http://joannabriggs.org/research/critical-appraisal-tools.html [Accessibility verified June 11, 2018]) and applied them to be used in Review Manager Software 5.3 (Review Manager (RevMan) [Computer program]. Version 5.3. Copenhagen: The Nordic Cochrane Centre, The Cochrane Collaboration, 2014).

The queries of the included studies are briefly explained as follows: (a) cases from the same country; (b) validity of data collection (i.e., valid clinical and

radiographic assessment or valid retrospective analysis); (c) evaluation of confounding factors such as the inclusion of OKCs based on histological criteria other than that recommended by the World Health Organization [5] and the presence of recurrent OKCs, syndromic OKCs, and orthokeratocysts in the study sample; and (d) strategies to deal with confounding factors.

We also assessed the risk of bias of the OKCs occupying the posterior maxilla, and large cysts may have occurred concomitantly in the posterior region and molar area (dentate area).

Summary measures and synthesis of results

Meta-analysis was performed in the Review Manager software 5.3, and the risk difference (RD) between the proportions of individuals having primary OKCs (event) in the posterior region versus in the tooth-bearing area with a confidence interval (CI) of 95% was the effect measure. Inverse variance method was applied in a random-effect model. *P* value, from the *Z* test, for the meta-analysis summary effect of < 0.05 was considered to provide evidence to the effect estimates. The heterogeneity among studies was obtained from the chi-squared test.

Results

Study selection and included studies

The first hit retrieved 2615 records from the traditional databases and 38 articles from another source. The distribution of the searched records and the number of studies finally included [3, 6–38] are shown in the flow diagram (Fig. 1).

Excluded studies

The studies that were not included in the present systematic were excluded for several reasons:

- No specification if the OKC was located in the mandible or maxilla [39–80]
- No specification if the OKC was located in the dentate area or in the posterior region of the jaws [39–44, 51, 60, 63, 78, 81–135]
- Impossibility to distinguish nonsyndromic from syndromic OKCs [46, 60, 63, 69, 76, 80, 81, 83, 89, 92, 99, 100, 102–106, 111, 119, 126, 129, 136–154]
- Impossibility to distinguish OKCs from orthokeratocysts [83, 99, 100, 103, 106, 119, 153, 155–160]
- Impossibility to distinguish primary OKCs from recurrent OKCs [48, 66, 81, 88, 89, 99, 103, 105, 107, 119, 126, 129, 137, 142, 153–155, 159, 161, 162]
- Less than ten OKCs in the study [44, 54, 55, 59, 61, 64, 68, 72, 75, 93, 148, 163–174]
- No specification on the quantity of OKCs among the total number of odontogenic cysts/tumors [1, 157, 175–197]
- Selective outcome bias (the posterior region or the dentate area may have been one of the exclusion criteria or OKCs in the posterior region were counted together with lesions in the dentate area) [86, 89, 95, 99, 108, 110, 113–115, 120, 125, 130, 132, 151, 173, 188, 192, 193, 198–214]

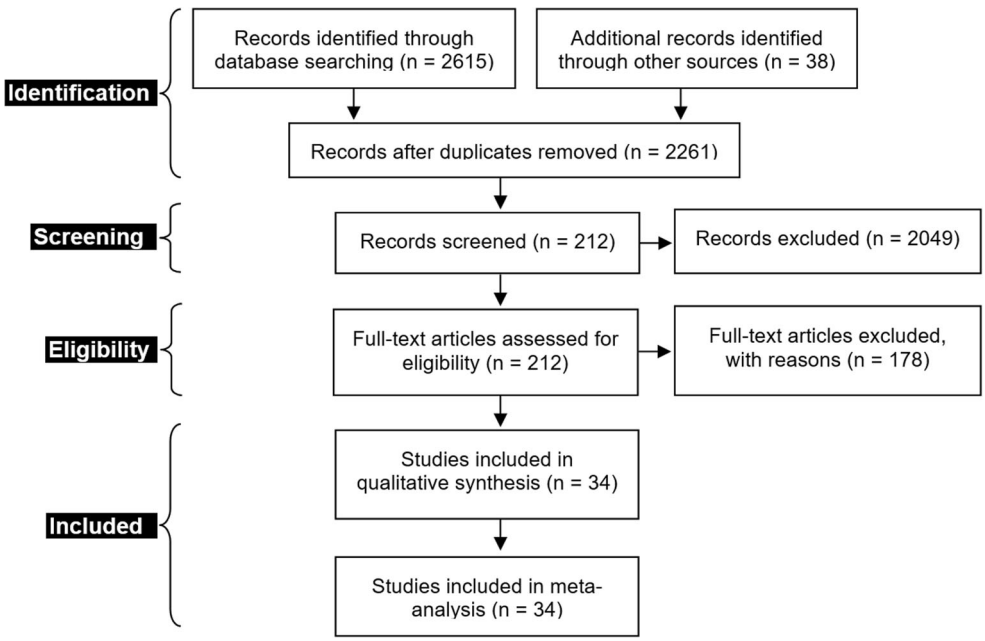

Fig. 1 Flow diagram

Study characteristics, risk of bias across studies, and synthesis of results of the included studies

Thirty-four articles met the criteria as set for the meta-analysis [3, 6–38].

Four studies were nonrandomized clinical trials [3, 6–8]. Thirty studies were retrospective observational case–controls [9–38]. The characteristics of the studies are shown in Table 1. The risk of bias across studies [215] is expressed in Fig. 2.

Mandibular OKCs

Out of the 2376 mandibular OKCs, 1401 were located in the posterior region (59%) and the remaining 975 OKCs were located in the tooth-bearing area (41%). The RD of 0.21 [0.03, 0.38] of the pooled values pointed that the difference in the clinical risk of having the OKC between the posterior region and tooth-bearing area is 21%, the posterior region being more susceptible to develop the lesion. In other words, patients have an estimated difference of 21% in the probability to have OKCs in the posterior region of the mandible. The CI and the *P* value of < 0.02 showed that there is strong evidence to support this statement (Table 1 and Fig. 3).

Maxillary OKCs

In the maxilla, however, 566 out of the 750 OKCs were located in the tooth-bearing area (75.5%) and the remaining 184 OKCs were located in the posterior region (24.5%). The RD of 0.43 [0.30, 0.55] of the pooled values pointed that the difference in the clinical risk of having the OKC between the posterior region and tooth-bearing area is 43%, the tooth-bearing area being more susceptible to develop the lesion. In other words, patients have an estimated difference of 43% in the probability to have OKCs in the tooth-bearing area of the maxilla. The CI and the *P* value of < 0.00001 showed that there is strong evidence to support this statement (Table 1 and Fig. 4).

Discussion

Thirty-four articles met the criteria as set for the qualitative synthesis and all of them were included in the meta-analysis [3, 6–38]. Forty-four percent of these studies did not base their histological diagnosis on the criteria as defined by the *WHO Atlas of Head and Neck Tumours 2005* [3, 6, 8–10, 13–15, 18, 28–32, 34] and 15% of the studies did not report at all on what histological criteria they based their findings [11, 23, 33, 36, 38]. Regarding the detection of confounding factors, that may have introduced bias and influenced the results expressed in the meta-analysis, in 64 to 73% of the included studies, no mention was made of the presence of recurrent [8, 10–13, 15–17, 20–22, 26–30, 32–34, 36–38] and/or syndromic OKCs [6, 9–11, 14–17, 20–22, 24, 25, 27, 29–34, 37, 38] nor the presence of orthokeratotic cysts [6, 8–11, 13, 14, 16, 17, 20–29, 32–34, 36–38]. Only 11% of the studies presented sufficient details to avoid these biases [3, 7, 19, 35] (Fig. 2). Furthermore, a considerable heterogeneity was detected among studies reporting mandibular and maxillary OKCs ($I^2 = 98\%$) due to sparse overlap of confidence intervals for the results of individual studies (Figs. 3 and 4). It is a consequence of the biological diversity of the included patients in the present study, in terms of ethnicity, age, gender, etc., that the occurrence of OKCs in either jaw is unpredictable. Therefore, we incorporated the random-effect model in the meta-analysis considering that the effects estimated were not identical, but followed some distribution [216].

Yet, the results of the present systematic review unequivocally point to a higher prevalence of OKCs for the mandible as compared to the maxilla, the ratio being about 3:1. Six studies evaluated only mandibular OKCs [6, 10, 28, 30, 31, 36].

The OKCs in the mandible did occur more often in the third molar area/ascending ramus as compared to the tooth-bearing area anterior of the wisdom tooth, the ratio being approximately 3:2. Considering the large number of pooled data, this was highly statistically relevant.

The results in the maxilla are seemingly quite different in that the presentation in the posterior maxilla is far less than that in the tooth-bearing area. In fact, there is no nontooth-bearing area of the maxilla that is exposed to the oral cavity, apart from the tuberosity. The posterior area literally would be the molar area and beyond. It is obvious that the anatomical borders in the maxilla are not as clearly distinguishable as those in the mandible. Cysts appearing in the bicuspid/molar area often extend toward the tuberosity or vice versa. Many authors did not even mention the tuberosity area as a separate location of an OKC. When they mentioned the location to be in the molar area, we counted it as being in the tooth-bearing area (dentate area), even though the cyst may have been extended to the tuberosity. The increased risk of having OKCs in the dentate area of the maxilla reflects that phenomenon (Fig. 5). In other words, our results with regard to the presentation in the maxilla are not reliable with regard to the site of origin of the OKC.

Since 30 out of the 34 included studies were retrospective observational, we should assume that in many occasions the diagnosis of OKC came after the cyst had been enucleated, leaving the patient and the surgeon with a

Table 1 Characteristics of included studies

Authors[a]	Mandible			Maxilla		
	Posterior[b]	X	Dentate[c]	Posterior	X	Dentate
Ali and Baughman, 2003		Anterior—14 Canine—41 Premolar—41 1st and 2nd molar—33			Anterior—14 Canine—41 Premolar—41 1st and 2nd molar—33	
	137	X	129	30	X	102
Baitaneh and Qudah, 1998		1st and 2nd molar—8		N/A		
	23	X	8			
Bande et al., 2010		Symphysis—3 Body—5			Anterior—2	
	38	X	8	2	X	2
Berge et al., 2016		Unclear			Unclear	
	60	X	7	15	X	10
Boffano et al., 2010		Incisor/canine—13 Premolar—42 Molar—59			Incisor/canine—16 Premolar—18 Molar—36	
	77	X	114	0	X	70
Chuong et al., 1982[d]		Unclear			Unclear	
	12	X	4	4	X	0
de Souza et al., 2010[e]		Unclear			Unclear	
	40	X	5	2	X	7
Deepthi et al., 2016		Anterior—13			Anterior—6	
	44	X	13	11	X	6
El-Gehani et al., 2009		Unclear			Incisor/canine—6	
	28	X	7	11	X	6
el-Hajj and Anneroth, 1996[f]		Symphysis—5 Body—6			Upper front—3 Premolar/molar—1	
	14	X	11	4	X	4
González-Alva et al., 2008[g]		Incisor—8 Premolar—8 Molar—75			Incisor—4 Premolar—1 Molar—9	
	14	X	91	0	X	14
Irvine and Bowerman, 1985[h]		Symphysis—1 Body—3 (1 bilateral)		N/A		
	8	X	5			
Jing et al., 2007[i]		Anterior—52 Premolar—58 Molar—107			Anterior—23 Premolar—39 Molar—45	
	253	X	217	0	X	107
Kambalimath et al., 2014		Unclear			Unclear	
	6	X	3	2	X	1
Khosravi et al., 2013[j]		Unclear			Unclear	
	188	X	35	54	X	57
Leung et al., 2016		Anterior—4			Anterior—2	
	79	X	4	20	X	2
MacDonald et al., 2013[k]		Unclear			Incisors to 1st premolar—2	
	7	X	0	1	X	2
Madras and Lapointe, 2008[l]		Anterior—3 Anterior/body—1 Premolar—4 Body—1			Canine and sinus—1	
	2	X	9	4	X	1

Table 1 (continued)

Authors[a]	Mandible			Maxilla		
	Posterior[b]	X	Dentate[c]	Posterior	X	Dentate
Mamabolo et al., 2011[m]		Anterior—4 Canine/premolar—6 Premolar—1 Canine/premolar/molar—1			Anterior—8 Premolar—3 Molar—2	
	1	X	12	0	X	13
Manzoor et al., 2015		Unclear			Unclear	
	49	X	18	8	X	7
McIvor, 1972[n]		Unclear		N/A		
	23	X	14			
Nakamura et al., 1995		Incisor—4 Incisor/premolar—5 Incisor/molar—1 Premolar—6 Premolar/molar—2 Molar—24			Incisor—7 Incisor/premolar—4 Incisor/molar—5 Premolar—3 Premolar/molar—2 Molar—13	
	25	X	42	1	X	34
Ngeow et al., 2000[o]		Anterior—4		N/A		
	26	X	4			
Ong and Siar, 1995[p]		Body—8 Body across midline—4		N/A		
	10	X	12			
Park and Kim, 1985[q]		Cross symphysis—8 Premolar—2 Molar—1			Anterior teeth—2 Premolar—3 Molar—2 Antrum—9	
	10	X	11	0	X	16
Peker et al., 2016		Anterior—5 Premolar—5 Molar—38			Anterior—12 Premolar—3 Molar—12	
	32	X	48	0	X	27
Ribeiro Junior et al., 2012[r]		Premolar—1 Molar—2			Anterior—1	
	7	X	3	0	X	1
Sharifian and Khalili, 2011[s]		Anterior—19 Premolars—15 Molar—14			Anterior—29 Premolars—2 Molar—2	
	44	X	48	11	X	33
Simiyu et al., 2013		Anterior—2 Body—8			Anterior—5 Premolar—1 Maxillary sinus—1	
	5	X	10	0	X	7
Stoelinga, 2001[t]		Anterior—6 Body—11			Bicuspid-molar—7 Between canines—7	
	39	X	17	0	X	14
Tabrizi et al., 2014		Symphysis—14 Body—24		N/A		
	53	X	38			
Therkildsen et al., 2014		Premolar—6 Molar—6			Incisor—1 Molar—1	
	2	X	12	0	X	2
Urs et al., 2014		Anterior—2			Anterior—2	
	6	X	2	1	X	2
Vedtofte and Praetorius, 1979		Anterior—6			Anterior—8	

Table 1 (continued)

Authors[a]	Mandible			Maxilla		
	Posterior[b]	X	Dentate[c]	Posterior	X	Dentate
		Body—8			Premolar/molar—11	
	39	X	14	3	X	19
Synthesis of studies	2376 mandibular OKCs			750 maxillary OKCs		
	1401 (59%)	X	975 (41%)	184 (24.5%)	X	566 (75.5%)
	RD—0.21 [0.03, 0.38] $P < 0.02$ $I^2 = 98\%$ ($P < 0.00001$)			RD—0.43 [0.30, 0.55] $P < 0.00001$ $I^2 = 98\%$ ($P < 0.00001$)		

N/A, not applicable or not possible to describe; *RD*, risk difference; *P*, a value for the summary effect of < 0.05 was considered statistically significant; I^2, heterogeneity

[a] In alphabetic order

[b] Mandibular ramus/angle and maxillary tuberosity, inclusive those OKCs involved with the third molars

[c] Alveolar ridge or beyond, inclusive those OKCs involved with teeth

[d] Eight recurrent OKCs of the original report were not accounted [14]

[e] Seven OKCs located in the anterior + posterior region and four cases of unspecified location were not accounted [15]

[f] Twenty-nine OKCs were not considered because there were five syndromic OKCs among this number. "Orthokeratosis alone was found in three of the included OKCs"—high risk of bias for strategies to deal with confounding factors [18]

[g] Eleven OKCs from syndromic patients, 40 OKCs described as "extent location," and 13 OKCs with "multiple location" were not accounted [19]

[h] One patient that had two OKCs, two recurrent, and one giant OKC occupying the right ramus and mandibular body was not accounted [6]

[i] Eleven OKCs with unspecified location were not accounted [20]

[j] Twenty-eight OKCs that occurred simultaneously in the anterior and posterior regions were not accounted [22]

[k] Eighteen recurrent OKCs and one primary that occurred in posterior/anterior region of the mandible were not accounted [24]

[l] Five recurrent OKCs, six that occurred in the mandibular body/ramus, and one unspecified maxillary OKC were not accounted [25]

[m] Nine maxillary and ten mandibular OKCs with unknown location and two located in the molar–ramus area were not accounted [26]

[n] Ten maxillary OKCs were not accounted because the site was unclear [28]

[o] Eighteen maxillary OKCs were not accounted because there was one orthokeratocyst in this sample. One orthokeratocyst, seven OKCs in the anterior/posterior mandible, and two with unknown location were also not accounted [30]

[p] Ten maxillary OKCs with unknown location and three cysts located simultaneously in the mandible/maxilla with doubtful location were not accounted. "There is only one orthokeratinized OKC"—high risk of bias for strategies to deal with confounding factors [31]

[q] Three OKCs with unknown location were not accounted [32]

[r] Eleven syndromic OKCs were not accounted [7]

[s] One hundred three OKCs with multiple unspecified locations were not accounted [34]

[t] Five recurrent OKCs and seven orthokeratocysts were not accounted [3]

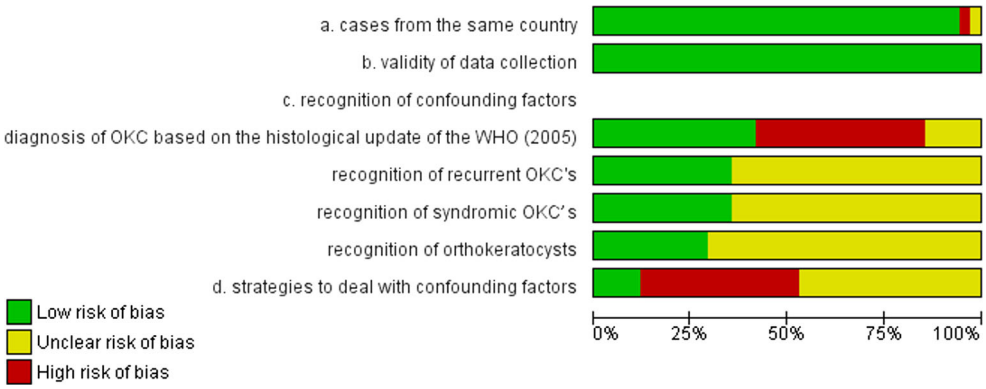

Fig. 2 Risk of bias across studies

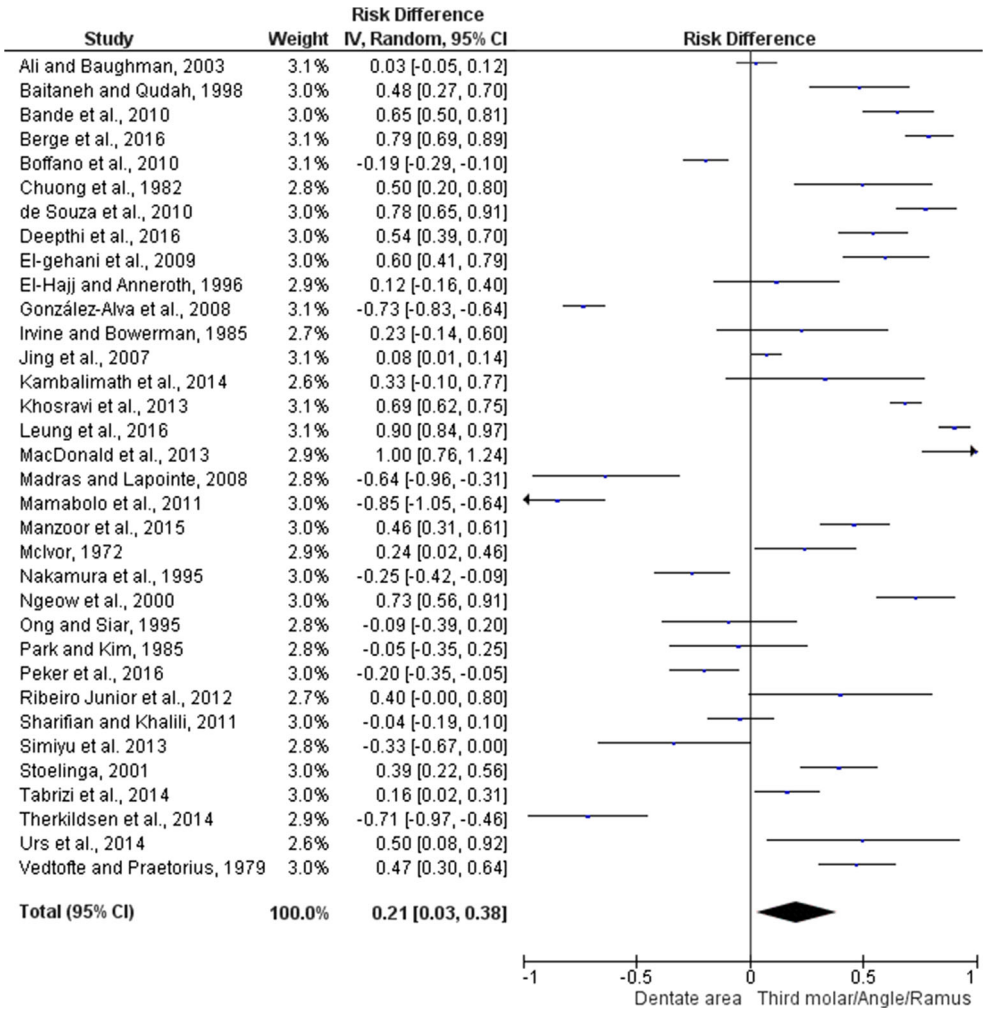

Fig. 3 Meta-analysis of the mandibular OKCs

dilemma so as to what treatment should be followed if any. In most instances, it would have been rather small lesions particularly in the mandible. They probably did not have a clear connection with the covering mucosa, and in case of a lateral periodontal presentation, the periodontium of the neighboring tooth would have been exposed. The latter precludes the use of Carnoy's solution in that area, because of possible irreparable damage to the periodontium of the tooth involved. In these small cysts, a wait-and-see policy might be the best option with a yearly follow-up for the first 5 years and beyond that every 2 years. The patients need to be informed about the chance of recurrence, which may occur over as long a period of 25 years [3]. In case of larger cysts (> 1 cm in diameter), one should consider treatment of the bony cavity with Carnoy's solution at a second intervention, particularly when the cyst has been removed piecemeal.

In case of maxillary cysts, the same applies for the small cysts in the tooth-bearing area as described for the mandible. The larger cysts that extended to the posterior area and most likely have penetrated the maxillary sinus would require a different approach. If the diagnosis is made after primary enucleation, one may choose to open the cavity again and wipe the bone cavity out with Carnoy's solution. It is not advisable to do the same with the thin bony walls of the sinus, because it may cause bone necrosis. If the cyst has reached the tuberosity area, it may pay off to take a wedge out of the soft tissue of the tuberosity as well and have it examined. One of the authors (PS) has seen several clusters of epithelial islands and even microcysts in this area, most likely the source of a newly developing cyst [3]. If the biopsy is positive, extra surveillance is warranted.

Conclusion

In conclusion, a substantial amount of OKCs occur in the tooth-bearing area of the jaws. They simply often come as surprise diagnosis and are, thus, unexpected findings. Yet,

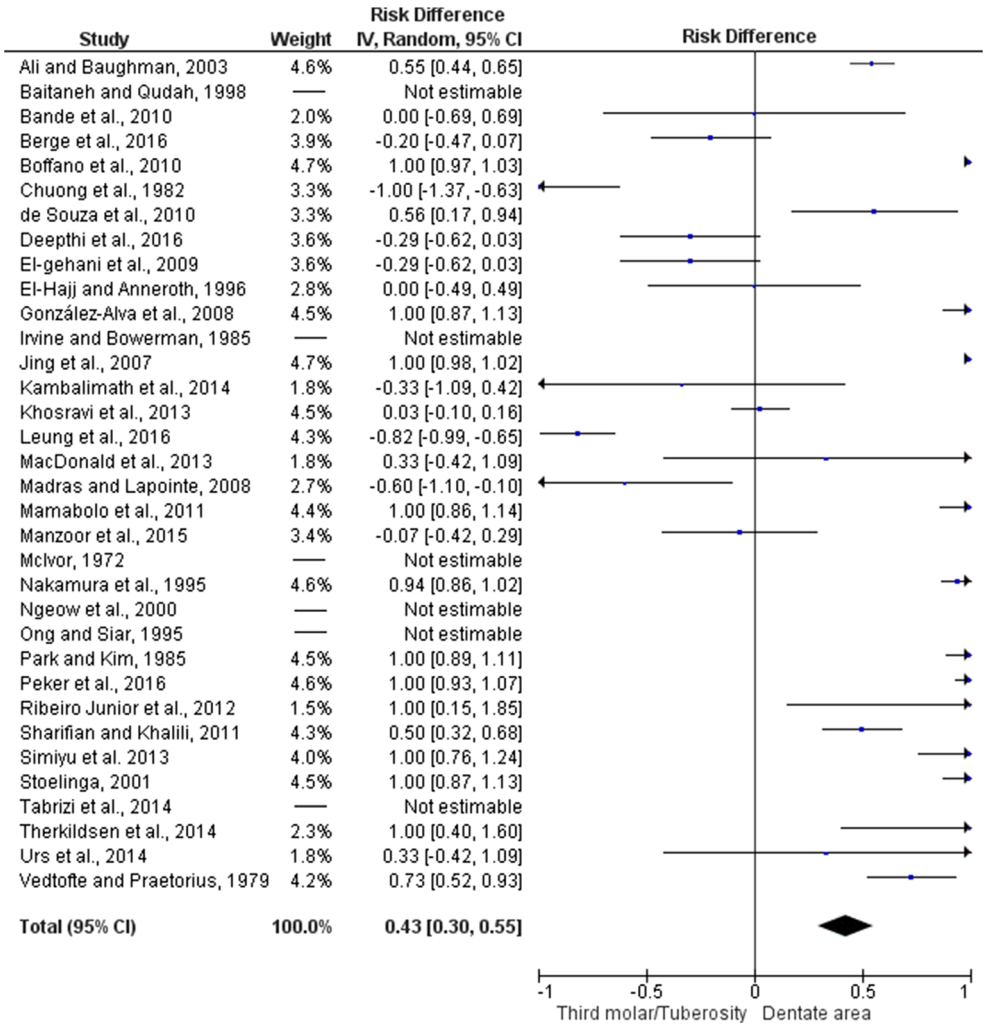

Fig. 4 Meta-analysis of the maxillary OKCs

it does not warrant routine biopsies of every cystic lesion. In fact, a false-negative histopathological diagnosis, particularly when the cyst has been infected, is likely to occur in many instances because inflammation causes the epithelium to lose its characteristic appearance and these changes may be local [3, 217]. There is, thus, a considerable chance of a nonrepresentative biopsy. In general, when cysts in the tooth-bearing area present with signs or symptoms compatible with those of OKCs, such as no bony expansion, scalloped margins, or even a multilobular appearance, an aspiration biopsy is probably the best option. The presence of keratin flakes or a protein level of less than 4 g per 100 ml is indicative for an OKC [116]. Yet, inflammatory changes may also preclude a correct diagnosis when using this method. In case of a positive diagnosis, careful enucleation and, where possible, excision of the attached mucosa should be carried out followed by treatment with Carnoy's solution.

The small OKCs, approximately 1 cm in diameter or less, require a different approach. A wait-and-see policy, with yearly follow-up for the first 5 years and every 2 years thereafter, is strongly advocated. Possible recurrences are easily manageable without much suffering for the patient involved.

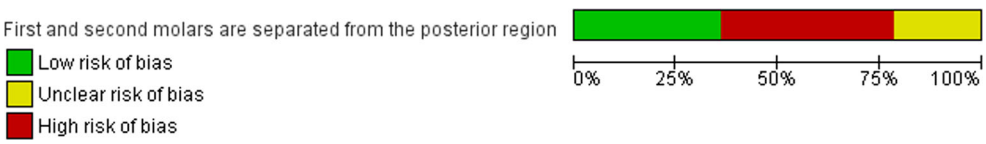

Fig. 5 Risk of bias across studies addressing maxillary OKCs occupying the posterior region

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent For this type of study, formal consent is not required.

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References

1. Chapelle KA, Stoelinga PJ, de Wilde PC, Brouns JJ, Voorsmit RA (2004) Rational approach to diagnosis and treatment of ameloblastomas and odontogenic keratocysts. Br J Oral Maxillofac Surg 42:381–390
2. Al-Moraissi EA, Pogrel MA, Ellis E III (2016) Does the excision of overlying oral mucosa reduce the recurrence rate in the treatment of the keratocystic odontogenic tumor? A systematic review and meta-analysis. J Oral Maxillofac Surg 74:1974–1982
3. Stoelinga PJ (2001) Long-term follow-up on keratocysts treated according to a defined protocol. Int J Oral Maxillofac Surg 30:14–25
4. Moher D, Liberati A, Tetzlaff J, Altman DG (2009) PRISMA group. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. PLoS Med 6:e1000097
5. Philipsen HP (2005) Keratocystic odontogenic tumour. In: Barnes L, Eveson JW, Reichart P, Sidransky (eds) Head and neck tumours WHO classification of tumours. IARC Press, Lyon, pp 306–307
6. Irvine GH, Bowerman JE (1985) Mandibular keratocysts: surgical management. Br J Oral Maxillofac Surg 23:204–209
7. Ribeiro Junior O, Borba AM, Alves C, De Gouveia M, Coracin F, Guimarães Júnior J (2012) Keratocystic odontogenic tumors and Carnoy's solution: results and complications assessment. Oral Dis 18:548–557
8. Vedtofte P, Praetorius F (1979) Recurrence of the odontogenic keratocyst in relation to clinical and histological features. A 20-year follow-up study of 72 patients. Int J Oral Surg 8:412–420
9. Ali M, Baughman RA (2003) Maxillary odontogenic keratocyst—a common and serious clinical misdiagnosis. J Am Dent Assoc 134:877–883
10. Bataineh AB, al Qudah M (1998) Treatment of mandibular odontogenic keratocysts. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 86:42–47
11. Bande CR, Prashant MC, Sumbh B, Pandilwar PK (2010) Prevalence, treatment and recurrence of odontogenic keratocyst in central India. J Maxillofac Oral Surg 9:146–149
12. Berge TI, Helland SB, Sælen A, Øren M, Johannessen AC, Skartveit L, Grung B (2016) Pattern of recurrence of nonsyndromic keratocystic odontogenic tumors. Oral Surg Oral Med Oral Pathol Oral Radiol 122:10–16
13. Boffano P, Ruga E, Gallesio C (2010) Keratocystic odontogenic tumor (odontogenic keratocyst): preliminary retrospective review of epidemiologic, clinical, and radiologic features of 261 lesions from University of Turin. J Oral Maxillofac Surg 68:2994–2999
14. Chuong R, Bruce Donoff R, Guralnick W (1982) The odontogenic keratocyst. J Oral Maxillofac Surg 40:797–802
15. de Souza LB, Gordon-Nunez MA, Nonaka CF, de Medeiros MC, Torres TF, Emiliano GB (2010) Odontogenic cysts: demographic profile in a Brazilian population over a 38-year period. Med Oral Patol Oral Cir Bucal 15:e583–e590
16. Deepthi PV, Beena VT, Padmakumar SK, Rajeev R, Sivakumar R (2016) A study of 1177 odontogenic lesions in a South Kerala population. J Oral Maxillofac Pathol 20:202–207
17. El-Gehani R, Orafi M, Elarbi M, Subhashraj K (2009) Benign tumours of orofacial region at Benghazi, Libya: a study of 405 cases. J Craniomaxillofac Surg 37:370–375
18. el-Hajj G, Anneroth G (1996) Odontogenic keratocysts—a retrospective clinical and histologic study. Int J Oral Maxillofac Surg 25:124–129
19. González-Alva P, Tanaka A, Oku Y, Yoshizawa D, Itoh S, Sakashita H, Ide F, Tajima Y, Kusama K (2008) Keratocystic odontogenic tumor: a retrospective study of 183 cases. J Oral Sci 50:205–212
20. Jing W, Xuan M, Lin Y, Wu L, Liu L, Zheng X, Tang W, Qiao J, Tian W (2007) Odontogenic tumours: a retrospective study of 1642 cases in a Chinese population. Int J Oral Maxillofac Surg 36:20–25
21. Kambalimath DH, Kambalimath HV, Agrawal SM, Singh M, Jain N, Anurag B (2014) Prevalence and distribution of odontogenic cyst in Indian population: a 10 year retrospective study. J Maxillofac Oral Surg 13:10–15
22. Khosravi N, Razavi SM, Kowkabi M, Navabi AA (2013) Demographic distribution of odontogenic cysts in Isfahan (Iran) over a 23-year period (1988-2010). Dent Res J (Isfahan) 10:162–167
23. Leung YY, Lau SL, Tsoi KY, Ma HL, Ng CL (2016) Results of the treatment of keratocystic odontogenic tumours using enucleation and treatment of the residual bony defect with Carnoy's solution. Int J Oral Maxillofac Surg 45:1154–1158
24. MacDonald D, Gu Y, Zhang L, Poh C (2013) Can clinical and radiological features predict recurrence in solitary keratocystic odontogenic tumors? Oral Surg Oral Med Oral Pathol Oral Radiol 115:263–271
25. Madras J, Lapointe H (2008) Keratocystic odontogenic tumour: reclassification of the odontogenic keratocyst from cyst to tumour. J Can Dent Assoc 74:165–165h
26. Mamabolo M, Noffke C, Raubenheimer E (2011) Odontogenic tumours manifesting in the first two decades of life in a rural African population sample: a 26 year retrospective analysis. Dentomaxillofac Radiol 40:331–337
27. Manzoor A, Ahmed W, Umair M, Bukhari GA, Niaz MO (2015) Assesment of recurrence of odontogenic keratocysts treated with different surgical modalities—nine years follow up of disease. Pakistan Oral Dental J 35:408–415
28. McIvor M (1972) The radiological features of odontogenic keratocysts. Br J Oral Surg 10:116–125
29. Nakamura T, Ishida J, Nakano Y, Ishii T, Fukumoto M, Izumi H, Kaneko K (1995) A study of cysts in the oral region. Cysts of the jaw. J Nihon Univ Sch Dent 37:33–40
30. Ngeow WC, Zain RB, Yeo JF, Chai WL (2000) Clinicopathologic study of odontogenic keratocysts in Singapore and Malaysia. J Oral Sci 42:9–14
31. Ong ST, Siar CH (1995) Odontogenic keratocysts in a Malaysian population: clinical, radiological and histological considerations. Ann Dent Univ Malaya 2:9–14
32. Park TW, Kim SR (1985) Clinical and radiographic study of odontogenic keratocyst. Oral Radiol 1:43–53
33. Peker E, Öğütlü F, Karaca I, Gültekin E, Çakir M (2016) A 5 year retrospective study of biopsied jaw lesions with the assessment of concordance between clinical and histopathological diagnoses. J Oral Maxillofac Pathol 20:78–85
34. Sharifian MJ, Khalili M (2011) Odontogenic cysts: a retrospective study of 1227 cases in an Iranian population from 1987 to 2007. J Oral Sci 53:361–367

35. Simiyu BN, Butt F, Dimba EA, Wagaiyu EG, Awange DO, Guthua SW, Slootweg PJ (2013) Keratocystic odontogenic tumours of the jaws and associated pathologies: a 10-year clinicopathologic audit in a referral teaching hospital in Kenya. J Craniomaxillofac Surg 41:230–234
36. Tabrizi R, Omidi M, Dehbozorgi M, Hekmat M (2014) Correlation of radiographic features and treatments with the frequency of recurrence in odontogenic keratocysts of the mandible. J Craniofac Surg 25:e413–e417
37. Urs AB, Arora S, Singh H (2014) Intra-osseous jaw lesions in paediatric patients: a retrospective study. J Clin Diagn Res 8: 216–220
38. Therkildsen NM, Andersen K, Blomlöf J (2014) Diagnostic sensitivity and specificity in a retrospective clinical, radiographic and histopathological study of 166 cystic jaw lesions. Oral Surg 7:33–38
39. Abdolali F, Zoroofi RA, Otake Y, Sato Y (2017) Automated classification of maxillofacial cysts in cone beam CT images using contourlet transformation and spherical harmonics. Comput Methods Prog Biomed 139:197–207
40. Abdullah BH, Jabbar Abdul Qader OA, Mussedi OS (2016) Retrospective analysis of 1286 oral and maxillofacial biopsied lesions of Iraqi children over a 30 years period. Pediatr Dent J 26:16–20
41. Ahmad M, Jenny J, Downie M (2012) Application of cone beam computed tomography in oral and maxillofacial surgery. Aust Dent J 57:82–94
42. Al Sheddi MA (2012) Odontogenic cysts. A clinicopathological study. Saudi Med J 33:304–308
43. Ali K, Munir F, Rehman A, Abbas I, Ahmad N, Akhtar UM (2014) Clinico-radiographic study of odontogenic cysts at a tertiary care centre. J Ayub Med Coll Abbottabad 26:92–94
44. Asaumi JI, Murakami J, Yanagi Y, Hisatomi M, Matsuzaki H, Maki Y, Unetsubo T, Honda Y, Konouchi H (2007) Diagnosis of multiple synchronously occurring cystic jaw lesions in 23 patients: prognostic implications related to basal cell nevus syndrome. Oral Radiol 23:16–18
45. Ataíde AP, Fonseca FP, Santos Silva AR, Jorge Junior J, Lopes MA, Vargas PA (2016) Distribution of oral and maxillofacial lesions in pediatric patients from a Brazilian southeastern population. Int J Pediatr Otorhinolaryngol 90:241–244
46. Ba K, Li X, Wang H, Liu Y, Zheng G, Yang Z, Li M, Shimizutani K, Koseki T (2010) Correlation between imaging features and epithelial cell proliferation in keratocystic odontogenic tumour. Dentomaxillofac Radiol 39:368–374
47. Baghaei F, Zargaran M, Najmi H, Moghimbeigi A (2014) A clinicopathological study of odontogenic cysts and tumors in Hamadan, Iran. J Dent (Shiraz) 15:167–172
48. Brannon RB (1977) The odontogenic keratocyst. A clinicopathologic study of 312 cases. Part II. Histologic features. Oral Surg Oral Med Oral Pathol 43:233–255
49. Curran AE, Damm DD, Drummond JF (2002) Pathologically significant pericoronal lesions in adults: histopathologic evaluation. J Oral Maxillofac Surg 60:613–617
50. Daley TD, Wysocki GP, Pringle GA (1994) Relative incidence of odontogenic tumors and oral and jaw cysts in a Canadian population. Oral Surg Oral Med Oral Pathol 77:276–280
51. Dhanuthai K, Banrai M, Limpanaputtajak S (2007) A retrospective study of paediatric oral lesions from Thailand. Int J Paediatr Dent 17:248–253
52. Dimba EA, Gichana J, Limo AK, Wakoli KA, Chindia ML, Awange DO (2007) An audit of oral disease at a Nairobi centre, 2000-2004. Int Dent J 57:439–444
53. Ferreira O Jr, Damante JH, Lauris JRP (2004) Simple bone cyst versus odontogenic keratocyst: differential diagnosis by digitized panoramic radiography. Dentomaxillofac Radiol 33:373–378
54. Franklin CD, Jones AV (2006) A survey of oral and maxillofacial pathology specimens submitted by general dental practitioners over a 30-year period. Br Dent J 200:447–450
55. Gultelkin SE, Tokman B, Turkseven MR (2003) A review of paediatric oral biopsies in Turkey. Int Dent J 53:26–32
56. Jaeger F, de Noronha MS, Silva ML, Amaral MB, Grossmann SM, Horta MC, de Souza PE, de Aguiar MC, Mesquita RA (2017) Prevalence profile of odontogenic cysts and tumors on Brazilian sample after the reclassification of odontogenic keratocyst. J Craniomaxillofac Surg 45:267–270
57. Jamshidi S, Shojaei S, Roshanaei G, Modabbernia S, Bakhtiary E (2015) Jaw intraosseous lesions biopsied extracted from 1998 to 2010 in an Iranian population. Iran Red Crescent Med J 17:e20374
58. Jones AV, Franklin CD (2006) An analysis of oral and maxillofacial pathology found in adults over a 30-year period. J Oral Pathol Med 35:392–401
59. Jones K, Jordan RC (2010) Patterns of second-opinion diagnosis in oral and maxillofacial pathology. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 109:865–869
60. Kreidler JF, Raubenheimer EJ, van Heerden WF (1993) A retrospective analysis of 367 cystic lesions of the jaw—the Ulm experience. J Craniomaxillofac Surg 21:339–341
61. Krishnan R, Ramesh M, Paul G (2014) Retrospective evaluation of pediatric oral biopsies from a dental and maxillofacial surgery centre in Salem, Tamil Nadu, India. J Clin Diagn Res 8:221–223
62. Lapthanasupkul P, Juengsomjit R, Klanrit P, Taweechaisupapong S, Poomsawat S (2015) Oral and maxillofacial lesions in a Thai pediatric population: a retrospective review from two dental schools. J Med Assoc Thail 98:291–297
63. Ledesma-Montes C, Hernandez-Guerrero JC, Garces-Ortiz M (2000) Clinico-pathologic study of odontogenic cysts in a Mexican sample population. Arch Med Res 31:373–376
64. Lei F, Chen JY, Wang WC, Lin LM, Huang HC, Ho KY, Chen YK (2015) Retrospective study of oral and maxillofacial lesions in older Taiwanese patients. Gerodontology 32:281–287
65. Lima Gda S, Fontes ST, de Araujo LM, Etges A, Tarquinio SB, Gomes AP (2008) A survey of oral and maxillofacial biopsies in children: a single-center retrospective study of 20 years in Pelotas-Brazil. J Appl Oral Sci 16:397–402
66. Manor E, Kachko L, Puterman MB, Szabo G, Bodner L (2012) Cystic lesions of the jaws—a clinicopathological study of 322 cases and review of the literature. Int J Med Sci 9:20–26
67. Mohajerani H, Esmaeelinejad M, Mofidian R (2016) Ability of oral and maxillofacial surgery residents in diagnosing jaw cysts: a retrospective 20 years study. J Clin Diagn Res 10:zc42–zc45
68. Mosadomi A (1975) Tumors, cysts and allied lesions of the jaws and oral mucosa in Lagos, Nigeria, 1969-74. Int J Oral Surg 4: 219–224
69. Mosqueda Taylor A, Irigoyen Camacho ME, Díaz Franco MA, Torres Tejero MA (2002) Odontogenic cysts. Analysis of 856 cases. Med Oral 7:89–96
70. Prockt AP, Schebela CR, Maito FD, Sant'Ana-Filho M, Rados PV (2008) Odontogenic cysts: analysis of 680 cases in Brazil. Head Neck Pathol 2:150–156
71. Rachanis CC, Shear M (1978) Age standardized incidence rates of primordial cyst (keratocyst) on the Witwatersrand. Community Dent Oral Epidemiol 6:296–299
72. Raitz R, Assuncao Junior JN, Correa L, Fenyo-Pereira M (2009) Parameters in panoramic radiography for differentiation of radiolucent lesions. J Appl Oral Sci 17:381–387
73. Saghravanian N, Zare-Mahmoodabadi R, Ghazi N, Hosseinpour S (2015) Odontogenic cysts: a 40-year retrospective clinicopathological study in an Iranian population. Cumhuriyet Dent J 18:272–281

74. Sibda A, Meer S, Altini M (2012) Analysis of demographic data obtained from pathology records. Sadj 67:376–379
75. Sixto-Requeijo R, Diniz-Freitas M, Torreira-Lorenzo JC, Garcia-Garcia A, Gandara-Rey JM (2012) An analysis of oral biopsies extracted from 1995 to 2009, in an oral medicine and surgery unit in Galicia (Spain). Med Oral Patol Oral Cir Bucal 17:e16–e22
76. Tkaczuk AT, Bhatti M, Caccamese JF Jr, Ord RA, Pereira KD (2015) Cystic lesions of the jaw in children: a 15-year experience. JAMA Otolaryngol Head Neck Surg 141:834–839
77. Yazdani J, Kahnamouii SS (2009) Developmental odontogenic cysts of jaws: a clinical study of 245 cases. J Dent Res Dent Clin Dent Prospects 3:64–66
78. Bodner L, Manor E (2010) Cystic lesions of the jaws—a review and analysis of 269 cases. Eur J Plast Surg 33:277–282
79. Schlieve T, Miloro M, Kolokythas A (2014) Does decompression of odontogenic cysts and cystlike lesions change the histologic diagnosis? J Oral Maxillofac Surg 72:1094–1105
80. Singh HP, Nayar A, Raj A, Kumar P (2012) Are all odontogenic keratocysts keratocystic odontogenic tumors? Correlation between imaging features and epithelial cell proliferation. J Clin Imaging Sci 30:3–3
81. Ahlfors E, Larsson A, Sjogren S (1984) The odontogenic keratocyst: a benign cystic tumor? J Oral Maxillofac Surg 42: 10–19
82. Ali MA (2011) Biopsied jaw lesions in Kuwait: a six-year retrospective analysis. Med Princ Pract 20:550–555
83. Anand VK, Arrowood JP Jr, Krolls SO (1995) Odontogenic keratocysts: a study of 50 patients. Laryngoscope 105:14–16
84. Avelar RL, Antunes AA, Santos TDS, Andrade ESDS, Dourado E (2008) Odontogenic tumors: clinical and pathology study of 238 cases. Braz J Otorhinolaryngol 74:668–673
85. Bassey GO, Osunde OD, Anyanechi CE (2014) Maxillofacial tumors and tumor-like lesions in a Nigerian teaching hospital: an eleven year retrospective analysis. Afr Health Sci 14:56–63
86. Bataineh AB, Rawashdeh MA, Al Qudah MA (2004) The prevalence of inflammatory and developmental odontogenic cysts in a Jordanian population: a clinicopathologic study. Quintessence Int 35:815–819
87. Becconsall-Ryan K, Love RM (2011) Range and demographics of radiolucent jaw lesions in a New Zealand population. J Med Imaging Radiat Oncol 55:43–51
88. Buckley PC, Seldin EB, Dodson TB, August M (2012) Multilocularity as a radiographic marker of the keratocystic odontogenic tumor. J Oral Maxillofac Surg 70:320–324
89. Chow HT (1998) Odontogenic keratocyst: a clinical experience in Singapore. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 86: 573–577
90. Cunha JF, Gomes CC, de Mesquita RA, Andrade Goulart EM, de Castro WH, Gomez RS (2016) Clinicopathologic features associated with recurrence of the odontogenic keratocyst: a cohort retrospective analysis. Oral Surg Oral Med Oral Pathol Oral Radiol 121:629–635
91. da Silva LP, Gonzaga AK, Severo ML, Barros CC, de Medeiros AM, de Souza LB, da Silveira ÉJ (2018) Epidemiologic study of odontogenic and non-odontogenic cysts in children and adolescents of a Brazilian population. Med Oral Patol Oral Cir Bucal 23:e49–e53
92. Del Corso G, Righi A, Bombardi M, Rossi B, Dallera V, Pelliccioni GA, Marchetti C, Foschini MP (2014) Jaw cysts diagnosed in an Italian population over a 20-year period. Int J Surg Pathol 22:699–706
93. Dib LL, Curi MM, Chammas MC, Pinto DS, Torloni H (1996) Ultrasonography evaluation of bone lesions of the jaw. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 82:351–357
94. Dovigi EA, Kwok EY, Eversole LR, Dovigi AJ (2016) A retrospective study of 51,781 adult oral and maxillofacial biopsies. J Am Dent Assoc 147:170–176
95. El Gehani R, Krishnan B, Orafi H (2008) The prevalence of inflammatory and developmental odontogenic cysts in a Libyan population. Libyan J Med 3:75–77
96. Gao Z, Ni QW, Gao W, Liu YP, Zhang Q (2017) Application of endoscopy to treat mandibular keratocystic odontogenic tumors. Braz J Med Biol Res 50:e6209
97. Goteti SH (2016) Odontogenic tumors: a review of 675 cases in eastern Libya. Niger J Surg 22:37–40
98. Jaafari-Ashkavandi Z, Akbari B (2017) Clinicopathologic study of intra-osseous lesions of the jaws in southern Iranian population. J Dent (Shiraz) 18:259–264
99. Jattan R, De Silva HL, De Silva RK, Rich AM, Love RM (2011) A case series of odontogenic keratocysts from a New Zealand population over a 20-year period. N Z Dent J 107:112–116
100. Lam KY, Chan AC (2000) Odontogenic keratocysts: a clinicopathological study in Hong Kong Chinese. Laryngoscope 110: 1328–1332
101. Li N, Gao X, Xu Z, Chen Z, Zhu L, Wang J, Liu W (2014) Prevalence of developmental odontogenic cysts in children and adolescents with emphasis on dentigerous cyst and odontogenic keratocyst (keratocystic odontogenic tumor). Acta Odontol Scand 72:795–800
102. Lipovec A, Hren NI (2004) Keratocysts in the jaws. Radiol Oncol 38:187–192
103. Magnusson BC (1978) Odontogenic keratocysts: a clinical and histological study with special reference to enzyme histochemistry. J Oral Pathol Med 7:8–18
104. Meara JG, Li KK, Shah SS, Cunningham MJ (1996) Odontogenic keratocysts in the pediatric population. Arch Otolaryngol Head Neck Surg 122:725–728
105. Meara JG, Shah S, Li KK, Cunningham MJ (1998) The odontogenic keratocyst: a 20-year clinicopathologic review. Laryngoscope 108:280–283
106. Meningaud JP, Oprean N, Pitak-Arnnop P, Bertrand JC (2006) Odontogenic cysts: a clinical study of 695 cases. J Oral Sci 48: 59–62
107. Minami M, Kaneda T, Ozawa K, Yamamoto H, Itai Y, Ozawa M, Yoshikawa K, Sasaki Y (1996) Cystic lesions of the maxillomandibular region: MR imaging distinction of odontogenic keratocysts and ameloblastomas from other cysts. AJR Am J Roentgenol 166:943–949
108. Mohajerani H, Esmaeelinejad M, Sabour S, Aghdashi F, Dehghani N (2015) Diagnostic factors of odontogenic cysts in Iranian population: a retrospective study over the past two decades. Iran Red Crescent Med J 17:e21793
109. Probst FA, Probst M, Pautke C, Kaltsi E, Otto S, Schiel S, Troeltzsch M, Ehrenfeld M, Cornelius CP, Müller-Lisse UG (2015) Magnetic resonance imaging: a useful tool to distinguish between keratocystic odontogenic tumours and odontogenic cysts. Br J Oral Maxillofac Surg 53:217–222
110. Raj A, Ramesh G, Nagarajappa R, Pandey A (2017) Prevalence of odontogenic lesions among the Kanpur population: an institutional study. J Exp Ther Oncol 12:35–42
111. Rubini C, Mascitti M, Santarelli A, Tempesta A, Limongelli L, Favia G, Maiorano E (2017) Odontogenic tumors: a retrospective clinicopathological study from two Italian centers. Pathologica 109:35–46
112. Sanatkhani M, Zarch HH, Pakfetrat A, Falaki F (2011) Odontogenic cysts: a clinical and radiographic study of 58 cases. Aust J Basic & Appl Sci 5:329–333
113. Selvamani M, Devi AY, Basandi PS, Madhushankari GS (2014) Prevalence and clinicopathological comparison of kerotocystic odontogenic tumor and orthokeratinized odontogenic cyst in

South Indian sample population: a retrospective study over 13 years. J Pharm Bioallied Sci 6:S127–S130
114. Tekkesin MS, Olgac V, Aksakalli N, Alatli C (2012) Odontogenic and nonodontogenic cysts in Istanbul: analysis of 5088 cases. Head Neck 34:852–855
115. Tortorici S, Amodio E, Massenti MF, Buzzanca ML, Burruano F, Vitale F (2008) Prevalence and distribution of odontogenic cysts in Sicily: 1986-2005. J Oral Sci 50:15–18
116. Voorsmit RA, Stoelinga PJ, van Haelst UJ (1981) The management of keratocysts. J Maxillofac Surg 9:228–236
117. Wziatek-Kuczmik D, Radzioch J, Pajak J, Niedzielska I, Drugacz J (2007) Ameloblastomas and odontogenic cysts of the mandible—comparative analysis based on clinical, radiological and morphological material of the Department of Maxillofacial Surgery, Medical University of Silesia, Katowice. Chirurgia Polska 9:212–217
118. Yoshiura K, Higuchi Y, Araki K, Shinohara M, Kawazu T, Yuasa K, Tabata O, Kanda S (1997) Morphologic analysis of odontogenic cysts with computed tomography. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 83:712–718
119. Zachariades N, Papanicolaou S, Triantafyllou D (1985) Odontogenic keratocysts: review of the literature and report of sixteen cases. J Oral Maxillofac Surg 43:177
120. Zhao YF, Wei JX, Wang SP (2002) Treatment of odontogenic keratocysts: a follow-up of 255 Chinese patients. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 94:151–156
121. AlSheddi MA, AlSenani MA, AlDosari AW (2015) Odontogenic tumors: analysis of 188 cases from Saudi Arabia. Ann Saudi Med 35:146–150
122. August M, Faquin WC, Troulis MJ, Kaban LB (2003) Dedifferentiation of odontogenic keratocyst epithelium after cyst decompression. J Oral Maxillofac Surg 61:678–683
123. da Silva LP, Serpa MS, Tenório JR, do Nascimento GJ, de Souza-Andrade ES, Veras-Sobral AP (2016) Retrospective study of 289 odontogenic tumors in a Brazilian population. Med Oral Patol Oral Cir Bucal 21:e271–e275
124. da-Costa DO, Maurício AS, de-Faria PA, da-Silva LE, Mosqueda-Taylor A, Lourenço SD (2012) Odontogenic tumors: a retrospective study of four Brazilian diagnostic pathology centers. Med Oral Patol Oral Cir Bucal 17:e389–e394
125. Gill S, Chawda J, Jani D (2011) Odontogenic tumors in Western India (Gujarat): analysis of 209 cases. J Clin Exp Dent 3:e78–e83
126. Grossmann SM, Machado VC, Xavier GM, Moura MD, Gomez RS, Aguiar MC, Mesquita RA (2007) Demographic profile of odontogenic and selected nonodontogenic cysts in a Brazilian population. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 104:e35–e41
127. Maurette PE, Jorge J, de Moraes M (2006) Conservative treatment protocol of odontogenic keratocyst: a preliminary study. J Oral Maxillofac Surg 64:379–383
128. Oginni FO, Stoelinga PJ, Ajike SA, Obuekwe ON, Olokun BA, Adebola RA, Adeyemo WL, Fasola O, Adesina OA, Akinbami BO, Iwegbu IO, Ogunmuyiwa SA, Obimakinde OS, Uguru CC (2015) A prospective epidemiological study on odontogenic tumours in a black African population, with emphasis on the relative frequency of ameloblastoma. Int J Oral Maxillofac Surg 44:1099–1105
129. Partridge M, Towers JF (1987) The primordial cyst (odontogenic keratocyst): its tumour-like characteristics and behaviour. Br J Oral Maxillofac Surg 25:271–279
130. Rao K, Smitha, Umadevi HS, Priya NS (2011) Clinicopathological study of 100 odontogenic cysts reported at V S Dental College—a retrospective study. J Adv Dent Res 1: 51–58
131. Rao K, Kumar S (2014) The use of enucleation and chemical cauterization (Carnoy's) in the management of odontogenic keratocyst of the jaws. Indian J Otolaryngol Head Neck Surg 66: 8–12
132. Sánchez-Burgos R, González-Martín-Moro J, Pérez-Fernández E, Burgueño-García M (2014) Clinical, radiological and therapeutic features of keratocystic odontogenic tumours: a study over a decade. J Clin Exp Dent 6:e259–e264
133. Tabrizi R, Özkan BT, Dehgani A, Langner NJ (2012) Marsupialization as a treatment option for the odontogenic keratocyst. J Craniofac Surg 23:e459–e461
134. Worawongvasua R, Tiensuwanb M (2015) Odontogenic tumors in Thailand: a study of 590 Thai patients. J Oral Maxillofac Surg Med Pathol 27:567–576
135. Akram S, Naghma AMA, Shakir MM (2013) Prevalence of odontogenic cysts and tumors in Karachi, Pakistan. J Dow Uni Health Sci 7:20–24
136. Azevedo RS, Cabral MG, dos Santos TC, de Oliveira AV, de Almeida OP, Pires FR (2012) Histopathological features of keratocystic odontogenic tumor: a descriptive study of 177 cases from a Brazilian population. Int J Surg Pathol 20:154–160
137. Brannon RB (1976) The odontogenic keratocyst. A clinicopathologic study of 312 cases. Part I. Clinical features. Oral Surg Oral Med Oral Pathol 42:54–72
138. Habibi A, Saghravanian N, Habibi M, Mellati E (2007) Keratocystic odontogenic tumor: a 10-year retrospective study of 83 cases in an Iranian population. J Oral Sci 49:229–235
139. Hodgkinson DJ, Woods JE, Dahlin DC, Tolman DE (1978) Keratocysts of the jaw. Clinicopathologic study of 79 patients. Cancer 41:803–813
140. Jones AV, Craig GT, Franklin CD (2006) Range and demographics of odontogenic cysts diagnosed in a UK population over a 30-year period. J Oral Pathol Med 35:500–507
141. Kauke M, Safi AF, Grandoch A, Nickenig HJ, Zoller J, Kreppel M (2017) Volumetric analysis of keratocystic odontogenic tumors and non-neoplastic jaw cysts—comparison and its clinical relevance. J Craniomaxillofac Surg
142. Mello LA, Gurgel CA, Ramos EA, de Souza RO, Schlaepfer-Sales CB, de Azevedo RA, dos Santos JN (2011) Keratocystic odontogenic tumour: an experience in the northeast of Brazil. Srp Arh Celok Lek 139:291–297
143. Myoung H, Hong SP, Hong SD, Lee JI, Lim CY, Choung PH, Lee JH, Choi JY, Seo BM, Kim MJ (2001) Odontogenic keratocyst: review of 256 cases for recurrence and clinicopathologic parameters. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 91:328–333
144. Ochsenius G, Escobar E, Godoy L, Penafiel C (2007) Odontogenic cysts: analysis of 2,944 cases in Chile. Med Oral Patol Oral Cir Bucal 12:E85–E91
145. Oda D, Rivera V, Ghanee N, Kenny EA, Dawson KH (2000) Odontogenic keratocyst: the northwestern USA experience. J Contemp Dent Pract 1:60–74
146. Radden BG, Reade PC (1973) Odontogenic cysts. A review and a clinicopathological study of 368 odontogenic cysts. Aust Dent J 18:218–225
147. Radden BG, Reade PC (1973) Odontogenic keratocysts. Pathology 5:325–334
148. Rengaswamy V (1977) Clinical statistics of odontogenic cysts in West Malaysia. Br J Oral Surg 15:160–165
149. Sansare K, Raghav M, Mupparapu M, Mundada N, Karjodkar FR, Bansal S, Desai R (2013) Keratocystic odontogenic tumor: systematic review with analysis of 72 additional cases from Mumbai, India. Oral Surg Oral Med Oral Pathol Oral Radiol 115:128–139
150. Titinchi F, Nortje CJ (2012) Keratocystic odontogenic tumor: a recurrence analysis of clinical and radiographic parameters. Oral Surg Oral Med Oral Pathol Oral Radiol 114:136–142
151. Tsukamoto G, Sasaki A, Akiyama T, Ishikawa T, Kishimoto K, Nishiyama A, Matsumura T (2001) A radiologic analysis of

dentigerous cysts and odontogenic keratocysts associated with a mandibular third molar. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 91:743–747
152. Eversole LR, Sabes WR, Rovin S (1975) Aggressive growth and neoplastic potential of odontogenic cysts: with special reference to central epidermoid and mucoepidermoid carcinomas. Cancer 35: 270–282
153. Kakarantza-Angelopoulou E, Nicolatou O (1990) Odontogenic keratocysts: clinicopathologic study of 87 cases. J Oral Maxillofac Surg 48:593–599
154. Yang S, Park Y, Choi S, Kim J, Kim C (2011) A retrospective study of 220 cases of keratocystic odontogenic tumor (KCOT) in 181 patients. Asian J Oral Maxillofac Surg 23:117–121
155. Anniko M, Anneroth G, Bergstedt H, Ramström G (1981) Jaw cysts with special regard to keratocyst recurrence—a long-term follow-up. Arch Otorhinolaryngol 233:261–269
156. Guler N, Sencift K, Demirkol O (2012) Conservative management of keratocystic odontogenic tumors of jaws. Sci World 2012: 680397
157. Muzio L, Mascitti M, Santarelli A, Rubini C, Bambini F, Procaccini M, Bertossi D, Albanese M, Bondì V, Nocini PF (2017) Cystic lesions of the jaws: a retrospective clinicopathologic study of 2030 cases. Oral Surg Oral Med Oral Pathol Oral Radiol 124:128–138
158. Noy D, Rachmiel A, Zar K, Emodi O, Nagler RM (2017) Sporadic versus syndromic keratocysts—can we predict treatment outcome? A review of 102 cysts. Oral Dis 23:1058–1065
159. Pitak-Arnnop P, Chaine A, Oprean N, Dhanuthai K, Bertrand JC, Bertolus C (2010) Management of odontogenic keratocysts of the jaws: a ten-year experience with 120 consecutive lesions. J Craniomaxillofac Surg 38:358–364
160. Stoelinga PJ, Bronkhorst FB (1988) The incidence, multiple presentation and recurrence of aggressive cysts of the jaws. J Craniomaxillofac Surg 16:184–195
161. Luo HY, Li TJ (2009) Odontogenic tumors: a study of 1309 cases in a Chinese population. Oral Oncol 45:706–711
162. Haring JI, Van Dis ML (1988) Odontogenic keratocysts: a clinical, radiographic, and histopathologic study. Oral Surg Oral Med Oral Pathol 66:145–153
163. Al-Khateeb TH, Bataineh AB (2006) Pathology associated with impacted mandibular third molars in a group of Jordanians. J Oral Maxillofac Surg 64:1598–1602
164. Barry JY, Le CH, Baumann J, Skinker L, Chiu AG, Chang EH (2016) Endoscopic resection of maxillary sinus keratocystic odontogenic tumors. Laryngoscope 126:2216–2219
165. Eliasson S, Isacsson G, Kondell PA (1989) Lateral periodontal cysts. Clinical, radiographical and histopathological findings. Int J Oral Maxillofac Surg 18:191–193
166. Garlock JA, Pringle CA, Hicks ML (1998) The odontogenic keratocyst—a potential endodontic misdiagnosis. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 85:452–456
167. Hofer B, Hardt N, Voegeli E, Kinser J (1985) A diagnostic approach to lytic lesions of the mandible. Skelet Radiol 14:164–172
168. Ide F, Horie N, Shimoyama T, Saito I, Tanaka A, Kusama K (2009) Infrequent clinicopathologic features of keratocystic odontogenic tumour: a 29-year multi-institutional retrospective review. Oral Surg 2:1–9
169. Paul S, Kapoor V, Kumar M, Narula R, Kapoor U (2014) A clinical study of cysts of the maxillofacial region; and an assessment of clinico-radiologico-pathological variables affecting the formulation of a comprehensive patient need based treatment plan. Indian J Dent 5:69–74
170. Smith I, Shear M (1978) Radiological features of mandibular primordial cysts (keratocysts). J Maxillofac Surg 6:147–154
171. Stoelinga PJW, Peters JH (1973) A note on the origin of keratocysts of the jaws. Int J Oral Surg 2:37–44
172. Sumer AP, Danaci M, Ozen Sandikci E, Sumer M, Celenk P (2009) Ultrasonography and Doppler ultrasonography in the evaluation of intraosseous lesions of the jaws. Dentomaxillofac Radiol 38:23–27
173. Vigneswaran AT, Shilpa S (2015) The incidence of cysts and tumors associated with impacted third molars. J Pharm Bioallied Sci 7:S251–S254
174. Kolokythas A, Fernandes RP, Pazoki A, Ord RA (2007) Odontogenic keratocyst: to decompress or not to decompress? A comparative study of decompression and enucleation versus resection/peripheral ostectomy. J Oral Maxillofac Surg 65:640–644
175. Al Yamani AO, Al Sebaei MO, Bassyoni LJ, Badghaish AJ, Shawly HH (2011) Variation of pediatric and adolescents head and neck pathology in the city of Jeddah: a retrospective analysis over 10 years. Saudi Dent J 23:197–200
176. Alcantara BA, Carli ML, Beijo LA, Pereira AA, Hanemann JÁ (2013) Correlation between inflammatory infiltrate and epithelial lining in 214 cases of periapical cysts. Braz Oral Res 27:490–495
177. Avelar RL, Antunes AA, Carvalho RW, Bezerra PG, Oliveira Neto PJ, Andrade ES (2009) Odontogenic cysts: a clinicopathological study of 507 cases. J Oral Sci 51:581–586
178. Bekiroglu N, Mete S, Ozbay G, Yalcinkaya S, Kargul B (2015) Evaluation of panoramic radiographs taken from 1,056 Turkish children. Niger J Clin Pract 18:8–12
179. Biswas D, Crank S (2007) Aetiopathology of maxillary swelling—a 3-year prospective study. Eur Arch Otorhinolaryngol 264:1295–1299
180. Butt FM, Ogeng'o J, Bahra J, Chindia ML (2011) Pattern of odontogenic and nonodontogenic cysts. J Craniofac Surg 22: 2160–2162
181. DelBalso AM (1995) Lesions of the jaws. Semin Ultrasound CT MR 16:487–512
182. Gupta B, Ponniah I (2010) The pattern of odontogenic tumors in a government teaching hospital in the southern Indian state of Tamil Nadu. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 110: e32–e39
183. Kalyanyama BM, Matee MI, Vuhahula E (2002) Oral tumours in Tanzanian children based on biopsy materials examined over a 15-year period from 1982 to 1997. Int Dent J 52:10–14
184. Kilinc A, Gundogdu B, Saruhan N, Yalcin E, Ertas U, Urvasizoglu G (2017) Odontogenic and nonodontogenic cysts: an analysis of 526 cases in Turkey. Niger J Clin Pract 20:879–883
185. Kondori I, Mottin RW, Laskin DM (2011) Accuracy of dentists in the clinical diagnosis of oral lesions. Quintessence Int 42:575–577
186. Lawal AO, Adisa AO, Sigbeku OF (2012) Cysts of the oro-facial region: a Nigerian experience. J Oral Maxillofac Pathol 16:167–171
187. Lustig JP, Schwartz-Arad D, Shapira A (1999) Odontogenic cysts related to pulpotomized deciduous molars: clinical features and treatment outcome. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 87:499–503
188. Mortensen H, Winther JE, Birn H (1970) Periapical granulomas and cysts. An investigation of 1,600 cases. Scand J Dent Res 78: 241–250
189. Munsamy C, Mahomed F, Rikhotso E (2011) A 20-year retrospective study of oral and maxillofacial histopathology in a South African paediatric population sample. Sadj 66:268–271
190. Nunez-Urrutia S, Figueiredo R, Gay-Escoda C (2010) Retrospective clinicopathological study of 418 odontogenic cysts. Med Oral Patol Oral Cir Bucal 15:e767–e773
191. Sidhu SS, Parkash H (1975) Odontogenic cystic lesions of the jaws. Aust Dent J 20:371–374
192. Spatafore CM, Griffin JA Jr, Keyes GG, Wearden S, Skidmore AE (1990) Periapical biopsy report: an analysis of over a 10-year period. J Endod 16:239–241

193. Syrjanen S, Tammisalo E, Lilja R, Syrjanen K (1982) Radiological interpretation of the periapical cysts and granulomas. Dentomaxillofac Radiol 11:89–92
194. Ulmansky M, Lustmann J, Balkin N (1999) Tumors and tumor-like lesions of the oral cavity and related structures in Israeli children. Int J Oral Maxillofac Surg 28:291–294
195. Varinauskas V, Gervickas A, Kavoliuniene O (2006) Analysis of odontogenic cysts of the jaws. Medicina (Kaunas) 42:201–207
196. Villasis-Sarmiento L, Portilla-Robertson J, Melendez-Ocampo A, Gaitan-Cepeda LA, Leyva-Huerta ER (2017) Prevalence and distribution of odontogenic cysts in a Mexican sample. A 753 cases study. J Clin Exp Dent 9:e531–e538
197. Yakin M, Jalal JA, Al-Khurri LE, Rich AM (2016) Oral and maxillofacial pathology submitted to Rizgary teaching hospital: a 6-year retrospective study. Int Dent J 66:78–85
198. Asutay F, Atalay Y, Turamanlar O, Horata E, Burdurlu MC (2016) Three-dimensional volumetric assessment of the effect of decompression on large mandibular odontogenic cystic lesions. J Oral Maxillofac Surg 74:1159–1166
199. Guven O, Keskin A, Akal UK (2000) The incidence of cysts and tumors around impacted third molars. Int J Oral Maxillofac Surg 29:131–135
200. Koivisto T, Bowles WR, Rohrer M (2012) Frequency and distribution of radiolucent jaw lesions: a retrospective analysis of 9,723 cases. J Endod 38:729–732
201. Kontogiannis TG, Tosios KI, Kerezoudis NP, Krithinakis S, Christopoulos P, Sklavounou A (2015) Periapical lesions are not always a sequelae of pulpal necrosis: a retrospective study of 1521 biopsies. Int Endod J 48:68–73
202. Neville BW, Damm DD, Brock T (1997) Odontogenic keratocysts of the midline maxillary region. J Oral Maxillofac Surg 55:340–344
203. Ortega A, Fariña V, Gallardo A, Espinoza I, Acosta S (2007) Nonendodontic periapical lesions: a retrospective study in Chile. Int Endod J 40:386–390
204. Soluk Tekkesin M, Tuna EB, Olgac V, Aksakalli N, Alatli C (2016) Odontogenic lesions in a pediatric population: review of the literature and presentation of 745 cases. Int J Pediatr Otorhinolaryngol 86:196–199
205. Tsukamoto G, Makino T, Kikuchi T, Kishimoto K, Nishiyama A, Sasaki A, Matsumura T (2002) A comparative study of odontogenic keratocysts associated with and not associated with an impacted mandibular third molar. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 94:272–275
206. Ansari SR, Rehman AU, Rehman B (2010) Frequency and demography of commonly occurring odontogenic cysts in Khyber Pakhtunkhwa (Pakistan). Pakistan Oral Dent J 30: 41–46
207. Apajalahti S, Hagström J, Lindqvist C, Suomalainen A (2011) Computerized tomography findings and recurrence of keratocystic odontogenic tumor of the mandible and maxillofacial region in a series of 46 patients. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 111:29–37
208. Farmand M, Makek M (1983) Late results following the Broschprocedure for treating large mandibular ramus cysts. J Maxillofac Surg 11:211–215
209. Macdonald-Jankowski DS, Li TK (2010) Keratocystic odontogenic tumour in a Hong Kong community: the clinical and radiological features. Dentomaxillofac Radiol 39:167–175
210. Sekerci AE, Nazlim S, Etoz M, Deniz K, Yasa Y (2015) Odontogenic tumors: a collaborative study of 218 cases diagnosed over 12 years and comprehensive review of the literature. Med Oral Patol Oral Cir Bucal 20:e34–e44
211. Servato JP, Prieto-Oliveira P, de Faria PR, Loyola AM, Cardoso SV (2013) Odontogenic tumours: 240 cases diagnosed over 31 years at a Brazilian university and a review of international literature. Int J Oral Maxillofac Surg 42:288–293
212. Shudou H, Sasaki M, Yamashiro T, Tsunomachi S, Takenoshita Y, Kubota Y, Ninomiya T, Kawazu T, Mori Y (2012) Marsupialisation for keratocystic odontogenic tumours in the mandible: longitudinal image analysis of tumour size using 3D visualised CT scans. Int J Oral Maxillofac Surg 41:290–296
213. Siriwardena BS, Tennakoon TM, Tilakaratne WM (2012) Relative frequency of odontogenic tumors in Sri Lanka: analysis of 1677 cases. Pathol Res Pract 208:225–230
214. Tawfik MA, Zyada MM (2010) Odontogenic tumors in Dakahlia, Egypt: analysis of 82 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 109:e67–e73
215. The Joanna Briggs Institute. Critical appraisal tools. http://joannabriggs.org/research/critical-appraisal-tools.html [Accessibility verified June 11, 2018]
216. Higgins JPT, Green S (2011) Cochrane handbook for systematic reviews of interventions version 5.1.0. The Cochrane Collaboration. Available from http://handbook-5-1.cochrane.org/ [Accessibility verified August 8, 2018]
217. Rodu B, Tate AL, Martinez MG (1987) The implications of inflammation in odontogenic keratocysts. J Oral Pathol 16:518–519