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REVIEW ARTICLE

Peripheral calcifying cystic odontogenic tumour and peripheral dentinogenic ghost cell tumour: an updated systematic review of 117 cases reported in the literature

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ABSTRACT

Purpose: To integrate the available data published on peripheral calcifying cystic odontogenic tumour (CCOT) and peripheral dentinogenic ghost cell tumour (DGCT) into a comprehensive analysis of its clinical and radiologic features.

Methods: An electronic search was undertaken in May, 2016. Eligibility criteria included publications reporting cases of peripheral CCOTs/DGCTs having enough clinical, radiological and histological information to confirm a definite diagnosis. Demographic data, lesion site and size, treatment approach and recurrence were analyzed.

Results: Hundred and thirty-eight lesions were found (65 publications), and 117 lesions (63 publications) with enough information were analyzed (55 CCOTs, 50 DGCTs, 12 unknown). Mean age of patients was 51.3 ± 23.4 (min–max, 1–92), with higher mean age for the DGCTs variant. The lesions were more prevalent in the mandible, anterior region of the jaws, and in the second, sixth and eighth decades, with an equal sexual distribution. About 20% of all lesions showed signs of erosion of the underlying bone, with a higher rate for DGCTs. The mean lesion size was 1.3 ± 0.8 (min–max, 0.4–3.0). Time of follow-up was informed for 37 lesions, with a mean ± SD of 30.2 ± 21.0 months (min–max, 6–84). Almost all lesions were treated by conservative surgery; only three recurrences were reported.

Conclusions: Peripheral CCOTs/DGCTs are rare lesions. Most of the lesions were treated by simple excision with or without curettage of the underlying bone. As the recurrence rate is very low, a conservative approach seems to be enough for the great majority of cases.

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Introduction

The calcifying cystic odontogenic tumour (CCOT), term used since it was coined by the World Health Organization (WHO) in 2005,[1] was first recognized as a distinct clinicopathologic entity in 1962 by Gorlin et al. [2] The lesion is a benign cystic neoplasm of odontogenic origin, characterized by an ameloblastoma-like epithelium with ghost cells that may calcify. The lesion may present as an intraosseous (central) or extraosseous (peripheral) process.[1] The dentinogenic ghost cell tumour (DGCT) is the solid variant of CCOT and is characterized by ameloblastoma-like islands of epithelial cells associated with ghost cells and varying amounts of dysplastic dentin.[1]

The peripheral CCOT and peripheral DGCT are extremely rare lesions, and thus there are limited details in the literature regarding their clinical and radiologic features. Only few studies [2–8] have reported a handful of cases, and almost all other publications are comprised of single-case reports. The epidemiological study of such rare lesions is of great importance because it provides information that can improve the diagnostic accuracy and could help pathologists and surgeons to make informed decisions and refine the treatment plan to optimize the clinical outcome. The aim of the present study was to integrate the available data published in the literature on peripheral CCOT/DGCT into an updated comprehensive analysis of its clinical and radiologic features, as well as to report the frequency of recurrence.

Materials and methods

This study followed the PRISMA Statement guidelines.[9] A review protocol does not exist.

Search strategies

An electronic search without time restrictions was undertaken in May 2016 in the following databases: PubMed/Medline, Web of Science and Scopus. The following terms were used in the search strategies: (calcifying cystic odontogenic tumour) OR (calcifying odontogenic cystic tumour) OR (calcifying odontogenic cyst) OR (Gorlin cyst) OR (dentinogenic ghost cell tumour) OR (odontogenic ghost cell tumour)

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OR (epithelial odontogenic ghost cell tumour) OR (calcifying ghost cell odontogenic tumour)

Moreover, Google Scholar was also checked. A manual search of related journals, including *Acta Odontologica Scandinavica, Acta Oto-Laryngologica, Annals of Otology Rhinology and Laryngology, British Journal of Oral and Maxillofacial Surgery, Cancer, Head & Neck, Head and Neck Pathology, International Journal of Oral and Maxillofacial Surgery, Journal of Dental Research, Journal of Craniofacial Surgery, Journal of Cranio-Maxillofacial Surgery, Journal of Laryngology and Otology, Journal of Maxillofacial and Oral Surgery, Journal of Oral and Maxillofacial Surgery, Journal of Oral Pathology and Medicine, Laryngoscope, Oral Diseases, Oral Oncology, Oral Surgery Oral Medicine Oral Pathology Oral Radiology, Otolaryngology – Head and Neck Surgery,* and *Quintessence International*, was performed. The reference list of the identified studies and the relevant reviews on the subject were also scanned for possible additional studies.

Inclusion and exclusion criteria

Eligibility criteria included publications written in any European language reporting cases of peripheral CCOTs and/or DGCTs. The studies needed to have enough clinical, radiological and histological information to confirm a definite diagnosis of peripheral CCOT/DGCT. Randomized and controlled clinical trials, cohort studies, case-control studies, cross-sectional studies, case series and case reports were included. Unless any of following publication categories had reported cases with enough clinical, radiological and histological data, they were excluded from the present review: immunohistochemical studies, histomorphometric studies, radiological studies, genetic expression studies, histopathological studies, cytological studies, cell proliferation/apoptosis studies, *in vitro* studies and review papers.

Study selection

The titles and abstracts of all reports identified through the electronic searches were read independently by the authors. For studies appearing to meet the inclusion criteria, or for which there were insufficient data in the title and abstract to make a clear decision, the full report was obtained. Disagreements were solved by discussion between the authors. The clinical and radiological aspects, as well as the histological description of the lesions were thoroughly assessed by one of the authors, an expert in oral pathology (R.S.G.), in order to confirm the diagnosis of peripheral CCOT.

Data extraction

The authors independently extracted data using specially designed forms. Any disagreements were solved by discussion. For each of the included studies, the following data were extracted, when available: year of publication, number of patients, patient's sex, age and race, follow-up period, duration of the lesion previously to treatment, lesion location (maxilla/mandible), anterior/posterior location (three categories: (a) anterior: lesions in the incisors/canine region; (b) premolar region; (c) posterior: lesions in the molars/retromolar region), recurrence, recurrence period, lesion size, histological features and presence of erosion of the subjacent cortical bone. The lesion size was determined according to the largest diameter. Because of the limited information available on the studies regarding the histopathological aspects of each case reported, no attempt was made to characterize the microscopic variants of CCOT (simple unicystic, odontoma-associated, ameloblastomatous proliferating associated with benign odontogenic tumours other than odontoma). Ghost cell odontogenic carcinoma was not considered for this study. Contact with authors for possible missing data was performed.

Analyses

Differences in the frequency between the cystic (CCOTs) and solid (DGCTs) variants of the lesion for the variables gender, lesion location (maxilla/mandible), anterior/posterior location and erosion of the subjacent cortical bone were analyzed by Pearson's chi-squared or Fisher's exact tests, depending on the expected count of events in a 2×2 contingency table. The performed tests to analyze the difference in the mean values between the groups (cystic and solid variants) for continuous variables (patient's age, lesion size) were Student's *t*-test or Mann–Whitney test, depending on the normality. Kolmogorov–Smirnov test was performed to evaluate the normal distribution of the variables, and Levene's test evaluated homoscedasticity. The degree of statistical significance was considered $p < .05$. All data were statistically analyzed using the Statistical Package for the Social Sciences (SPSS) version 23 software (SPSS Inc., Chicago, IL).

Results

Literature search

The study selection process is summarized in Figure 1. The search strategy in the databases resulted in 2482 papers. Search in Google Scholar resulted in seven eligible papers not found in the three main databases. A number of 1586 articles were cited in more than one database (duplicates). Of the resulted 903 studies, 629 were excluded for not being related to the topic. Additional hand-searching of journals and reference lists of selected studies yielded one additional paper. The full-text reports of the remaining 275 articles led to the exclusion of 210 because they did not meet the inclusion criteria: 161 clinical series/case reports of central CCOTs/DGCTs not including peripheral lesions, 26 immunohistochemical studies, 6 histopathological studies, 6 radiological studies of central lesions, 5 genetic/glycoprotein/cytokeratin expression studies, 5 review papers and 1 cell proliferation/apoptosis study. Thus, a total of 65 publications were included in the review.

Description of the studies and analyses

Sixty-three publications [2,3,5,7,8,10–67] were included in the present review, reporting 117 cases of peripheral

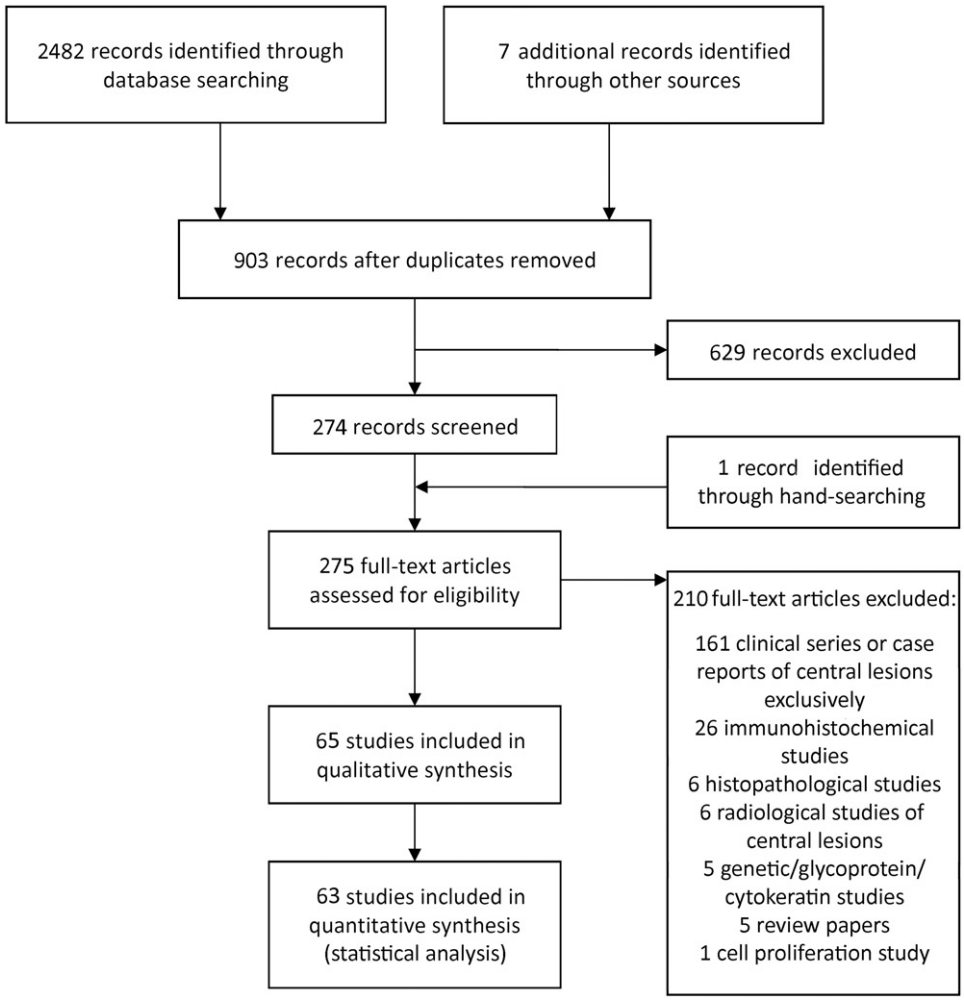

Figure 1. Study screening process.

CCOTs/DGCTs. It is important to mention the particularities of two publications not included in the analysis. The first one is Johnson et al.,[6] who reported 57 cases of CCOTs/DGCTs, of which 17 were peripheral lesions. The authors did not provide any kind of detail about these 17 peripheral lesions, besides the fact that any of them had recurred, and due of that these lesions were not included. The second publication is Habibi et al.,[68] who reported 60 cases of CCOTs/DGCTs, of which 4 were peripheral ones, but again the authors did not provide any kind of detail about them. Thus, of the total of 138 peripheral CCOTs/DGCTs found in the literature in 65 publications, 21 lesions from these two studies [6,68] were not accounted for the statistical analyses.

The other 63 publications reported 117 cases of peripheral CCOTs/DGCTs, of which three recurred (2.56%). From the 117 lesions, 50 were solid DGCTs and 55 were CCOTs. It was not possible to certify whether the lesions were CCOT or DGCT in 12 cases from 5 articles.[3,11,19, 21,45] The three recurrences were cystic lesions, two occurring 3 years after the excision [25,52] and the other one without information about the time of recidive.[8]

Table 1 presents the demographic and clinical features of all 117 lesions included in the analysis. There was an almost equal distribution of the lesions between males and females. The age of the patient was known for 101 lesions; when all these lesions ($n = 101$) were considered, there were peaks of prevalence in the second, sixth and eighth decades (Figure 2). The bimodal distribution of the cases according to age was observed for the cystic variant, but not for the solid one. The lesions were more prevalent in the mandible than in the maxilla (Figure 3), and at the anterior region than in the posterior region, for both cystic and solid variants. About 20% of all lesions showed signs of erosion of the subjacent osseous surface. The difference in mean size between the cystic and solid types was not statistically significant. Treatment of the lesions was reported in 116 cases, with conservative surgery in 114 cases (110 excisions, 3 enucleations, 1 marsupialization) and 2 cases treated by marginal resection. The three primary lesions that recurred were treated by simple excision. Two of the recurrent lesions were treated by conservative excision, after which no recurrence was reported after 12 [52] and 40 months.[25] There was no information about the treatment and follow-up of the third recurrence.[8] Time of follow-up was informed for 37 lesions, with a mean ± SD of 30.2 ± 21.0 months (min–max, 6–84). The patients' race was informed in 55 cases. Twenty-five lesions (45.5%) occurred in whites, 11 in blacks, 6 in Indians, 6 in Persians, 3 in Asians, 2 in Hispanics and 2 in Turks.

Table 1. Demographic and clinical features of peripheral CCOTs and peripheral dentinogenic ghost cell tumours (DGCT) described in the literature.

	All lesions	CCOT variant	DGCT variant	*p* Value
Sample size (*n*)	117[c]	55	50	
Age (year), mean ± SD (min–max)	51.3 ± 23.4 (1–92)	41.8 ± 25.0 (1–80)	57.0 ± 19.2 (7–92)	.002[a,e]
Men	46.8 ± 24.9 (1–82)	33.3 ± 26.1 (1–74)	55.8 ± 19.5 (10–82)	.003[a,e]
Women	55.9 ± 20.9 (7–92)	49.1 ± 22.0 (10–80)	58.6 ± 19.2 (7–92)	.142[a,e]
p value[d]	0.048[a]	0.041[a]	0.621[a]	
Gender, *n* (%)				
Men	53 (52.0)	20 (47.6)	28 (57.1)	.364[b,f]
Women	49 (48.0)	22 (52.4)	21 (42.9)	
Unknown	15	13	1	
Jaw, *n* (%)				
Maxilla	39 (34.8)	21 (40.4)	13 (27.1)	.161[b,f]
Mandible	73 (65.2)	31 (59.6)	35 (72.9)	
Unknown	5	3	2	
Location				
Incisor-canine	58 (62.4)	28 (68.3)	24 (60.0)	.105[b,f]
Premolar	19 (20.4)	5 (12.2)	12 (30.0)	
Molar-retromolar	16 (17.2)	8 (19.5)	4 (10.0)	
Unknown	24	14	10	
Bone erosion, *n* (%)				
Yes	21 (21.6)	7 (17.1)	13 (29.5)	.176[b,f]
No	76 (78.4)	34 (82.9)	31 (70.5)	
Unknown	20	14	6	
Lesion size (cm), mean ± SD (min-max)	1.3 ± 0.8 (0.4–3.0)	1.1 ± 0.7 (0.5–3.0)	1.5 ± 0.8 (0.4–3.0)	.101[a,e]

SD: standard deviation.
[a]Student's *t*-test.
[b]Pearson's chi-squared test.
[c]It was not possible to certify whether the lesions were CCOT or DGCT in 12 cases.
[d]Comparison of mean age between men and women.
[e]Comparison of mean values between the CCOT and DGCT variants.
[f]Comparison of the frequency of the variables between the CCOT and DGCT variants.

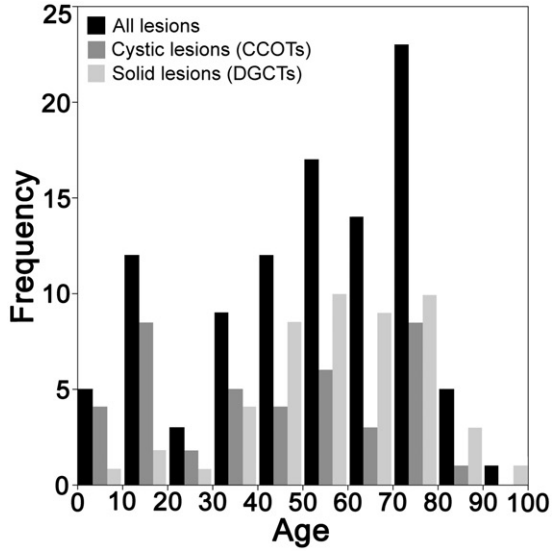

Figure 2. Age distribution of the peripheral calcifying cystic odontogenic tumours ($n = 101$; for 16 lesions the age of the patient was not informed), also discriminated for the peripheral cystic (CCOTs, $n = 41$) and peripheral solid lesions (DGCTs, $n = 49$). The total number of cystic and solid variants does not add up to the total number of lesions, because it was not possible for some cases to certify whether they were cystic or solid.

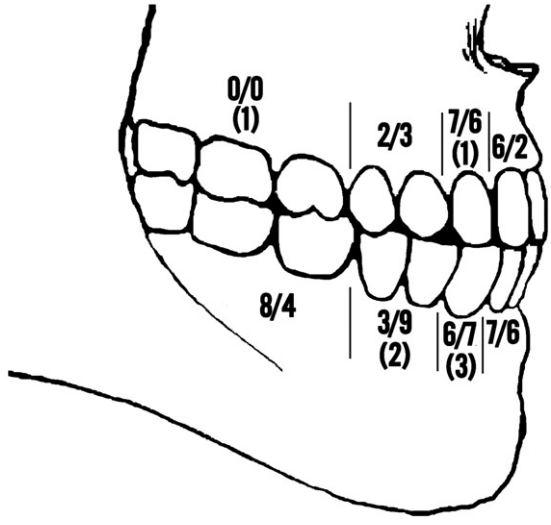

Figure 3. Distribution of the known precise locations ($n = 83$) of the peripheral calcifying cystic odontogenic tumour (CCOTs)/peripheral dentinogenic ghost cell tumour (DGCTs). The number between parentheses represents lesions for which a distinction between CCOT and DGCT is not known. For the rest of the lesions ($n = 34$), the location was 'anterior' for 2 lesions in the maxilla and 5 in the mandible, 'posterior' for 3 lesions in the maxilla, unknown for 6 lesions in the maxilla and 13 in the mandible, and for 5 lesions was not informed whether the lesion was located in the maxilla or mandible.

Discussion

Gorlin et al. [2] were the first to describe the lesion under the term calcifying odontogenic cyst (COC) in 1962, even though the authors mentioned an earlier publication from 1960 describing a lesion with the same characteristics.[10] In 1971, the WHO described the lesion as a non-neoplastic cystic lesion and preferred to use the term COC.[69] In 1992, the WHO classified the lesion as an odontogenic tumour but the classification continued to be based on the monistic concept that all COCs (cystic and solid lesions) are neoplastic in nature.[70] One decade before the WHO 1992 classification, Praetorius et al. [20] were probably the first ones who introduced a classification based on a dualistic concept in which the lesions are divided into two entities, i.e. cyst and neoplastic.

This was followed by other several authors.[5,32,71,72] The WHO classification terminology was finally changed again in 2005, renaming the lesion as CCOT and DGCT.[1] From the first description of the lesion back in the 1960s until today, different terminologies and classifications have been proposed and practiced in the literature. In spite of various terminologies and classifications, discrepancies have prevailed over the usage of a terminology, and some authors still prefer to use older terminologies.[73] For this reason, the clinical and radiological aspects and the histological description of the lesions were thoroughly assessed by an expert in oral pathology, leading to the exclusion of the cases of two publications [6,68] that did not provide enough information to reassert the diagnosis of peripheral CCOT/DGCT.

Although the lesions were classified by the WHO [1] as variants of CCOTs, there is a debate whether CCOT and DGCT are variants of a single process or distinct cystic and neoplastic conditions. Still, we followed the WHO 2005 classification [1] and analyzed and compared peripheral DGCTs and CCOTs as distinct variants.

The present review of the literature revealed that the peripheral CCOT/DGCT is a rare lesion, with only 138 cases since the first description of the lesion in 1960 by Spirgi.[10] The great majority of the cases described appear as isolated case reports or small case series. Of the 138 cases described in the literature, 117 were more deeply analyzed in the present review, with only three recurrences reported. This suggests that the recurrence of peripheral CCOTs/DGCTs is unusual. It was observed an almost clear bimodal age distribution of the lesions, with the highest prevalence in the second and in the sixth/eighth decades of life. This was the exact same age distribution observed by Buchner et al.,[32] who also performed a literature review 25 years ago, but identified only 45 lesions at that time. It is interesting to note that the bimodal distribution of the cases according to age was not observed in the peripheral variant of DGCT. The peripheral DGCTs showed a higher incidence in later mean ages than its central counterpart (57.0 vs. 39.7 years) when compared to the results of a recent review,[74] and the same was observed for the cystic variant (41.8 vs. 30.3 years).[4] The peripheral lesions didn't show a sex predilection, but affected women at a higher age than men ($p = .048$). A previous review on central lesions observed that men and women tend to have the same mean age, with men tending to be slightly older.[4]

The lesions were predominantly found in the anterior region of the jaws. Moreover, they were more commonly observed in the mandible (65.2%), a pattern not observed for central lesions, which do not seem to have any particular predilection for either maxilla or mandible.[1,4]

The erosion of the underlying alveolar bone was not so commonly seen in peripheral CCOTs/DGCTs (about 20% of all lesions). The erosion may be so slight that it does not appear in radiographs and can only be detected during the surgical procedure.[54] This low rate may be related to the benign behaviour of these lesions.

Most of the studies reported simple excision with or without curettage of the underlying bone as the treatment of choice for peripheral CCOT/DGCT. As the recurrence rate was very low, a conservative treatment seems to be enough for the great majority of cases. The reported cases of recurrences occurred after 3 years. Therefore, an adequate follow-up of no less than 3 year should be considered.

The results of the present study have to be interpreted with caution because of its limitations. First, all included studies were retrospective reports, which inherently results in flaws. These problems are manifested by the gaps in information and incomplete records. Second, many of the cases have a short follow-up. This might have led to an underestimation of the actual recurrence rate, because a longer follow-up period can lead to an increase in the recurrence rate. However, it is hard to define what it would be considered a short follow-up period to evaluate the recurrence of CCOTs/DGCTs. Third, the great majority of the cases described were published as isolated case reports or small case series.

Conclusions

Peripheral CCOTs/DGCTs are rare lesions with a low rate of recurrence. The mean age of patients was 51.3 ± 23.4, with higher mean age for the DGCTs variant. The lesions were more prevalent in the mandible, anterior region of the jaws, and in the second, sixth and eighth decades, with an equal sexual distribution. About 20% of all lesions showed signs of underlying bone erosion, with a higher rate for DGCTs. Most of the lesions were treated by simple excision with or without curettage of the underlying bone. As the recurrence rate was very low, a conservative treatment seems to be enough for the great majority of cases.

Disclosure statement

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of this article.

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