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Odontogenic Cysts: Presentation of a Simplified Classification System

Malas V[1,2] and Rasmusson L[3,4]*

[1]*Department of Head-Neck and Plastic Surgery, Maxillofacial Surgery Section, Örebro University Hospital, Sweden*

[2]*Research Department, Örebro University, Sweden*

[3]*Department of Oral and Maxillofacial surgery, The Sahlgrenska Academy, University of Gothenburg, Sweden*

[4]*Maxillofacial Unit, Linköping University Hospital, Sweden*

Abstract

Objective: This study examined the prevalence, surgical treatment strategies, postoperative complications and recurrence rates of odontogenic cysts in the University Hospital in Örebro.

Material and Methods: Data were retrieved from patient's records of 234 patients who received the diagnosis odontogenic cyst during the years 2011-2021 at the maxillofacial surgery clinic in Örebro. The collected data covered patient gender and age, health declaration, presence of symptoms, tobacco use, diagnosis, localization of cyst, size of the cyst, radiographical examination, date of surgery, histopathological diagnosis, treatment, recurrence, reported complications and if hospitalization was needed.

Results: A total of 211 patients were included in the study. 87.7% of the patients were diagnosed with ICD-10 code K099 and 12.3% with K092. The most common odontogenic cyst was dentigerous cyst (25%), followed by the radicular cyst (20.5%) and the third most common cyst (20%) was the Odontogenic Keratocyst (OKC). The most common treatment reported was tooth extraction in combination with cyst enucleation (45.9%) followed by only enucleation (33.5%). Recurrence was reported in 17 of the cases and of these, 11 were OKC. Recurrence was most common the first three years. Postoperative complications were reported in 37% of the cases and the most common complications were postoperative infections.

Conclusion: The prevalence, surgical treatment strategies, postoperative complications and recurrence rates of odontogenic cysts in the University Hospital in Örebro is similar to data presented in other studies. A large variation in the diagnosis of cysts has been noted, which is why the authors have suggested a simplified classification system to improve the quality of future research and simplify the classification for the odontogenic cysts in the clinic.

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Correspondence:

Viktoria Malas, Department of Head-Neck and Plastic Surgery, Maxillofacial Surgery Section, Örebro University Hospital, Research Department, Örebro University, 701 85 Örebro, Sweden,

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Introduction

The definition of an odontogenic cyst is a cavity with epithelial lining with origin from the odontogenic epithelium. The odontogenic epithelium includes the reduced enamel epithelium, epithelial cell rest of Serres and the epithelial cell rest of Malassez [1].

Johnson et al. 2014 [2] concluded that the odontogenic cysts were 2.25 times more common than the odontogenic tumors. According to the World Health Organization (WHO), the classification of odontogenic tumors is mainly divided into malignant and benign tumors, based on their biologic behavior. In the 2022 edition of classification, the WHO presents cysts of the jaws without any subdivision. However, in some reports the authors prefer to present the odontogenic cysts as subdivisions such as inflammatory odontogenic cysts and developmental odontogenic cysts to simplify and emphasize the origin of the cysts [3]. The inflammatory cysts include the radicular and residual cysts and the developmental cysts include the gingival cyst, dentigerous cyst, Orthokeratinized OKC, lateral periodontal, calcified odontogenic, and glandular odontogenic cysts [3,4].

Johnson et al. [2] reported that the radicular cyst was the most common cyst found in the jaws. The dentigerous cyst and the previously coined odontogenic keratocystic tumor were the second most frequent odontogenic cysts with a prevalence of 20.6% and 11.7% respectively. Other than the odontogenic cysts, they also reported the incidence of the odontogenic tumors. According to

the presented literature, the ameloblastomas were the most common odontogenic tumors with a prevalence of 37.9% [2].

There are several reports that consider the radicular cyst as the most common cyst of the jaw [2,3,5]. A radicular cyst develop from peri-radicular inflammation as a consequence from tooth devitalization [3]. The epithelium of periapical cysts has its origin from the Malassez cell rests [4] and the typical radiographic finding is a unilocular radiolucency at the apex of the tooth. The radiolucency is often presented with well-defined borders and usually not larger than 1 cm in diameter [6]. The standard treatment for the radicular cyst is enucleation. It is possible for the lesion to resolve with endodontic treatment. If the lesion is not properly removed, it can develop into a residual cyst [4].

The residual cyst is a subtype of the radicular cyst and is characterized by a well-defined radiolucency located at a site of a previous tooth extraction if the radicular cyst was failed to be removed during the tooth extraction. The lateral radicular cyst is no longer a separate group but is classified as a radicular cyst with lateral localization [3].

The most common developmental odontogenic cysts are the dentigerous cyst and the Odontogenic Keratocyst (OKC) [2].

The dentigerous cyst, also called follicular cyst, are the second most common odontogenic cysts and can prevent tooth eruption due to a fluid that dilate the tooth follicle. It is usually present at the crown of a tooth that is either unerupted or under development [7-9]. The radiographic findings are usually a unilocular radiolucency in conjunction with an unerupted tooth. The lesion is often well-defined and origin from the Cemento-Enamel Junction [10].

The OKC is characterized as a locally aggressive lesion in the jaw with a high risk for growing and recurrence [11-13] In the 2022 edition of the WHO classification, the OKC is still classified as a cyst although it has earlier been classified as a tumor [3,14]. The typical radiographic features of the OKC are a unilocular radiolucent lesion with a smooth border. Presence of impacted tooth, displacement of adjacent tooth, root resorption and bone expansion can be seen with both OKC and ameloblastomas [15]. The incident of the OKC have been reported in patients between 10 and 30 years with a small predominance among men [16,17]. The most common site of occurrence is in the angle of the mandible and ramus of the mandible [18]. In a systematic review by Johnson et al. the recurrence rate is reported to 0% to 50% with differences depending on treatment strategy. Enucleation with supplementary measures had a 30.3% recurrence rate. Marsupialization with supplementary measures had a recurrence rate of 15.8%. In cases where enucleation with Carnoy's solution had been performed, the presented recurrence rate was 7.9% [19].

Ameloblastoma is the most common odontogenic tumor [2] in the jaws and has an incidence rate on 0.92 per million people per year [20]. The ameloblastomas originate from the odontogenic epithelium which includes the dental lamina, the reduced enamel epithelium and both Malassez and Serres epithelial rests [21]. The mean age is 34 and it is most common in the third decade of life. It has a slight predominance in the mandible *versus* the maxilla and the multicystic form is the most common type [20].

Study objective

The current study aims to report the incidence, recurrence rate and differences regarding surgical treatment strategies of odontogenic cysts and tumors in Örebro County. The secondary aim is to identify factors associated with the risk of developing odontogenic cysts and/or tumors in the jaws.

Materials and Methods

Design

The study was designed as a retrospective study. All patients who received the diagnosis odontogenic cyst (ICD code K099, K092) during the years 2011-2021 at the maxillofacial surgery clinic in Örebro was included in the study. Patients with following factors were excluded: Diagnose of an odontogenic cyst but without histopathological verification, diagnosed odontogenic cyst without treatment, surgery date prior or after 2011-2021.

Subjects and procedures

Data from patient's records was collected in the Maxillofacial Unit in Örebro University Hospital. For those patients who had more than one diagnosed cyst under this period, the first cyst to be surgically removed was included in the study. If the patient had an OKC and an additional diagnosis of radicular cyst or inflammatory tissue, the OKC was selected for inclusion.

Unidentified data were collected from patient charts. Each chart was opened only once; to prevent data from the same chart being recorded twice, a note was made in the chart that data had been extracted from it. The following data were retrieved from the hospital charts: Patient gender and age, health declaration, presence of symptoms, tobacco use, diagnosis, localization of cyst, size of the cyst, radiographical examination, date of surgery, histopathological diagnosis, treatment, recurrence, reported complications and if hospitalization was needed.

Statistics

Mean values, ranges and Standard Deviations (SDs) were calculated for the descriptive statistics. When comparing two groups Fisher's exact test were used for binary variables and Fisher's permutation test for numerical variables. Two-sided p-value was used for all analyses and $p<0.05$ was considered statistically significant.

When testing for postoperative complications and recurrence rate, 11 of the patients were not included in these tests as they had not undergone any surgery.

Ethics

The study was approved by the regional ethics committee in Uppsala (Dnr 2022-02119-01).

Results

In the period 2011-2021, 234 patients were diagnosed with an odontogenic cyst. A total of 23 patients were excluded:

- 7 patients had their surgery date outside the study interval
- 1 patient died before treatment
- 9 patients did not receive any treatment
- 2 patients only had teeth extracted and no histopathologic analysis
- 1 patient only had apicectomy done and no histopathologic analysis
- 1 patient did not have any cyst at the surgery and therefore

Table 1: A total of 211 patients were included in the study, of these 121 were men and 90 women.

		n (%)	Mean ± SD (range)
Female		90 (42.7)	
Male		121 (57.3)	
Age at surgery or examination		211	49.5 ± 19.6 (9-93)
Symptom	No	122 (61.0)	
	Yes	78 (39.0)	
Diseases	No	111 (55.5)	
	Yes	89 (44.5)	
Medications	No	107 (53.2)	
	Yes	94 (46.7)	
Allergies	No	134 (76.1)	
	Yes	42 (23.9)	
Smoking	No	113 (73.4)	
	Yes	41 (26.6)	
Snuffing	No	138 (89.6)	
	Yes	16 (10.4)	
Diagnostic code (ICD10)	K099	179 (87.7)	
	K092	25 (12.3)	
Localization	Maxilla right	27 (12.8)	
	Maxilla left	27 (12.8)	
	Mandible right	56 (26.5)	
	Mandible left	86 (40.8)	
	Maxilla front	8 (3.8)	
	Mandible front	7 (3.3)	
Size	≤ 2x2 cm	91 (77.1)	
	> 2x2 cm	27 (22.9)	
Radiograph	OPG	25 (12.3)	
	CBCT	63 (31.0)	
	OPG+CBCT	74 (36.5)	
	Computer tomography	11 (5.4)	
	Intraoral	17 (8.4)	
	OPG + Intraoral	13 (6.4)	
Year of operation		200	2016 ± 2.4 (2011-2022)
Keratocyst	No	159 (79.9)	
	Yes	40 (20.1)	
Histopathologic diagnosis			
Treatment	Enucleation	70 (33.5)	
	Fenestration	9 (4.3)	
	Extraction + enucleation	96 (45.9)	
	Fenestration + extraction	1 (0.5)	
	None	16 (7.7)	
	Retrograde root canal + enucleation	13 (6.2)	
	biopsy	4 (1.9)	
Recurrence	1-3 year	10 (4.7)	
	4-7 year	5 (2.4)	
	No year specified	2 (0.9)	
	No recurrence	183 (86.7)	
	No surgery	11 (5.2)	
Recurrence diagnosis	Keratocyst	11 (64.7)	
	None Keratocyst	3 (17.6)	
	Data not available	3 (17.6)	
Complications	No	126 (63.0)	
	Yes	37 (37.0)	
Hospitalization	No	194 (97.0)	
	Yes	6 (3.0)	

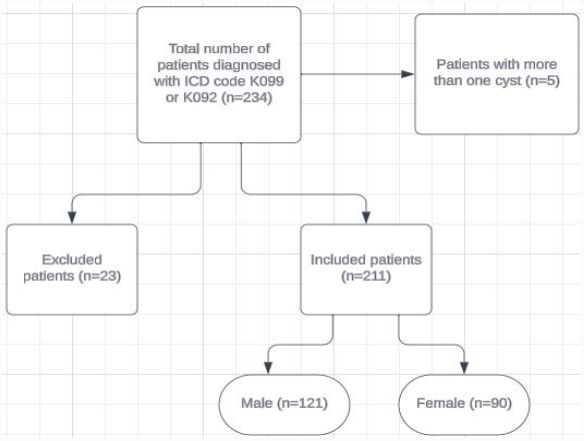

Figure 1: Showing total number of patients included in the study. For those patients with more than one cyst only one cyst per patient was included in the study.

no histopathologic analysis

- 1 patient moved to another city
- 1 patient had not yet received treatment when the data was collected.

Five patients had more than one diagnosed cyst. In one case, the patient had a diagnosed dentigerous cyst and a diagnose of inflammatory tissue. In this case, the dentigerous cyst was included. Another patient had two OKC diagnosed and the first OKC that was removed was included in the study. A third patient had an OKC, and inflammatory tissue diagnosed and the OKC was included in the study. A fourth patient had one OKC and one radicular cyst diagnosed and the OKC was included in the study. The fifth patient had two dentigerous cysts diagnosed and the first one that was surgically removed was included in the study.

A total of 211 patients were included in the study, of these 121 were men and 90 women (Table 1 and Figure 1). Of all patients, 78 patients reported symptoms at the time of examination such as bad taste, pericoronitis, swelling, pressure/tension, pain and discharge due to infection. Patients with symptoms had a higher risk of complications compared to those without symptoms, p=0.013 (Table 2). Of the patients included in the study, 44.5% stated that they had one or more systemic diseases and 46.7% were taking at least one prescribed medicine. 26.6% of the patients stated that they were smokers and 10.4% used snuff (Table 1).

A total of 87.7% of the patients were diagnosed with ICD-10

Table 2: Patients with symptoms had a higher risk of complications compared to those without symptoms.

	No recurrence N=183		Recurrence N=17		Two sided p-value	No reported complication N=126		Reported complications N=74		Two sided p-value
Gender (% female)	183	44.30%	17	35.30%	>0.30	126	40.50%	74	48.60%	>0.30
Age (at surgery or examination) (mean ± SD (range))	183	47.6 ± 19.2 (9-93)	17	56.6 ± 19.2 (19-84)	0.08	126	49.5 ± 21.7 (9-93)	74	46.5 ± 14.5 (10-73)	>0.30
Symptoms (% yes)	174	40.20%	15	26.70%	>0.30	119	31.90%	70	51.40%	0.013
Diseases (% yes)	174	45.40%	16	43.80%	>0.30	120	43.30%	70	48.60%	>0.30
Medication (% yes)	175	46.90%	16	50.00%	>0.30	124	44.40%	67	52.20%	>0.30
Allergies (% yes)	153	23.50%	14	21.40%	>0.30	109	22.90%	58	24.10%	>0.30
Snuffing (% yes)	137	10.20%	10	20.00%	>0.30	94	9.60%	53	13.20%	>0.30
Diagnosis (% K099)	178	90.40%	16	68.80%	0.043	123	84.60%	71	95.80%	0.023
Size of cyst (% up to 2 cm × 2 cm)	109	78.90%	9	55.60%	0.24	68	80.90%	50	72.00%	>0.30
Histopathologic diagnosis (% Keratocyst)	181	16.00%	17	64.70%	<0.001	124	22.60%	74	16.20%	>0.30
Recurrence (% yes)						126	9.50%	74	6.80%	>0.30
Complications (% yes)	183	37.70%	17	29.40%	>0.30					
Hospitalization (% yes)	183	3.30%	17	0.00%	>0.30	126	0.80%	74	6.80%	0.052
Position (% maxilla)	183	26.80%	17	47.10%	0.14	126	34.10%	74	18.90%	0.030>0.30
Smoking (% yes)	137	27.00%	10	10.00%	>0.30	94	27.70%	53	22.60%	>0.30
Radiograph OPG (% yes)	176	56.30%	16	37.50%	0.24	120	54.20%	72	55.60%	>0.30
Radiograph CBCT (% yes)	176	68.20%	16	68.80%	>0.30	120	63.30%	72	76.40%	0.083
Radiograph DT (% yes)	176	5.70%	16	6.30%	>0.30	120	5.80%	72	5.60%	>0.30
Radiograph Intraoral (% yes)	176	14.20%	16	18.80%	>0.30	120	18.30%	72	8.30%	0.085
Treatment Enucleation (% yes)	183	89.10%	17	88.20%	>0.30	126	87.30%	74	91.90%	>0.30
Treatment fenestration (% ja)	183	4.90%	17	5.90%	>0.30	126	5.60%	74	4.10%	>0.30
Treatment Extraction (% ja)	183	51.90%	17	11.80%	0.0024	126	41.30%	74	60.80%	0.011
No treatment (% yes)	183	4.40%	17	0.00%	>0.30	126	4.80%	74	2.70%	>0.30
Treatment retrograde root canal (% yes)	183	6.60%	17	0.00%	>0.30	126	7.90%	74	2.70%	0.23
Treatment biopsy (% yes)	183	1.60%	17	5.90%	>0.30	126	2.40%	74	1.40%	>0.30

Table 3: Most common odontogenic cyst.

		Patients with surgery (n=200)	
	All patients (n=211) (% of those reported diagnosis)	No recurrence	Recurrence (% of those with the same diagnosis)
Dentigerous cyst	50 (25.0)	48	2 (4.0)
Keratocyst	40 (20.0)	28	11 (28.2)
Radicular cyst	41 (20.5)	39	1 (2.5)
Other	69 (34.5)	65	3 (4.4)
No diagnosis reported	11	3	0

code K099 and 12.3% with K092. Patients diagnosed with K099 had a lower risk for recurrence in comparison with those diagnosed with K092, p=0.043 (Table 2) and a higher risk for complications compared to those diagnosed with K092, p=0.023 (Table 2). The most common odontogenic cyst was dentigerous cyst (25.0%), followed by the radicular cyst (20.5%) and the third most common cyst (20.0%) was the OKC (Table 3). Patients diagnosed with OKC had a higher recurrence rate $p \leq 0.001$ (Table 2). The left mandible was the most common localization of the cysts (40.8%) and the second most common localization was the right mandible (26.5%). The localization of cysts in the right and left maxilla was both 12.8%. Most cysts (77.1%) were smaller than 2 cm × 2 cm. The most common preoperative radiographic examination (36.5%) was a combination of panorama and CBCT (Table 1). Patients with a cyst in the maxilla had a lower risk of complications compared to those with a cyst in the mandible p=0.030 (Table 2).

The most common treatment reported was tooth extraction in combination with cyst enucleation (45.9%) followed by only enucleation (33.5%) (Table 1). Recurrence was most common the first three years (4.7%) and in 64.7% of the cases of the recurrence the diagnosis was OKC (Table 1). Patients treated with extraction had a lower risk of recurrence compared to those with treatment without extraction p=0.0024 (Table 2). Patients treated with extraction had a higher risk of complications compared to those who were treated without extraction p=0.011 (Table 2).

Postoperative complications were reported in 74 patients that had treatment for their cyst. Fifty-four of these had postoperative

Table 4: Current classification system of odontogenic cysts.

Diagnosis code	Collective name	Example	Excludes
K090	Odontogenic developmental cysts	Eruption cyst Dentigerous cyst Gingival cyst Lateral periodontal cyst	
K091	Non-Odontogenic developmental cysts in mouth and jaw region	Globulomaxillary cyst Incisive canal cyst Nasolabial cyst Nasopalatinal cyst	
K092	Other cysts in the jaw	aneurysmal hemorrhagic traumatic without further specification	Idiopatic bone cavity (Stafnes cyst) (K10.0)
K098	Other specified cysts in the mouth and jaw region not classified elsewhere	Dermoid-, epidermoid- and lymphoepithelial cyst of the mouth Epstein´s gem	
K099	Cyst in the mouth and jaw region, unspecified		

infections and the other 20 had other complications such as severe postoperative pain, sensory impairment, and alveolitis.

Discussion

The three most common cysts in this study were the dentigerous cyst (also known as follicular cyst in some literature), the radicular cyst and the OKC, which together accounted for 65.5% of all odontogenic cysts in this study sample. The study result is similar to other results from previous studies around the world. A 10-year retrospective study done on an Indian population concluded that these three cysts accounted for 74% of all odontogenic cysts [22]. Another study done on an English population presented similar study results [23]. A number of similar studies from different parts of the world have presented similar results [24-29].

Regarding gender, the present study found that the occurrence of cysts was more common in males (57.3%) compared to females (42.7%). The majority of results reported from previous studies have reported similar results with a predominance in men compared to women [22,24-26,30]. However, female dominance has been reported in a Brazilian study [26]. Studies have shown that men are more likely to ignore their oral health and have poorer oral hygiene habits than woman [31]. It is also more common with oral and facial trauma in men *vs.* woman [31,32]. These two factors could hypothetically explain the male predominance.

The left side of the mandible was the most common site (40.8%), followed by the right side of the mandible 26.5%. It was evenly distributed with the number of cysts in the upper jaw with 12.8% in both the right and left maxilla. Other studies have reported different results. Some authors have reported higher frequency in the maxilla *vs.* the mandible [23,25,26,30,33,34] while other authors have reported similar result as in the present study [22,27].

Seventy-eight patients (39%) reported symptoms prior to their surgery and according to our statistics they had an increased risk of developing postoperative complications such as postoperative infection. According to the authors, a possible explanation for this could be that the cysts have been secondarily infected and therefore the presence of bacteria has existed in the cyst cavity. In one study that reviewed OKC 46.1% of the patients reported symptoms such as pain, swelling and pus at their first visit [16]. Another study observed OKC over an 18-year period and concluded that swelling was the most common symptom [35]. There are multiple factors that have been shown to affect the wound healing [36]. This study result could possibly be explained by the presence of inflammatory tissue (microbial components) that affects the healing process. Previous studies have suggested poor macrophage function as an explanation for poor healing [37]. Another study discussed that the dysregulation of inflammatory cells can result in inflammatory disease states that impair the quality of healing [38].

Regarding the recurrence rate, the present study found that the OKC had more recurrences in comparison with other cysts, which agrees with the literature where it is described that the OKC have a high recurrence risk. One study reported 58.3% recurrence rate of OKC [39]. Blanas et al. reported 17% to 56% recurrence rate [40] and Johnson et al. reported recurrence rate up to 30.3% [19]. In the present study, the recurrence rate of the OKC was 28.2%. Factors such as histopathological features and surgical techniques have been suggested to explain the high recurrence rate for OKC. One study systematically reviewed recurrence rate in relation to treatment modalities and could not find any evidence to support the thesis [41].

Another study concluded that it is likely that parts of the basal layer of the epithelium of the oral mucosa cause recurrences [42]. A Chinese study that evaluated the recurrence rate for patients with surgical treatment of OKC concluded that treatment with enucleation alone have a higher recurrence rate and adjunctive treatment such as Carnoy's solution can decrease the recurrence rate [43].

Another article that studied the predictive factors of recurrence of OKC with focus on histopathological and immunohistochemical factors identified higher recurrence rate associated to factors such as presence of daughter cysts and epithelial islands [44]. Presence of involved teeth combined with cortical perforation has also been suggested to explain recurrence [45].

In the present study, extraction of involved teeth occurred to have a decreased risk for recurrence. Management of teeth involved in the cyst can be difficult to the surgeon [39]. In cases with supernumerary teeth and teeth with other obvious pathologies extraction may be a suitable option as a complement to enucleation of the cyst [39]. Some authors recommend extraction of teeth to reduce the risk of recurrence [44]. Zhao et al. reviewed 19 OKC and found three cases of recurrence that may be explained by incomplete epithelial removal around roots in the cyst cavity. They recommend removal of involved teeth in such cases [46]. Another aspect of management of involved teeth is to preserve masticatory performance after surgery. Tan et al. concluded that the number of lost teeth following surgery has an effect on patients' masticatory function and therefore in some cases teeth could be preserved when marsupialization was performed

in combination with secondary enucleation [47]. Published data regarding management of teeth involved within cystic lesions is limited and there are no clear guidelines for the surgeons. The need for further research is necessary.

Regarding the statistical analysis in Table 2, 49 tests were done. When 49 tests are done 3 (49 × 0.05 = 2.45) tests can become significant by chance even if there are no correlations, when considering $p<0.05$ as statistically significant (Type I error). Seven significant relationships were found. 5 at $p<0.05$ level, 1 at $p<0.01$ level and 1 at $p<0.001$ level. This means that the authors interpret the five $p<0.05$ with caution.

One interesting finding of the study is the variation in diagnosis of the odontogenic cysts. The study was carried out by searching for diagnosis codes in the medical record system used at the hospital in question. The International Statistical Classification of Diseases and Related Health Problems is the international classification that has existed for around 100 years. It is the World Health Organization (WHO) that has been responsible for this since 1948 [48]. ICD-10-SE is the Swedish version of the classification system and is used to be able to make overview statistical compilations and analyses within the health care system in Sweden according to the National Board of Health and Welfare.

As for cysts in the mouth and jaw region, these go under the ICD-10 code K090, which has five subgroups: K090, K091, K092, K098, and K099. In this study, the codes K099 and K092 were used to select patients for the study. This is considered a limitation of the study as one could imagine a larger patient base with an expanded search including the additional three subgroups (K090, K091, and K098). The reason why K099 and K092 were used in particular is because they are the most frequently used diagnoses for cysts before PAD verification in the department in question. Correct diagnostic codes are probably not used. Instead, it is likely that the code K099 is used as a general diagnosis code for odontogenic cysts, suggesting that clinician's misses diagnosing according to the previously described subgroups. It is likely that K092 is also incorrectly used as a non-specific diagnosis code since the definition "other cysts in the jaw" can be interpreted non-specifically.

An interesting finding in this study is that the different diagnosis codes (K099 and K092) were associated with different risk of recurrence and complication. The patients classified with the diagnosis code K099 had a lower risk of recurrence than those with K092 and a higher risk of complication than those with K092.

As it might be difficult to place OKC in the correct classification group, a possible explanation to this finding could be that OKC have been diagnosed both as K092 and K099 but predominantly K092, which would explain that K092 has an increased risk of recurrence. Regarding the risk of complications, it is difficult to draw any conclusions as cysts have probably been misdiagnosed. To improve the quality of future research work, it is important to clarify the classification for the odontogenic cysts and also that the surgeons are calibrated.

Because of this, the authors have created a schematic picture of the current classification system of odontogenic cysts in Sweden with the aim of making it easier for surgeons to make the correct diagnosis (Table 4) [49].

Conclusion

The prevalence, surgical treatment strategies, postoperative complications and recurrence rates of odontogenic cysts in the University Hospital in Örebro is similar to data presented in other studies. A large variation in the diagnosis of cysts has been noted, which is why the authors have suggested a simplified classification system to improve the quality of future research and simplify the classification for the odontogenic cysts in the clinic.

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