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RESEARCH ARTICLE

Odontogenic cysts: A 40- year retrospective clinicopathological study in an Iranian population

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ABSTRACT

Objectives: To focus on distribution of different histologic types of odontogenic cysts (OCs), patient gender and age, and cyst location in an Iranian population and compare results with others studies in different geographic locations and/or racial populations.

Materials and Methods: A 40-year retrospective study of 1189 patients with OCs that were diagnosed from 1971 to 2011 in the Department of Oral and Maxillofacial Pathology, Faculty of Dentistry, Mashhad, Iran, were presented in this study.

Results: In contrast to most of previous studies that have been reported inflammatory OCs as the most common type, in the current study, among cystic lesions of the jaws most were developmental in origin (62.30%). The most commonly diagnosed inflammatory and developmental odontogenic cysts were the periapical cyst (30.45%) and dentigerous cyst (26.80%) respectively. Developmental cysts were happened more in males whereas inflammatory types showed more tendencies to females. The patients' mean age was 27.20 ± 15.93 years, with a peak of occurrence in the second decade of life followed by the third decade. A high posterior mandibular incidence (41.85%) was observed, whereas some previous reports showed anterior maxilla predilection.The most common histological types of developmental cysts were aggressive [Dentigerous cyst (DC), odontogenic keratocyst (OKC) and calcifying odontogenic cyst (COC)]. Furthermore, these three varieties representing more than half of all studied cysts (59.30%) at our Service.

Conclusions: Knowledge of the incidence of odontogenic cysts and their clinicopathologic features, including most common location, gender and age distribution in different ethno-geographic backgrounds is necessary for the accurate diagnosis and differentiating aggressive lesions from other kinds of cysts.

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INTRODUCTION

Cyst is defined as a pathologic cavity with liquid, semiliquid or gaseous contents, enveloped by an epithelial tissue membrane.[1] Odontogenic cysts (OCs) constitute an important aspect of oral and maxillofacial pathology. They are relatively common lesions with different clinical behavior. Therefore, these lesions have been the focus of many studies.[2]

According to World Health Organization (WHO) International Classification, OCs are best classified as inflammatory and developmental. Inflammatory cysts develop as a result of inflammation, whereas the inciting factors that initiate the formation of developmental cysts remain unknown.[3]

Since OCs are the most important part of oral and maxillofacial cysts and there is little information in the English-language literature about the clinicopathologic features of OCs in Iranian population, we retrospectively investigated OCs in Iranian patients diagnosed at Department of Oral and Maxillofacial Pathology, Mashhad University of Medical Sciences over a period of 40 years.

The clinical signs and symptoms as well as biologic behavior of these lesions including recurrence, mode of growth, and malignant change differ by type of OCs. As some of these lesions represent variable behaviors, considering the set of criteria such as age, gender, and location of lesions could be useful for management of them.[1]

The clinicopathological features of these cysts vary with race and geographic location. Therefore, we focused on distribution of different histologic types of OCs, patient gender and age, and cyst location in an Iranian population and compared results with others studies in different geographic locations and/or racial populations.

MATERIALS AND METHODS

Case records from patients with OCs diagnosed between 1971 and 2011 were retrieved from the files of the department of Oral and Maxillofacial Pathology, Faculty of Dentistry, Mashhad, Iran. Hematoxylin and eosin stained sections were re-evaluated by three pathologists according to the 1992 WHO histologic classification.[4] The histologic criteria for the diagnosis of calcifying odontogenic cyst (COC) were those proposed by Buchner.[5]

The necessary clinical information, including patient age and gender, and cyst location was obtained from records. For cyst location the following scheme was used. The maxilla and mandible were divided into 4 anatomical regions, 2 on either side: anterior (from the midline to the distal surface of the canine), posterior (from the mesial aspect of the first premolar to the distal side of the third molar). Statistical analysis was performed using SPSS software. Data were analyzed by applying $\chi 2$ Test. Data were considered significant at $P < 0.05$.

RESULTS

A total of 1189 out of 10165 patients (11.70 %) were affected by OCs during the study.

Histologic type

The distribution of histological types and frequency of OCs is presented in Table 1. Of 1189 OCs, 448 (37.70%) were inflammatory, and 741 (62.30%) were developmental.

The most commonly diagnosed inflammatory and developmental OCs were the periapical cyst (30.45%) and DC (26.80%) respectively.

Table 1. Distribution of histologic type of 1331 odontogenic cysts

Cysts	n	%
Inflammatory cysts		
Radicular cyst (periapical cyst)	362	30.45
Residual cyst	77	6.50
Paradental cyst	9	0.75
Developmental cysts		
DC	319	26.80
OKC	298	25
COC	89	7.5
Eruption cyst	8	0.70
Botryoid odontogenic cyst	3	0.25
Orthokeratinized odontogenic cyst	4	0.35
Lateral periodontal cyst	9	0.75
Gingival cyst of infant	1	0.1
Glandular odontogenic cyst	10	0.85
Total	1189	100

Age

Patients ranged in age at the time of diagnosis from 1 to 90 years with a mean age of 27.20 ± 15.93. OCs had a peak of occurrence in the second decade of life followed by the third decade. Figure 1 depicts the distribution by age (decades) of OCs.

Developmental cysts were more common in the first two decades of the life, whereas with an increase in age, inflammatory cysts become the most frequently observed with statistically a significant difference ($p<0.05$).

Sex

Overall, OCs were diagnosed more frequently in males (55.25%). Table 2 shows the distribution of OCs by patients' gender. Developmental cysts were happened more in males (60.20%), whereas inflammatory types showed more tendencies to females (51.10%). Statistically significant differences were not observed between type of OCs and patients gender ($p=0.25$).

Location

Six hundred and ninety four cysts (58.34%) out of 1189 cysts occurred in the mandible. The posterior area of mandible was the

Table 2. Frequency of odontogenic cysts based on sex origin

	Male (n; relative %)	Female (n; relative%)
Inflammatory cysts		
Radicular cyst (periapical cyst)	179 (49.45)	183 (50.55)
Residual cyst	34 (44.15)	43 (55.85)
Paradental cyst	6 (66.70)	3 (33.30)
Developmental cysts		
DC	219 (60.50)	143 (39.50)
OKC	182 (61)	116 (39)
COC	52 (58.5)	37 (41.5)
Eruption cyst	4 (50)	4 (50)
Botryoid odontogenic cyst	1 (33)	2 (67)
Orthokeratinized odontogenic cyst	3 (75)	1 (25)
Lateral periodontal cyst	4 (44.5)	5 (55.5)
Gingival cyst of infant	0	1 (100)
Glandular odontogenic cyst	7 (70)	3 (30)

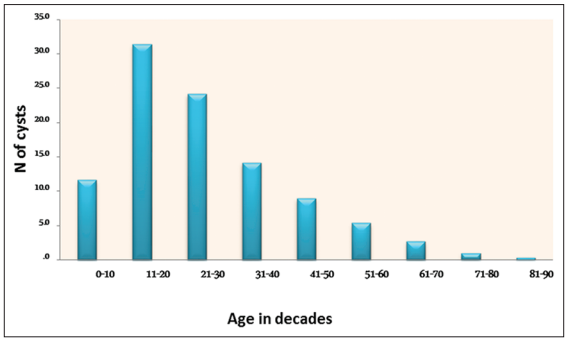

Figure 1. Distribution of odontogenic cysts in different decades of age

most frequent site (41.84%) followed by posterior maxilla (28.50%). OKC (25.9%) and periapical cyst (37.7%) were the most common cysts in posterior mandible and anterior maxilla respectively. The mandibular and maxillary distribution of OCs is presented in Table 3.

DISCUSSION

OCs are one of the most important lesions affecting the jaws. Since many of them possess similar histological, clinical and radiographic characteristics, the diagnosis should be based on careful examination of clinical and radiographic aspects, as well as histopathologic features.[3,6,7]

Unfortunately, no extensive data concerning inflammatory and developmental OCs have been reported on the Iranian population. In research literature on Iran, only Sharifian et al.[8] report has been published. This article evaluated OCs in 20-year period, whereas in the present study a 40-year retrospective clinicopathological study in an Iranian population was performed. It is important

to note that only Grossmann et al.[9] evaluated OCs in longer period than the present study. Comparative incidence of OCs in different published studies is presented in Table 4.

Table 3. Distribution of odontogenic cyst location

Location	N (relative %)
Posterior mandible	498 (41.85)
Anterior mandible	196 (16.50)
Posterior maxilla	339 (28.50)
Anterior maxilla	156 (13.15)

Complete clinical data is important for diagnosis and, consequently, the execution of retrospective studies.[7] In the present study, some records were incomplete which shows that some practitioners are careless when filling out clinical data. In order to guarantee the most reliable epidemiologic profile of OCs in our population, unclassified cysts due to insufficient clinical data were excluded from this study.

In the most recent classification, WHO considered OKC as "keratocystic odontogenic tumor".[10] However, Neville et al.[1] still regard this lesion as an odontogenic cystic lesion. In order to compare the this study results with previous reports, OKC is included in this study.

Table 4. Comparative incidence of odontogenic cysts in different published studies

Published series	Years covered	Country of study	Total number of cases	Most common histological types	Sex predilection	Most common involved area
Ioannidou et al.[4]	1977-1987	Athena	508	Radicular cyst (59.6) Residual cyst (28.4)	Male	Maxilla
Ledesma-Montes et al[10]	1986-1996	Mexico	304	Radicular cyst (38.8) DC (35.5)	Male	Anterior maxilla & Posterior Mandible
Arotiba et al[20]	1982-1996	Nigeria	65	Radicular cyst (61.9) DC (19)	Female	Maxilla
Grossmann et al[7]	1953-2003	Brazil	2812	Radicular cyst (61) DC (25.3)	Female	Anterior Maxilla
Jones et al.[13]	1975-2003	Britain	7121	Radicular cyst (52.3) DC (18.1)	Male	Anterior Maxilla
Ochsenius et al.[3]	1976-2004	Chile	2944	Inflammatory (50.7) DC (18.5)	Male	Anterior Maxilla

Contd...

Table 4. (Contiuned)

Published series	Years covered	Country of study	Total number of cases	Most common histological types	Sex predilection	Most common involved area
Meningaud et al[11]	1995-2005	France	695	Radicular cyst (53.5) DC (22.3)	Male	Posterior Mandible
El Gehani et al.[15]	1990-2005	Libya	326	Radicular cyst (68.1) DC (15)	Male	Anterior Maxilla
Tortorici et al [19]	1986-2005	Italy	1310	Radicular cyst (84.5) DC (11.4)	Male	Anterior Maxilla
Prockt et al.[14]	1985-2005	Brazil	680	Radicular cyst (72) DC (22)	Female	Anterior Maxilla
Núñez-Urrutia et al.[18]	1997-2006	Spain	418	Radicular cyst (50.2) DC (21.8)	Male	Posterior Mandible
Moctezuma-Bravo et al.[9]	1989-2006	Mexico	103	DC (56) OKC (33)	Female	Posterior Mandible
Acikgoz et al.[21]	2000-2008	Turkey	452	Radicular cyst (54.7) DC (26.6)	Male	Anterior Maxilla
Sharifian et al.[6]	1987-2007	Iran	1227	Radicular cyst (37.8) Dentigerous cyst (27.4)	Male	Mandible
Souza et al.[5]	1970-2007	Brazil	1019	Radicular cyst (61.4) DC (20.1)	Female	Posterior mandible
Butt et al[17]	1991-2010	Kenya	194	Radicular cyst DC (57.2)	Male	Undetermined
AlSheddi[16]	1984-2010	Saudi Arabia	470	Radicular cyst (64.3) DC (25.1)	Male	Mandible

Contd...

Table 4. (Contiuned)

Published series	Years covered	Country of study	Total number of cases	Most common histological types	Sex predilection	Most common involved area
Bataineh et al.[12]	1989-2011	Jordan	654	Radicular cyst (41.7) DC (24.8)	Male	Anterior maxilla
Present study	1971-2011	Iran	1331	Radicular cyst (24.94) DC (21.71)	Male	Posterior Mandible

Studies involving different populations demonstrated a higher frequency of inflammatory cysts.[2,13-15] In contrast to most of previous studies that have been reported inflammatory OCs as the most common type, a higher prevalence of developmental cysts was observed in the current study. A higher proportion of developmental cysts has also been showed in Mexican population.[11,12] It can be explained by the socioeconomic conditions of the population which may influence the relative frequency of inflammatory and developmental OCs.[7]

Radicular cysts are the most common inflammatory cysts.[16] In this study, radicular cysts were the most common type, accounting for 30.45% of all specimens followed by dentigerous cysts. This finding can be explained by the fact that,as in other countries, dental caries are observed in high percentage of the Iranian population. This finding being similar to most of previous studies.[2,7-9,12-15,17-23]

The second most frequently diagnosed lesion was DC (26.80%). This figure is in contrast with those published in other works from Athens and Turkey.[6,24] and Mexico[11] which showed residual cyst and OKC as the second most common cyst respectively.

In our series OKC was the third most common type of OC (25%). This is in contradiction to the results reported by Prockt et al.,[17] who reported residual cyst as the third most common type in Brazilian population.

In agreement with other studies,[11,12,19,23,24] the results of this study showed an increase in the incidence of OCs in second and third decades of life. In contrast, Bataineh et al. have shown a peak incidence in third and fourth decades of life in Jordanian population.[14]

Out of the total of 1189 biopsies in our department, there are 55.25% males and 44.75% females in agreement with previous reported findings,[2,8,12-15,18-22,24] whereas female predilection was reported in Brazilian and Nigerian population.[7,9,17,23]

In some of these studies it has been suggested that the greater occurrence in males is possibly associated with poor oral hygiene and more susceptibility to trauma than women as risk factors for cystic lesion formation.[24]

As mentioned before, developmental OCs were more common than inflammatory type in the present survey. It has been demonstrated that some OCs may show an aggressive behavior.[12,25-27] An important finding in our study was that the most common histological types of developmental cysts were aggressive (DC, OKC and COC). We found that these

cysts representing more than half of all OCs at our Service (59.30%). In our previous research,[6] we also showed local aggressive behavior of these cystic lesions by immunohistochemistry. Compared with other studies, a higher prevalence of glandular odontogenic cyst (GOC) as an aggressive cystic lesion was also observed in our study.

It should be noted that regarding COC classification and terminology, the clinical and histopathologic features of these cysts have generated disagreement and confusion. According to new WHO concept, COCs not only contain a neoplasm but also a cyst.[7] In order to permit comparison with previous studies, only cases belonging to the cystic variant were included in the study.

Since the GOC described as a distinct entity by Gardner et al. in 1988,[28] we revised the original diagnoses of hematoxylin/eosin-stained slides carefully before 1988. Some previous studies reported 2 cases of GOC, whereas this cyst has not been reported by other authors. Compared with a frequency of GOC reported in previous studies, we found a higher frequency for this cyst (10 cases) in our series. A study involving other Iranian population[8] has also shown a slightly higher prevalence of GOC (4 cases) than that observed in other countries. These data suggest that Iranian patients have a high risk of developing this aggressive cyst.We hypothesize developing cysts with aggressive behavior may be related to the racial characteristics of the population studied.

Therefore, diagnosis should be made by a well-trained pathologist becausethe clinical behavior of some OCs including DC, GOC, OKC, and COC is more aggressive than that of other types. Adequate treatment can be performed with accurate diagnosis, preventing their recurrence and more extensive tissue destruction.[3,12]

Regarding anatomic site, the posterior region of mandible (42.35%) was the most common location which is in agreement with the literature.[7,11,13,21] The anterior maxilla was an infrequent involved area in the current study, whereas studies from Libya,[18] Turkey,[24] Italy,[22] Jordan,[14] Chile,[3] Brazil,[9,16] and Britain[15] reported anterior maxilla as a most common site.

In conclusion, in our study population, cysts were mainly developmental in origin. The most frequent inflammatory odontogenic cyst was radicular cyst which also accounting for 30.45% of all specimens. The most common histological types of developmental cysts were aggressive representing more than half of studied cysts. Most of cystic lesions were observed in male patients and in the mandible, particularly posterior region. As the clinicopathologic features of these cystic lesions including patient age and sex, cyst location, and distribution vary with race and geographic location. Therefore, clinicopathologic features is necessary for the accurate diagnosis and differentiating aggressive lesions from other kinds of cysts.

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