Acta Odontologica Scandinavica. 2014; 72: 795–800

informa
healthcare

ORIGINAL ARTICLE

Prevalence of developmental odontogenic cysts in children and adolescents with emphasis on dentigerous cyst and odontogenic keratocyst (keratocystic odontogenic tumor)

NANNAN LI[1*], XING GAO[2*], ZIYUAN XU[3], ZHUO CHEN[1], LAIKUAN ZHU[3], JINRUI WANG[1] & WEI LIU[3]

[1]*School of Stomatology, Jilin University, Changchun, Jilin, China,* [2]*Department of Oral and Maxillofacial Surgery, XiangYa Hospital, Central South University, Changsha, Hunan, China, and* [3]*Shanghai Key Laboratory of Stomatology, Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, Shanghai, China*

Abstract

Objective. To investigate the incidence and prevalence of developmental odontogenic cysts in children and adolescents and compare the features of the two most common types, dentigerous cyst and keratocystic odontogenic tumor (KCOT). ***Study design.*** A retrospective review in a series of 369 patients with all histological diagnoses of developmental odontogenic cysts in children (≤12 years) and adolescents (13–18 years) was conducted. ***Results.*** Among these, 361 (97.8%) patients were diagnosed as dentigerous cyst ($n = 281$) and KCOT ($n = 80$), with the male-to-female ratios of dentigerous cyst and KCOT both being 2:1. The average age of the patients with KCOT was older than that of those with dentigerous cyst (14.7 years vs 11.8 years, $p < 0.001$). Dentigerous cyst (59.1%) was more common in children, but KCOT (78.8%) was more common in adolescents ($p < 0.001$). Dentigerous cyst (57.6%) predominantly located on the maxilla, but KCOT (60.3%) predominantly located on the mandible ($p = 0.010$). ***Conclusions.*** Adolescent patients with lesions located on the mandible would favor KCOT over dentigerous cyst. This study aids in better knowledge of the prevalence of developmental odontogenic cysts in a large pediatric population, and shows that a well-supported early diagnosis is indispensable for a more adequate treatment.

Key Words: *developmental odontogenic cysts, dentigerous cyst, keratocystic odontogenic tumor, pediatrics*

Introduction

Odontogenic cysts are the common osseous-destructive lesions affecting the jaws and they classified into developmental or inflammatory cysts based on their origin and pathogenesis. In children there is a relatively high rate of developmental cysts, whereas in adults inflammatory cysts are more common [1,2]. Dentigerous cyst is the most common developmental odontogenic cyst of the jaw bones and rarely recurs. Keratocystic odontogenic tumor (KCOT), formerly known as odontogenic keratocyst and primordial cyst, is classified into the group of developmental odontogenic cysts. KCOT is a locally aggressive benign odontogenic lesion with a relatively high recurrence potential. The other uncommon developmental odontogenic cysts include glandular odontogenic cyst, eruption cyst, gingival cyst of infant and lateral periodontal cyst [2,3].

Cystic lesions in the jaws share similarities in clinical and radiographic as well as symptomatology, especially between the dentigerous cyst and KCOT [4]. Radiographically, a dentigerous cyst usually presents as a unilocular and pericoronal radiolucent lesion of an unerupted or impacted tooth in the jaws. The radiographic picture alone is insufficient for definite diagnoses of DCs, because similar radiographic pictures can be found in other odontogenic lesions such as KCOT. The radiographic aspect of the KCOT, when involving the crown of an enclosed tooth, can simulate a dentigerous cyst. Therefore, the final pathologic diagnosis is based on microscopic examination of biopsy or surgical excision specimens.

Correspondence: Jinrui Wang, School of Stomatology, Jilin University, 1500 Qinghua Road, Changchun 130021, China. E-mail: linannan1734@126.com; or Wei Liu, Shanghai Key Laboratory of Stomatology, 639 Zhizaoju Road, Shanghai 200011, China. liuweb@hotmail.com.
*These authors contributed equally to this work.

(Received 3 August 2013; accepted 15 March 2014)

ISSN 0001-6357 print/ISSN 1502-3850 online
DOI: 10.3109/00016357.2014.913192

A review of the literature reveals that limited information regarding pediatric patients hidden away in several studies on developmental odontogenic cysts of all age-range patients were reported [5–23]. The issue of the prevalence and distribution of developmental odontogenic cysts has seldom been addressed in the Chinese population. Also, hardly any reports are available in the literature focusing on investigating the relationship between dentigerous cyst and KCOT in children and adolescents.

We, therefore, retrospectively reviewed a large series of 369 patients with all histological diagnoses of developmental odontogenic cysts in children and adolescents from eastern China to investigate the incidence and prevalence of developmental odontogenic cysts and compare the features of dentigerous cyst and KCOT in the current hospital-based study over a 20-year period.

Materials and methods

All the medical records of patients with the clinical and pathologic diagnosis of developmental odontogenic cyst from 1993–2012 in a computerized diagnostic index database of Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, were retrieved and reviewed retrospectively. Records of children (≤12 years) and adolescents (13–18 years) were selected. The tissue specimen of developmental odontogenic cyst was fixed in formalin, embedded in paraffin, and processed for routine histopathological examination from hematoxylin-eosin staining sections. The WHO criteria [1,2] were used when examining the histopathology of the sections. The patients with nevoid basal cell carcinoma syndrome were excluded in this study. If a patient had recurrent cysts, just the first diagnosis was included in the study. Information regarding patient's age, gender and location of lesion at the time of the initial diagnosis were documented in detail. The comparative and logistic regression analysis of dentigerous cyst and KCOT was performed to evaluate the parameters that would favor KCOT over a dentigerous cyst. This study was approved by the local institutional review board.

A descriptive analysis was performed on demographic parameters. Statistical analysis was carried out with the Chi-square test and Fisher's exact test among qualitative variables and the Student's *t*-test among quantitative variables. Logistic regression was applied to evaluate odds ratio (OR), indicative of being KCOT over dentigerous cyst. OR with 95% confidence interval (CI) and *p*-values were reported. All the tests were two-sided and p-values < 0.05 were considered statistically significant.

Results

Distribution of developmental odontogenic cysts

Table I shows the diagnosis and the relative frequency of developmental odontogenic cysts in pediatric populations between the current study and selected references from different countries.

A total of 369 patients in children and adolescents with developmental odontogenic cysts were identified for this study during a 20-year period. Among these, 361 (97.8%) patients were diagnosed as dentigerous cyst ($n = 281$) and KCOT ($n = 80$). Figures 1 and 2 show representative radiographic features of dentigerous cyst and KCOT, respectively. Figure 3 shows representative histological features of dentigerous cyst and KCOT.

In addition, six cases of glandular odontogenic cyst were males with an average age of 12.8 years (range = 6–18), one case (a 1-year-old male) with eruption cyst and one case (a 2-year-old female) with gingival cyst of infant were found; and no case of lateral periodontal cyst was found in this study.

Table I. The relative frequency of developmental odontogenic cysts in pediatric populations between the current study and selected references from different countries.

Country	China, current study	UK [6], 2006	Chile [7], 2007	Brazil [8], 2010	Turkey [9], 2012
Age range, years	1–18	1–16	0–15	0–16	2–17
Diagnosis (*n*, %)					
Dentigerous cyst	281 (76.2)	160 (59.7)	240 (68.2)	96 (89.7)	128 (66.0)
KCOT	80 (21.7)	100 (37.3)	98 (27.8)	6 (5.6)	64 (33.0)
Glandular odontogenic cyst	6 (1.6)	0	3 (0.9)	0	0
Eruption cyst	1 (0.3)	8 (3.0)	10 (2.8)	3 (2.8)	1 (0.5)
Gingival cyst of infant	1 (0.3)	0	0	1 (0.9)	0
Lateral periodontal cyst	0	0	1 (0.3)	1 (0.9)	1 (0.5)
Total	369	268	352	107	194

KCOT, keratocystic odontogenic tumor.

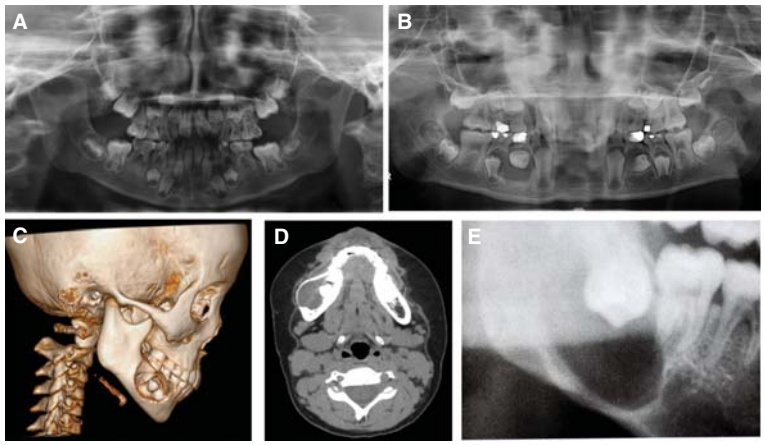

Figure 1. Representative radiographic features of a dentigerous cyst. Panoramic radiograph shows the dentigerous cysts in the (A) left maxilla and (B) right mandible. (C) Three-dimensional and (D) axial section of computed tomographic scan. (E) Dental radiograph.

Comparison of dentigerous cyst and KCOT

Table II shows the demographic features and location of dentigerous cyst and KCOT. Among the 281 patients with a dentigerous cyst, there were 166 children and 115 adolescents. These patients were 187 males and 94 females (ratio M:F = 2:1). The maxilla (160 cases) was more commonly involved than the mandible (118 cases) by dentigerous cysts. Of the 80 patients with KCOT, there were 17 children and 63 adolescents. These patients were 53 males and 27 females (ratio M:F = 2:1). The mandible (41 cases) was more commonly affected than the maxilla (27 cases) by KCOTs and there were nine cases of KCOT located at both the maxilla and mandible.

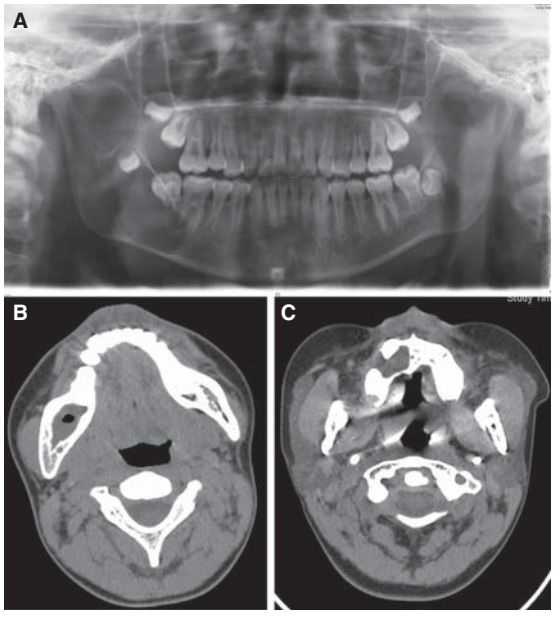

Figure 2. Representative radiographic features of keratocystic odontogenic tumor (KCOT). (A) Panoramic radiograph shows a KCOT in the right mandible. Computed tomographic scan shows KCOTs in the right (B) mandible and (C) maxilla.

To define the differences in demographic parameters between dentigerous cyst and KCOT, a comparative analysis was performed. The average age of the patients with dentigerous cyst was 11.8 years compared with those with KCOT at 14.7 years (Student's *t*-test, $p < 0.001$). Dentigerous cysts were more common in children (59.1%) and KCOTs were more common (78.8%) in adolescents (Fisher's exact test, $p < 0.001$). A significant difference in location was also observed (Fisher's exact test, $p = 0.010$), whereas a difference in gender was not observed between dentigerous cyst and KCOT.

Considering that dentigerous cyst shares some morphology similarity to KCOT, both clinically and radiographically, distinguishing KCOT from the cystic lesions has some difficulties. To evaluate the parameters that would favor KCOT over a dentigerous cyst, demographic parameters were analyzed by using the logistic regression model.

In univariate analysis, adolescence and location were significantly indicative for the likelihood of KCOT over dentigerous cysts. The likelihood of the patient being adolescent was higher (OR = 5.35; 95% CI = 2.98–9.61; $p < 0.001$) than being a child. The likelihood of lesion located on the mandible was higher (OR = 2.06; 95% CI = 1.20–3.54; $p = 0.009$) than that located on the maxilla. To further assess the influence of each factor, we did multivariate analysis. Adolescence and location retained statistical significance. The likelihood of the adolescent patient was higher than the child (adjusted OR = 5.31; 95% CI = 2.83–9.96; $p < 0.001$). The likelihood of the lesion being located on the mandible was higher than it being located on the maxilla (adjusted OR = 2.18; 95% CI = 1.24–3.85; $p = 0.007$).

Discussion

Developmental odontogenic cysts are probably related to the fact that during the pediatric age period

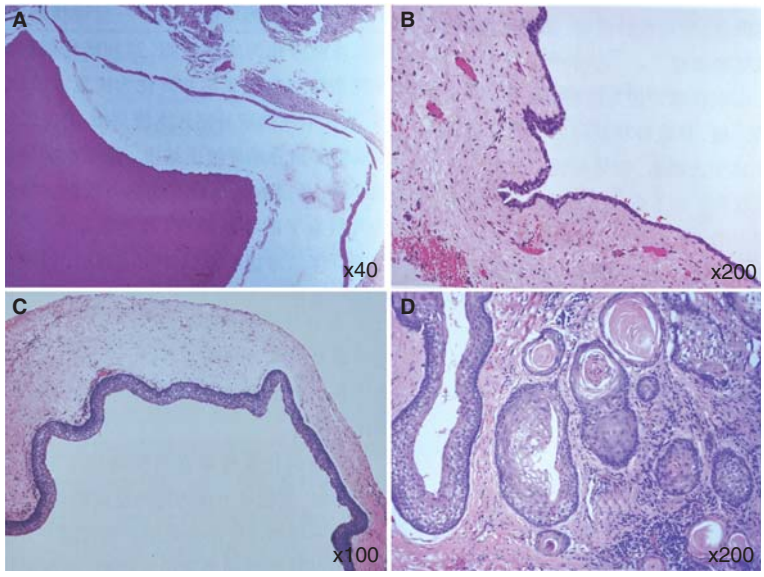

Figure 3. Representative histological features. (A) Dentigerous cyst shows the cystic lesion encloses a dental crown. (B) Dentigerous cyst shows cystic lining with thin non-keratinized stratified squamous epithelium. (C) Keratocystic odontogenic tumor (KCOT) shows a thin regular parakeratinized stratified squamous epithelium with basal palisading. (D) Daughter cysts and epithelial budding formation are also frequently observed in KCOT.

the jaws are involved in profound developmental processes. These include growth of the maxillofacial skeleton and development of the primary and permanent dentition, all of which can be associated with cyst formation. Dentigerous cyst and KCOT were the two most common types, accounting for over 95% of developmental odontogenic cysts in pediatric patients [5–9].

The correct diagnosis of dentigerous cyst and KCOT is necessary and important for correct surgical treatment and the institution of adequate follow-up. Therefore, knowledge of the incidence of dentigerous cyst and KCOT and their more common location of presentation and age distribution will help practitioners to determine a more likely correct clinical early diagnosis. The current study attempts to perform a descriptive analysis of 369 cases of developmental odontogenic cysts and to assess the relationship between dentigerous cyst and KCOT in a large series of dentigerous cysts ($n = 281$) and KCOT ($n = 80$) in children and adolescents from eastern China during the same 20-year period.

We found that dentigerous cysts and KCOT were the two most frequent developmental odontogenic

Table II. Comparison of dentigerous cyst and keratocystic odontogenic tumor (KCOT).

				Univariate LRA		Multivariate LRA	
Patients	Dentigerous cyst ($n = 281$)	KCOT ($n = 80$)	*p*-value	OR (95% CI)	*p*-value	OR (95% CI)	*p*-value
Age (years)							
Mean (SD)	11.8 (3.3)	14.7 (2.6)	<0.001				
Range	1–18	10–18					
Age group			<0.001				
Children	166 (59.1)	17 (21.2)		1.00 (ref)		1.00 (ref)	
Adolescents	115 (40.9)	63 (78.8)		5.35 (2.98–9.61)	<0.001	5.31 (2.83–9.96)	<0.001
Gender			1.00				
Female	94 (33.5)	27 (33.8)		1.00 (ref)			
Male	187 (66.5)	53 (66.2)		0.99 (0.58–1.67)	0.960		
Location*			0.010				
Maxilla	160 (57.6)	27 (39.7)		1.00 (ref)		1.00 (ref)	
Mandible	118 (42.4)	41 (60.3)		2.06 (1.20–3.54)	0.009	2.18 (1.24–3.85)	0.007
Unknown	3	3					

LRA, logistic regression analysis; OR, odds ratios; CI, confidence interval.
*There were nine cases of KCOT located at both maxilla and mandible.

cysts in children and adolescents, which constituted 97.8%, in agreement with over 95% reported in the literature [5–8]. In a comparison of our findings with those of other studies, the frequency of aggressive KCOT in China was higher than that in Brazil, but lower than that in other countries [5–8]. This result implies that the Chinese pediatric population may have a higher risk of developing aggressive cystic lesions than the Brazilian pediatric population.

We observed the average age of dentigerous cyst and KCOT, respectively, at the diagnosis was similar to that reported in the UK and Brazil [5,7]. Dentigerous cyst in the males obviously outnumbered the females in our study, but a marked female predilection was found in the UK [5]. For KCOT, a male predominance was found in our study, in accordance with the previous reports [5–7]. With regard to anatomic location of the jaws, little information exists in a pediatric population. Our study showed that dentigerous cyst predominantly located on the maxilla, but KCOT predominantly located on the mandible.

We evaluated the parameters that would favor KCOT over dentigerous cyst using logistic regression analysis. This will help practitioners to determine a more likely correct clinical early diagnosis. We found that the likelihood of KCOT over dentigerous cyst in the adolescent patient was higher than in the child and the lesion located on the mandible was associated with a higher neoplasia risk compared with maxilla.

In summary, the current study provides better knowledge of the prevalence of developmental odontogenic cysts in the largest retrospective series of pediatric patients among those published in the literature [5–8]. The demographic features and location of dentigerous cyst and KCOT in eastern China were elucidated, and our data indicated that adolescent patients with lesions located on the mandible were significantly indicative for being KCOT over dentigerous cysts. The knowledge of the incidence and distribution of pediatric developmental odontogenic cysts and their relationship constitute key aspects to provide a well-supported early diagnosis.

Acknowledgments

We thank Dr Jiang Li (Department of Oral Pathology, Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, Shanghai, China) for support and help in getting the medical records. This study was supported by the National Natural Science Foundation of China (81302358) and Changchun Science and Technology Project (12SF92).

***Declaration of Interest*:** The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

References

[1] Kramer IRH, Pindnorg JJ, Shear M. WHO International histological classification of tumors: histological typing of odontogenic tumors. 2nd edition. Springer-Verlag; Berlin, Germany: 1992.

[2] Barnes L, Eveson JW, Reichart P, Sidransky D. World Health Organization classification of tumors: pathology and genetics of head and neck tumors. IARC Press; Lyon, France: 2005.

[3] Butt FM, Ogeng'o J, Bahra J, Chindia ML. Pattern of odontogenic and nonodontogenic cysts. J Craniofac Surg 2011;22: 2160–2.

[4] de Avila ED, de Molon RS, Massucato EM, Hochuli-Vieira E. Relationship between the prevalence of the dentigerous cyst and the odontogenic keratocyst tumor and the current etiologic hypothesis. J Craniofac Surg 2009;20:2036–40.

[5] Jones AV, Craig GT, Franklin CD. Range and demographics of odontogenic cysts diagnosed in a UK population over a 30-year period. J Oral Pathol Med 2006;35:500–7.

[6] Ochsenius G, Escobar E, Godoy L, Peñafiel C. Odontogenic cysts: analysis of 2,944 cases in Chile. Med Oral Patol Oral Cir Bucal 2007;12:e85–91.

[7] de Souza LB, Gordón-Núñez MA, Nonaka CF, de Medeiros MC, Torres TF, Emiliano GB. Odontogenic cysts: demographic profile in a Brazilian population over a 38-year period. Med Oral Patol Oral Cir Bucal 2010;15: e583–90.

[8] Tekkesin MS, Olgac V, Aksakalli N, Alatli C. Odontogenic and nonodontogenic cysts in Istanbul: analysis of 5088 cases. Head Neck 2012;34:852–5.

[9] Pechalova PF, Bakardjiev AG, Beltcheva AB. Jaw cysts at children and adolescence: a single-center retrospective study of 152 cases in southern Bulgaria. Med Oral Patol Oral Cir Bucal 2011;16:e767–71.

[10] Ledesma-Montes C, Hernández-Guerrero JC, Garcés-Ortíz M. Clinico-pathologic study of odontogenic cysts in a Mexican sample population. Arch Med Res 2000;31:373–6.

[11] Mosqueda-Taylor A, Irigoyen-Camacho ME, Diaz-Franco MA, Torres-Tejero MA. Odontogenic cysts. Analysis of 856 cases. Med Oral 2002;7:89–96.

[12] Koseoglu BG, Atalay B, Erdem MA. Odontogenic cysts: a clinicalstudy of 90 cases. J Oral Sci 2004;46:253–7.

[13] Meningaud JP, Oprean N, Pitak-Arnnop P, Bertrand JC. Odontogenic cysts: a clinical study of 695 cases. J Oral Sci 2006;48:59–62.

[14] Grossmann SM, Machado VC, Xavier GM, Moura MD, Gomez RS, Aguiar MC, et al. Demographic profile of odontogenic and selected nonodontogenic cysts in a Brazilian population. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007;104:e35–41.

[15] Tortorici S, Amodio E, Massenti M, Buzzanca ML, Burruano F, Vitale F. Prevalence and distribution of odontogenic cysts in Sicily: 1986-2005. J Oral Sci 2008;50:15–18.

[16] Prockt AP, Schebela CR, Maito FD, Sant'Ana-Filho M, Rados PV. Odontogenic cysts: analysis of 680 cases in Brazil. Head Neck Pathol 2008;2:150–6.

[17] Avelar RL, Antunes AA, Carvalho RW, Bezerra PG, Oliveira Neto PJ, Andrade ES. Odontogenic cysts: a clinicopathological-study of 507 cases. J Oral Sci 2009;51:581–6.

[18] Nuñez-Urrutia S, Figueiredo R, Gay-Escoda C. Retrospective clinicopathological study of 418 odontogenic cysts. Med Oral Patol Oral Cir Bucal 2010;15:767–73.

[19] Sharifian MJ, Khalili M. Odontogenic cysts: a retrospective study of 1227 cases in an Iranian population from 1987 to 2007. J Oral Sci 2011;53:361–7.

[20] Butt FM, Ogeng'o J, Bahra J, Chindia ML. Pattern of odontogenic and nonodontogenic cysts. J Craniofac Surg 2011;22: 2160–2.

[21] Manor E, Kachko L, Puterman MB, Szabo G, Bodner L. Cystic lesions of the jaws – a clinicopathological study of 322 cases and review of the literature. Int J Med Sci 2012;9:20–6.
[22] Açikgöz A, Uzun-Bulut E, Özden B, Gündüz K. Prevalence and distribution of odontogenic and nonodontogenic cysts in a Turkish population. Med Oral Patol Oral Cir Bucal 2012; 17:e108–15.
[23] Lin HP, Wang YP, Chen HM, Cheng SJ, Sun A, Chiang CP. A clinicopathological study of 338 dentigerous cysts. J Oral Pathol Med 2013;42:462–7.