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ORIGINAL ARTICLE

Reclassification of odontogenic keratocyst as tumour. Impact on the odontogenic tumours prevalence

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AIM: The aim of this study was to establish the impact of the redefinition and reclassification of odontogenic keratocyst (OKC) as a tumour on the prevalence of odontogenic tumours (OT).
METHODS: We revised 15 435 files of a teaching head and neck histopathology service in the time period from January 1981 to December 2008 and 478 cases of OT were selected. The 342 cases from 1981 to 2004 were classified according 1992 to the World Health Organization (WHO)-classification (excluding keratocystic OT) while the 136 cases from 2005 onwards were classified according to the 2005 WHO-classification (including keratocystic OT). Age and gender were obtained from medical records. The frequency distribution and prevalence of OT from each periods of time were compared. A chi-square test was performed ($P < 0.05$ 95% confidence interval).
RESULTS: The prevalence of OT increases 92% in the 2005–2008 period; from 2.6% (1981–2004 period) to 5% (2005–2008 period) (*P* 0.000).The most frequent OT in the 1981–2004 period was odontoma (45% of all OT) while in the 2005–2008 period was Keratocystic Odontogenicv Tumour (38.9%). Conclusions. The redefinition of OKC as a tumour produced an increase in the frequency and prevalence of OT.
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Keywords: odontogenic keratocyst; keratocystic odontogenic tumour

Introduction

In 2005, the World Health Organization (WHO) redefined the odontogenic keratocyst (OKC) as a result of its biological behaviour, as a benign tumour of odontogenic origin. They named it keratocystic odontogenic tumour (KCOT) and included it in the group of benign odontogenic tumours (OT) derived from odontogenic epithelium with mature, fibrous stroma without odontogenic ectomesenchyme (Barnes *et al*, 2005). Regardless of the redefinition and reclassification of KCOT aroused controversy in different oral pathologist groups (Reichart and Philipsen, 2006; Madras and Lapointe, 2008), the impact of the redefinition and reclassification of KCOT on the prevalence and epidemiological profiles of OT it is not known. Therefore the principal objective of this report is to establish the frequency distribution and prevalence of OT before and after 2005 using an archive of a Mexican teaching service of head and neck histopathology.

Material and methods

The files of the Histopathology service of the Dental School, National Autonomous University of México were revised from January 1981 to December 2008. This histopathology service is the most important centre of diagnosis of head and neck pathology in all the country (México). All the cases with diagnosis of odontogenic cysts (OC) or OT were identified and selected.

To be included in this study, the cases should have biological material imbedded in paraffin and histological slides. If necessary, additional histological slides were obtained (5 μm) and stained with Haematoxylin and Eosin technique. All the selected cases were reviewed by two head and neck pathologist (DQR and FTR) and classified according to the 1991 WHO-classification to 1981–2004 cases (Kramer *et al*, 1992); and the 2005 WHO-classification (Barnes *et al*, 2005) to 2005 onwards cases. Demographical data (age at moment of diagnosis and gender) were obtained from medical records.

The prevalence of OT was established with regard to the total of biopsies per year. The frequency distribution and prevalence of OT from January 1981 to December 2004 were compared with frequency distribution and prevalence of OT obtained from January 2005 to December 2008. To statistical purposes, a chi square test was performed ($P < 0.05$ 95% confidence interval)

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using the EPI INFO 3.4.3 software package (Atlanta, GA, USA) (CDC).

Results

During the time period of our study (from January 1981 to December 2008), 15 435 files were revised (Table 1). From the 12 729 files analysed from 1981 to 2004, 1113 were OC while 342 were OT. The most frequent OC was the radicular cyst with 503 cases (45.1%) followed by dentigerous cyst with 385 cases (34.5%) and OKC with 213 cases (19.1%).On the other hand, OT were distributed as follows: 154 cases (45%) were odontomas; 76 (22%) were ameloblastomas; and 51 (14.9%) had a diagnosis of odontogenic myxoma. From January 2005 to December 2008, 2706 files were analysed, 209 were OC and 136 were OT. The most frequent OC was the radicular cysts with 119 cases (56.9%), followed by dentigerous cyst with 90 cases (43%). In regard to OT, the most frequent was KCOT with 53 cases (38.9%) followed by odontoma with 42 cases (30.8%) and ameloblastoma with 25 cases (18.3%). The frequency distribution of all OC and OT is shown in Table 2.

The prevalence of OT from 1981 to 2004 was 2.6%, while the prevalence of OT from 2005 to 2008 was 5%. This represents an increase in the prevalence of OT of 92% ($P = 0.000$). However, if the KCOT is excluded in the 2005–2008 period, the prevalence of OT is 3%, very similar to the 1981–2004 prevalence. On the other hand the prevalence of OC was very similar in both time periods: 8.7% (1981–2004) *vs* 7.7% (2005–2008) ($P > 0.05$). In the period 1981–2004, the prevalence of OT was very similar in both genders: 2.6% in female and 2.7% in males. In the period 2005–2008 a lightly male predominance in the OT prevalence was observed: 5.4% in males and 4.7% in females.

Discussion

Our results show that the redefinition of OKC as a neoplasia and its latter reclassification as a benign tumour derived from odontogenic epithelium with mature, fibrous stroma without odontogenic ectomesenchyme (Barnes *et al*, 2005) impacted the prevalence of OT. The prevalence of OKC varies from 11.6% to 19.1% (Ochsenius *et al*, 2002, 2007; Meningaud *et al*, 2006; González-Alva *et al*, 2008). In the present report, the prevalence of OKC from 1981 to 2004 was 19.1% and this OC occupied the 3rd place in the frequency distribution of all OC. These data agree with the previous one reported on Mexicans and other populations (Mosqueda Taylor *et al*, 2002; Jones *et al*, 2006; Habibi *et al*, 2007).

Although it has recently been suggested that ameloblastoma is the most frequent OT (Ledesma-Montes *et al*, 2007), it has been established that a distinct geographic variation of OT exists (Sriram and Shetty, 2008). However, it is not known if the geographical variation will be conserved when KOCT is included in the epidemiological studies of OT. In Asia, including India, the most frequent OT is the ameloblastoma, followed by adenomatoid OT and by odontogenic myxomas (Odukoya, 1995; Lu *et al*, 1998; Okada *et al*, 2007; Sriram and Shetty, 2008). In Americans, including Mexicans, the most common OT is odontoma followed by the ameloblastoma and odontogenic myxoma (Daley *et al*, 1994; Mosqueda-Taylor *et al*, 1997; Buchner *et al*, 2006; Guerrisi *et al*, 2007). In the present report, from 1981 to 2004 we obtained a prevalence of OT of 2.6%, and the same frequency distribution: odontoma was the most frequent OT, followed by ameloblastoma; and odontogenic myxoma. However, the distribution changes radically from 2005 onwards. When the KCOT was included into OT, they displaced to Odontoma to

Table 2 Frequency distribution of odontogenic cysts and odontogenic tumours in a Mexican sample in regard to different classifications

		1981–2004 $N = 12\,729$, n (%)	*2005–2007* $N = 2\,706$, *N* (%)
Odontogenic cyts	Total	1113 (100)	209 (100)
	Radicular	503 (45.1)	119 (56.9)
	Dentigerous	385 (34.5)	90 (43)
	OKC	213 (19.1)	–
	Paradental	8 (0.7)	2 (0.9)
	Gingival	3 (0.2)	–
	GOC	1 (0.08)	–
Odontogenic tumours	Total	342 (100)	136 (100)
	Odontoma	154 (45)	42 (30.8)
	Ameloblastoma	76 (22.2)	25 (18.3)
	Odontogenic myxoma	51 (14.9)	8 (5.8)
	AOT	23 (6.7)	2 (1.4)
	Odontogenic fibroma	20 (5.8)	2 (1.4)
	CEOT	3 (0.8)	2 (1.4)
	Cementoblastoma	2 (0.5)	–
	KCOT	–	53 (38.9)

n, number of biopsies; *N*, number of cases; %, frequency distribution; OKC, odontogenic keratocystic; GOC, glandular odontogenic cyst; AOT, adenomatoid odontogenic tumour; CCOT, calcifying cystic odontogenic tumour; CEOT, calcifying epithelial odontogenic tumour; KCOT, keratocystic odontogenic tumour.

Table 1 Prevalence of odontogenic tumours and odontogenic cysts in regard to gender in two different time periods

	No. biopsies			*Odontogenic tumors*			*Odontogenic cysts*		
Years	*Total*	♀	♂	*Total (%)*	♀ *(%)*	♂ *(%)*	*Total (%)*	♀ *(%)*	♂ *(%)*
1981–2004	12 729	7957	4772	342 (2.6)	212 (2.6)	130 (2.7)	1113 (8.7)	576 (7.2)	537 (11.2)
2005–2008	2706	1734	972	136 (5)	83 (4.7)	53 (5.4)	209 (7.7)	96 (5.5)	113 (11.6)
Total	15 435	9691	5744	478 (3.1)	295 (3)	183 (3.1)	1322 (8.5)	672 (6.9)	650 (11.3)

Females, (♀); males, (♂); prevalence, (%).

the 2nd place and now the most frequent OT is the KCOT. These data agree with very recent reports on Chinese and Libyan population (Luo and Li, 2008; El-Gehani *et al*, 2009). In these particular populations, the most frequent OT is KCOT (Luo and Li, 2008; El-Gehani *et al*, 2009). These data suggest that the amount of OKC could be enough to modify the prevalence of the OT and therefore the KCOT will occupy a preponderant place in the prevalence of OT.

Our data suggest that the redefinition and the reclassification of KOCT modified the prevalence and frequency distribution of OT. KCOT has a lightly male predominance, it is most frequent in the third decade of life and the mandible is the site of occurrence (González-Alva *et al*, 2008). This epidemiological profile of KOCT could modify the epidemiological profile of OT. We observed an increase in the prevalence of OT in males in the period 2005–2008. If this increase is related to KCOT should be established. A research protocol designed ex-profeso will be necessary to clarify this important issue.

In a Chinese series, the amount of OT without KOCT was 1054 while they increased to 1642 when KOCT was included (Jing *et al*, 2007). These data mean a 55.7% increase (Jing *et al*, 2007). We obtained an increase of 92% when KOCT was included as OT. In our opinion, this finding should be taken cautiously because it is result of a reclassification and not associated with a real increment in the cases of OT. The report of an increase of 92% in a period of time of 4 years in a very specific and uncommon neoplasias could be misunderstood by health carriers, specifically the managers of the preventive programmes of oral public health and to influence strategies of prevention. The oral pathologist should be awakened to this situation to clarify any misunderstanding in this issue.

Author contributions

Dr Luis A Gaitán-Cepeda was the coordinator of the research teams, Drs Daniel Quezada-Rivera and Fernando Tenorio-Rocha reviewed the cases and Dr Elba and Leyva-Huerta did the statistical analysis.

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