

Odontogenic tumors: a 14-year retrospective study in Santa Catarina, Brazil

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Abstract: Odontogenic tumors (OTs) are lesions that develop exclusively on maxillary bones, and form a heterogeneous group. They vary from hamartomatous lesions to benign and malign tumors. Although they are rarely observed in dentistry clinics, it is extremely important for the dentist to be aware of them. The aim of this study was to investigate the incidence of odontogenic tumors diagnosed in the population of Florianópolis, Santa Catarina, Brazil. Cases of odontogenic tumors were selected from the anatomopathological diagnostic services at Federal University of Santa Catarina from 1998 to 2011. Clinical data on these cases were collected from biopsy reports and patient files. Seventy-eight cases of odontogenic tumors were surveyed. Of these diagnoses, 51% were keratocystic odontogenic tumors (KCOTs); the remaining cases were mainly ameloblastomas and odontomas. The most frequently observed lesion in this retrospective study was KCOT (more than half of cases). Thus, this study shows that modifying the classification of the OTs altered the frequency of the lesions, possibly making KCOT the most common lesion observed in diagnostic services worldwide.

Descriptors: Odontogenic Tumors; Pathology, Oral; Epidemiology.

Introduction

Odontogenic tumors (OTs) develop exclusively on gnathic bones through the proliferation of odontogenic tissues (epithelial, mesenchymal or both). They form a heterogeneous group of hamartomatous lesions, benign and malign tumors. According to their origins, they are divided into epithelial, ectomesenchymal, and mesenchymal. Among the most frequent OTs are keratocystic odontogenic tumors (KCOTs), ameloblastomas, and odontomas.¹

Over time, OTs have passed through several modifications in their classification. In 1869, Broca used the term “odontoma” for any lesion arising from tissues that were part of the dental formation.² Later, several authors began to use and modify this terminology, and it was only in 1971 that the World Health Organization (WHO) published the first guide for the classification of OTs.³ In the most recent classification, “WHO Classification of Tumors - Head and Neck Tumours” in 2005,¹ there were several modifications. One was the inclusion of the keratocystic odontogenic tumor in the category of benign tumors; it was no longer categorized as an odontogenic cyst.² Information was also added

concerning lesions, including their definition, epidemiology, etiology, clinical and imaging features, and genetics.⁴

OTs are rare lesions in dentistry clinics, and their incidence varies based on geographic location. In Brazil, studies report that the incidence of these lesions varies between 2.4%,⁵ and 6.8%⁶ of all diagnosed oral lesions. Findings from other parts of Latin America are similar, such as those by Ledesma-Montes *et al.*,⁷ whose multicentric study (realized in Mexico and Guatemala) observed a 2.16% incidence of OTs, and in another study performed in Chile in which the authors found an incidence of 1.29% for OTs⁸ among all the oral lesions diagnosed in their services. Studies from other parts of the world, such as the one conducted by Jones and Franklin⁹ in the United Kingdom, found a lower frequency of 0.8% for all OTs. However, reports from Nigeria showed a greater prevalence of OTs, such as frequencies of 9.6% and 19% observed in studies by Ladeinde *et al.*¹⁰ and Odukoya,¹¹ respectively.

The aim of the present study was to investigate the incidence and main characteristics of OT cases that were diagnosed at *the Universidade Federal de Santa Catarina* in two diagnostic services, which are references to the Santa Catarina State.

Methodology

Cases for the study were selected from histopathological reports of two diagnostic services of the *Universidade Federal de Santa Catarina - UFSC*, the Oral Pathology Laboratory and the Pathological Anatomy Service, from 2006 to 2011 and from 1998 to 2010, respectively. Slides stained with hematoxylin and eosin (H&E) were selected from the histopathological diagnosis reports for evaluation under light microscopy (Olympus Corporation, Tokyo, Japan) and classified according to WHO criteria.¹ Clinical data were collected from biopsy reports stored by both services, as well as from the hospital records. Patient information included gender, age, ethnic group and, in KCOT cases, association with nevoid basal cell carcinoma syndrome (NBCCS), also known as Gorlin syndrome. Lesion-related data were the location, clinical and radiographic aspects of the lesions. Data were collected and, af-

Table 1 - Distribution of the cases diagnosed.

Diagnoses	n	%
Keratocystic odontogenic tumor	40	51
Ameloblastoma	18	23
Odontoma	13	17
Odontogenic myxoma	2	3
Calcifying cystic odontogenic tumor	2	3
Cementoblastoma	1	1
Central odontogenic fibroma	1	1
Calcifying epithelial odontogenic tumor	1	1
Total	78	100

Table 2 - Cases diagnosed distributed by gender.

Diagnoses	Male		Female	
	n	%	n	%
Keratocystic odontogenic tumor	15	62	10	38
Ameloblastoma	9	50	9	50
Odontoma	6	46	7	54
Odontogenic myxoma	1	50	1	50
Calcifying cystic odontogenic tumor	-	-	2	100
Cementoblastoma	-	-	1	100
Central odontogenic fibroma	1	100	-	-
Calcifying epithelial odontogenic tumor	-	-	1	100
Total	32	51	31	49

ter being filed in an initial format, they were later filed in a Microsoft Excel® (Microsoft Corporation, Redmond, USA) spreadsheet. Descriptive statistical analysis was performed with all collected data using Microsoft Excel® software. This study was approved by the Ethics Committee for Research with Human Beings at UFSC under number 1055/10.

Results

In our study, OTs corresponded to 3% of all oral lesions diagnosed by both services. In Table 1, we can observe the general prevalence of the lesions in these cases. These cases occurred more frequently in male patients (Table 2) and Caucasian (89%). The lesions were most prevalent in the age group between 21 and 30 years (Table 3). The preferential location was the posterior region of the mandible

Table 3 - Distribution according to the patients' age group.

Diagnoses	Age group	Mean age	Age Group							
			00-10	11-20	21-30	31-40	41-50	51-60	61-70	71-80
Keratocystic odontogenic tumor	05-71	35.5	1	5	7	3	3	3	2	1
Ameloblastoma	15-64	27.25	-	4	10	2	1	-	1	-
Odontoma	06-75	25	4	2	3	-	1	1	-	1
Odontogenic myxoma	08-32	20	1	-	-	1	-	-	-	-
Calcifying cystic odontogenic tumor	12-70	41	-	1	-	-	-	-	1	-
Cementoblastoma	37	37	-	-	-	1	-	-	-	-
Central odontogenic fibroma	18	18	-	1	-	-	-	-	-	-
Calcifying epithelial odontogenic tumor	31	31	-	-	-	1	-	-	-	-
Total	05-75	30.36	6	13	20	8	5	4	4	2

Table 4 - Distribution of the cases according to the predominant location of the lesion.

Diagnoses	Posterior mandible	Anterior mandible	Posterior maxilla	Anterior maxilla	Other	n
Keratocystic odontogenic tumor	22	8	5	2	3	40
Ameloblastoma	15	1	-	-	2	18
Odontoma	4	3	2	3	-	12
Odontogenic myxoma	2	-	-	1	-	3
Calcifying cystic odontogenic tumor	-	-	-	2	-	2
Cementoblastoma	1	-	-	-	-	1
Central odontogenic fibroma	1	-	-	-	-	1
Calcifying epithelial odontogenic tumor	1	-	-	-	-	1
Total	46	12	7	8	5	78

(Table 4), and the lesions presented as radiolucent images (74%), mixed radiolucent-radiopaque (10%) or radiopaque (4%). Among radiolucent lesions, the largest number of lesions was KCOT (64%), followed by ameloblastoma (34%) and myxoma (2%). Among mixed lesions, 57% were odontomas, followed by a calcifying cystic odontogenic tumor (CCOT, 28%) and a calcifying epithelial odontogenic tumor (CEOT, 15%). Among the radiopaque lesions two cases were odontomas (67%), and one case was cementoblastoma (33%). Nine cases (12%) had no information concerning radiographic characteristics: seven were odontoma (78%), one case was myxoma (11%), and one case was central odontogenic fibroma (COF, 11%).

KCOT was the most prevalent lesion in our study, with 51% of the cases diagnosed in both ser-

vices, and it occurred more frequently in male patients with a mean age of 35 years, varying between 5 and 75 years. These KCOT cases occurred in all areas of the maxillary bones, although the most frequent were the posterior mandible. Of all KCOT cases diagnosed in both services, 40% were associated with NBCCS. When we evaluated the KCOT cases associated with NBCCS, and the isolated KCOT cases, we verified that the mean age of the cases associated with the syndrome was 28 years, whereas the mean age for patients without the syndrome was 31 years. The ethnic group, and gender distributions were very similar between the two groups (KCOT cases associated with NBCCS, and isolated KCOT cases). We also checked the number of recurrence, and found seven cases of recurrence, of which two were associated with NBCCS. Occurrence of mul-

tiple lesions, all associated with the syndrome, was observed in four patients.

The second-most frequent lesion, ameloblastoma, did not show prevalence based on gender and the mean age of the patients was 27 years, varying between 15 and 64 years. All cases of ameloblastoma occurred in the mandible, mainly in the posterior area. Ameloblastoma occurred in two different clinical presentations, solid (61%) and unicystic (39%). All cases were histologically classified, being divided into follicular (33%), plexiform (22%), association between plexiform and follicular (96%), unicystic with mural proliferation (22%), unicystic with luminal proliferation (11%), and unicystic with intraluminal proliferation (6%).

The third-most prevalent lesion, odontoma, showed a slight preponderance for the female gender, and most of the cases occurred in young patients up to 10 years of age. The preferential location for this lesion was the posterior mandibular area.

Discussion

OTs are lesions rarely observed in dentistry clinics,^{5-7,9,12,13} and their frequency was 3% in our study, with a mean age of 30 years, varying between 5 and 75 years, and occurring more often in male patients. Their main location was in the posterior mandibular area. These data agree with findings from other studies performed in different countries.^{5,6,13-15} Our study demonstrated that most of the lesions occurred in Caucasian patients, which can be easily explained, as Caucasians correspond to 85.7% of the entire population in the Santa Catarina State according to data from the *Instituto Brasileiro de Geografia e Estatística* - IBGE.¹⁶

KCOT was the most frequent lesion observed in our study, comprising 51% of all lesions. Recently, KCOT has been reclassified by the WHO as a cystic lesion becoming a benign tumor.¹⁷ With this reclassification KCOT became the most frequent odontogenic tumor; however, it is still difficult to find a series of studies including this lesion as an OT.¹ The frequency of KCOTs in our study was 51% of all diagnosed cases, which was higher than the 28% to 36% frequency found in other studies.^{13,18-20} We verified that the mean age of KCOT cases was 35

years, and that the posterior mandible was the most affected site. Data found in our study corroborate the data found in other studies,^{13,18} and we found a greater prevalence for the male gender, in agreement with the results reported by Jing *et al.*¹⁹ and González-Alva *et al.*²¹ The association of KCOT with NBCCS was seen in 40% of the cases, which is very different from what was reported by González-Alva *et al.*,²¹ who only found an association of 6%. The same study reported data on the characteristics of the cases associated with NBCCS, reporting a greater prevalence in female patients (63.6%), which was different from the observations of the present study, wherein we found a higher prevalence in male patients (57% of cases associated with NBCCS). In our study, the age of NBCCS patients was between 5, and 56 years, with a mean of 28 years, which differed from the findings presented by González-Alva *et al.*,²¹ who presented cases whose age varied from 8 to 43 years with a mean of 19.5 years. Those authors also reported recurrence in only three cases, whereas it was observed in seven cases in our study.

Ameloblastoma is considered one of the most common lesions among OTs, and its incidence varies from 18%²² to 45%.²³ In our study, the incidence of ameloblastoma was 23% in all diagnosed cases, and these data are similar to findings from other studies.^{7,14,18,20} No gender-related preponderance was noted in our study, which was different from other studies in which the incidence in the female gender was higher.^{5,9,13,23,24} The most frequent clinical presentation was of the solid ameloblastoma (61%) and, histologically, of the follicular variant (33%). This is very similar to what was observed in the studies conducted by Ledesma-Montes *et al.*,⁷ Osterne *et al.*,¹³ Saghravanian *et al.*¹⁵ and Santos *et al.*⁵ However, Fregnani *et al.*²⁴ observed that the most frequent histological variant was the plexiform, which was observed in 53% of the cases. In our study, the posterior mandibular area was the preferred location, which was consistent with other studies.^{5,7,13,14,19,22-24}

Odontoma, also considered a frequent OT, was the third-most common lesion in our study. In some studies, it is shown to be the most frequent lesion,^{5,6,12,22} although our study is more consistent with those conducted by Avelar *et al.*,¹⁸ Fernandes

et al.,²³ Jing *et al.*,¹⁹ Jones and Franklin,⁹ Olgac *et al.*,¹⁴ Osterne *et al.*,¹³ and Saghravarian *et al.*¹⁵ in presenting an incidence of approximately 17%. Our study is in agreement with the data found in the literature that show a higher frequency in female patients.^{5,13,15,22} In the present study, the mean age was 23 years, a result that is also similar to those found in the literature.^{5,15,19,23} The preferential location for these lesions was the posterior mandibular area, which corroborated the findings of Jing *et al.*¹⁹ and Saghravarian *et al.*¹⁵

Other lesions had a lower incidence, such as myxoma (3%), CCOT (3%), cementoblastoma (2%), COF (1%), and CEOT (1%). This lower incidence can also be observed in other studies, such as those conducted by Avelar *et al.*,¹⁸ Jing *et al.*,¹⁹ and Santos *et al.*⁵ However, myxoma had a slightly higher incidence in some studies, such as in that conducted by Olgac *et al.*,¹⁴ who observed an incidence of 16%, being the third-most frequent lesion.

Very few studies have investigated OTs and, thus, a comparison between our results, and those published in the literature remains very difficult. Furthermore, the large ethnic diversity found worldwide, and even inside a single country with continental proportions, such as Brazil, creates substantial differences in the results. With the 2005 WHO

publication,¹ it has become important to conduct new studies using this new classification of OTs so that a new panorama can be defined for the incidence of this group of lesions. In addition, multicentric studies that can establish the worldwide characteristics of OTs are also important.

Conclusion

In our study, the most frequent lesion was KCOT, comprising almost half of the diagnosed cases. This shows that with the modification of the classification of OTs, KCOT is possibly the most common lesion observed in diagnostic services worldwide. However, there is a lack of studies demonstrating these frequencies, including data even showing KCOT as an OT.

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