

Odontogenic tumors: analysis of 188 cases from Saudi Arabia

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BACKGROUND AND OBJECTIVES: Odontogenic tumors (OTs) represent an uncommon group of lesions that arise from the tooth-forming apparatus. They pose a significant diagnostic and management challenge. There is a lack of data among the Saudi population. The aim of the study was to establish the relative frequency of the various histological types of OTs.

DESIGN AND SETTINGS: A retrospective study of 188 cases of OTs using the histopathology archives of the College of Dentistry, King Saud University.

METHODS: The histopathology archives of the College of Dentistry, King Saud University were reviewed from January 1984 to December 2010 for OTs. The age and gender of the patients, tumor site, and histopathologic typing were analyzed.

RESULTS: A total of 188 (4.3%) patients met the criteria for being classified as an OT. Odontogenic keratocystic tumor (36.7%) was the most commonly diagnosed, followed by ameloblastoma (25.0%), odontoma (14.9%), and odontogenic myxoma (6.4%). Two cases of malignant OTs (1.1%) are found. The male-to-female ratio was 1.4:1. The most frequently affected area was the posterior mandible (48.9%), followed by the anterior maxilla (22.9%).

CONCLUSION: This is a relatively large series of OTs revealing aspects of similarities and differences with those of previous studies of populations in Africa, Asia, and the Americas. The findings of the present study may be useful as a guide for clinicians who need to make clinical judgments prior to biopsy about the most probable diagnosis.

Odontogenic tumors (OTs) represent a rare group of lesions that arise from the tooth-forming apparatus. They constitute a group of heterogeneous diseases that range from hamartomatous tissue proliferations and benign neoplasms to malignant tumors with metastatic potential.¹ While they are uncommon, they can pose a significant diagnostic and management challenge. OTs are generally classified according to their presumed tissue of origin (e.g., epithelial, mesenchymal, or mixed lesion).² There have been several attempts to reclassify OTs according to their diverse histopathological features. The latest revised histopathological classification by the World Health Organization (WHO) was published in 2005 by the International Agency for Research on Cancer,³ and it included the reclassification of odontogenic keratocysts and calcifying odontogenic cysts as tumors.

Published cases of OTs have been found to vary depending on the affected population.^{4,5} In Saudi Arabia, a few case reports are available.^{6,7} The aim of the present study was to establish the relative frequency of the various histological types of OTs over a period of 27 years using the histopathology archives of the College of Dentistry, King Saud University. The latest WHO classification criteria were applied and the results were compared with previously reported cases of OTs from other populations.

METHODS

This study was conducted in compliance with the "Ethical Principles for Medical Research Involving Human Subjects" statement of the Helsinki Declaration and was approved by the Committee of Ethics in Research of the College of Dentistry Research Center

(IR: 0123).

Histopathology archives from January 1984 to December 2010 that are maintained at the Histopathology Laboratory of the College of Dentistry, King Saud University, Riyadh, were reviewed. These lesions included OTs, as well as keratocysts, including odontogenic keratocysts, keratocystic OTs, and calcifying odontogenic cysts. Sections stained with hematoxylin and eosin were re-examined to confirm the diagnosis in accordance with the revised histopathological classification of OTs by the WHO.³ The age and gender of the patients associated with each lesion, including the tumor site and histopathologic typing, were analyzed using SPSS, version 17.0 (SPSS Inc., Chicago, IL, USA). The Chi-square test was applied to analyze the statistical significance of the data when applicable. A *P* value less than .05 was considered statistically significant.

RESULTS

Of the 4408 lesions of the oral cavity and jaws that were reviewed, 188 (4.3%) met the criteria for being classified as an OT. Odontogenic keratocystic tumor (KCOT, 36.7%) was the most commonly diagnosed, followed by ameloblastoma (25.0%), odontoma (14.9%), and odontogenic myxoma (6.4%). Of the 47 cases of ameloblastoma, 8 (17.0%) were unicystic. In addition, 2 cases of malignant OTs were reported (1.1%) (Table 1).

For the tissues examined, the age of the corresponding patients at diagnosis ranged from 7 years to 82 years (mean, 29 years). The incidence of the OTs peaked between the second and third decades of life (Table 2). The male-to-female ratio was 1.4:1. A slight overall male predominance (59%) was observed, and the difference remained when the lesions were examined individually such as in ameloblastoma (*P*<.01) and KCOT (*P*<.00), except for the adenomatoid OT. For the latter, females were more often affected (87.5%). The mandible and maxilla were involved in 66.7% and 29.6% of the OT cases, respectively, and the most frequently affected area was the posterior mandible (48.9%), followed by the anterior maxilla (22.9%) (Table 3).

DISCUSSION

For large series studies of OTs that have been conducted for different populations, the age and gender of the affected patients, as well as the site of the OT lesions have been reported, and regional differences have been observed.^{8,9} However, the relative frequency of OTs in various populations is difficult to compare due to differences in the classification methods used and the exclusion of some recently recognized entities. Another

significant factor is the reclassification of odontogenic keratocysts as tumors.¹⁰ Only a limited number of studies of the population of Saudi Arabia are available. Therefore, to our knowledge, the present study represents the first large series for this country.^{6,7}

OTs are rather uncommon and represent a relatively small percentage of all biopsy specimens submitted to oral and maxillofacial histopathology laboratories worldwide. Correspondingly, in the current study, OTs represented 4.3% of all of the cases archived between January 1984 and December 2010. In other studies where odontogenic keratocysts were classified as tumors, the reported percentage of OT cases ranged from 3.9% to 19%.^{11,12,4} Interestingly, however, when OKCT and COC cases were excluded, OTs only constituted 1.8% of all cases, which is comparable to the incidence rates reported for OTs in North America (1.2%),⁸ Pakistan (1.7%),¹³ and Iran (1.9%).¹⁴ A significantly higher percentage (5.1%) was reported from the western province of Saudi Arabia.⁷ This might be attributed to the diverse ethnic background characterizing this part of the country. However, such a percentage should be interpreted with caution in the absence of well-defined inclusion/exclusion criteria.

The most common tumor identified was KCOT (36.7%), followed by ameloblastoma (25.0%), odontoma (14.9%), and odontogenic myxoma (6.4%).

Table 1. Relative frequency the odontogenic tumors reviewed (n=188).

Tumor	No. of cases	Percentage
Keratocystic odontogenic tumor	69	36.7
Ameloblastoma	47	25.0
Odontoma	28	14.9
Odontogenic myxoma	12	6.4
Calcifying cystic odontogenic tumor	11	5.8
Adenomatoid odontogenic tumor	8	4.3
Cementoblastoma	4	2.1
Ameloblastic fibro odontoma	3	1.6
Calcifying epithelial odontogenic tumor	2	1.1
Odontogenic fibroma	1	0.5
Ameloblastic fibroma	1	0.5
Ameloblastic carcinoma	1	0.53
Clear cell odontogenic carcinoma	1	0.5
Total	188	100.0

OT (Odontogenic tumor), KCOT (Keratocystic odontogenic tumor), CCOT (calcifying cystic odontogenic tumor), AOT (adenomatoid odontogenic tumor), CEOT (calcifying epithelial odontogenic tumor).

Table 2. Age distribution of the patients with odontogenic tumors (y).^a

Tumor	Age group (y)							Total (n)
	1-10	11-20	21-30 ^a	31-40	41-50	51-60	>60	
Keratocystic odontogenic tumor	2	15	24	10	9	2	2	64
Ameloblastoma	0	10	20	11	4	1	0	46
Odontoma	5	12	4	5	0	0	0	26
Odontogenic myxoma	0	4	3	3	0	1	1	12
Calcifying cystic odontogenic tumor	0	5	2	3	0	0	1	11
Adenomatoid odontogenic tumor	0	3	5	0	0	0	0	8
Cementoblastoma	0	3	1	0	0	0	0	4
Ameloblastic fibro odontoma	3	0	0	0	0	0	0	3
Calcifying epithelial odontogenic tumor	0	0	2	0	0	0	0	2
Odontogenic fibroma	0	1	0	0	0	0	0	1
Ameloblastic fibroma	0	0	0	1	0	0	0	1
Ameloblastic carcinoma	0	0	0	1	0	0	0	1
Clear cell odontogenic carcinoma	0	0	1	0	0	0	0	1
Total	10	53	62	34	13	4	4	180

^aPatient age was not reported for 8 cases. OT (Odontogenic tumor), KCOT (Keratocystic odontogenic tumor), CCOT (calcifying cystic odontogenic tumor), AOT (adenomatoid odontogenic tumor), CEOT (calcifying epithelial odontogenic tumor).

Table 3. Site distribution of the odontogenic tumors reviewed.^a

Tumor	Site				Total (n)
	Mandible		Maxilla		
	n	%	n	%	
Keratocystic odontogenic tumor	49	77.8	14	22.2	63
Ameloblastoma	38	86.4	6	13.6	44
Odontoma	11	42.3	15	57.7	26
Odontogenic myxoma	9	75	3	25	12
Calcifying cystic odontogenic tumor	5	45.5	6	54.5	11
Adenomatoid odontogenic tumor	5	62.5	3	37.5	8
Cementoblastoma	3	75	1	25	4
Ameloblastic fibro odontoma	1	50	1	50	2
Calcifying epithelial odontogenic tumor	0	0	2	100	2
Odontogenic fibroma	0	0	1	100	1
Ameloblastic fibroma	1	100	0	0	1
Ameloblastic carcinoma	1	100	0	0	1
Clear cell odontogenic carcinoma	1	100	0	0	1
Total	176				

^aTumor site was not reported for 12 cases.

Keratocystic OTs were not included in reports published prior to the latest update of WHO classification criteria for OTs; and this is an important consideration when comparing different studies. In the current study, the most frequent OT identified was ameloblastoma (43.5%) when KCOT and COC were excluded. Malignant OTs are extremely rare, with the reported incidence ranging from 9.7% to 0%.^{12,15} In the present study, only 2 cases of malignant tumors were diagnosed, which constituted 1.1% of all of the OT cases reviewed. However, the perception that malignant OTs are extremely rare, may be misleading. Therefore, malignancy or malignant transformation of a benign tumor should be suspected when a lesion exhibits atypical clinical and/or histopathological features.¹⁶

Many studies from Asia, Africa, and South America have identified ameloblastoma as the most common OT diagnosed.^{7,11,17,18} However, in some studies that were conducted according to the 2005 WHO classification criteria, KCOT was identified as the most common OT.^{4,5,19,20} In North America and Europe, odontoma is the most common OT.^{8,21} It is well-documented that geographic variations exist in the incidence of ameloblastoma and odontoma, although the relatively lower incidence of odontoma in developing countries may be attributed to the absence of strict adherence to a biopsy submission policy compared with developed countries. Any pathological tissue should be submitted for histopathological examination; however, many cases may be diagnosed clinically without submission of tissues for histopathological evaluation.

For the tumors retrospectively reviewed, the age of

the corresponding patients at diagnosis ranged from 7 to 82 years, and the incidence of OT peaked between the second and third decades of life. These results are consistent with those of previous reports.^{7,13,22} In addition, male dominance for OTs was found to be statistically significant, and this is consistent with some reports of other populations.^{13,20} Finally, the posterior mandible was identified as the most frequently affected anatomical site, consistent with the results of other studies.^{7,17,23,24}

Malignant OTs are typically rare. In the current study, these cases represented 1.1% of all of the cases examined. This incidence is comparable to that reported in the United States (1.5%)⁸ and Turkey (1.1%),²³ although a higher frequency has been reported in Egypt (3.7%),²⁵ Nigeria (5%),¹² and China (5.1%).¹¹

In conclusion, this analysis of a relatively large series of OTs revealed some similarities and some differences between our findings and those of previous studies of populations in Africa, Asia, and the Americas. The findings of the present study may be useful as a guide for clinicians who need to make clinical judgments prior to biopsy about the most probable diagnosis, and need to anticipate the risks associated with certain types of lesions. Further studies are also needed to characterize the incidence of OTs in different regions of the Saudi Arabia.

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