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Clinicopathologic Study of Odontogenic Tumors; 118 Cases

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Abstract: The aim of this study was to determine the relative frequency and clinicopathologic presentations of odontogenic tumors in an Iranian population. We reviewed the archives of 110000 files from 1997-2007 in the Department of Pathology of Shiraz Dental School and two main head and neck referral hospitals in Shiraz by using the criteria for histological classification of odontogenic tumors published by WHO in 2005. Information about clinical featuers (age, sex and location etc.) were obtained from the patients' chart and analyzed by χ^2-test. We found 118 cases of odontogenic tumors which were all benign. Keratocystic odontogenic tumor was the most frequent tumor (42.3%) followed in descending order by ameloblastoma (30.5%), odontoma (9.3%), calcifying epithelial odontogenic cyst (6.7%), myxoma and adenomatoid odontogenic tumor (2.5%). Odontogenic tumors are uncommon lesions in the Shirazian population. The overall male to female ratio was 0.9:1 and the mandible was obviously more affected than the maxilla (3.04:1).

Key words: Odontogenic tumors, keratocystic odontogenic tumor, ameloblastoma, patients, Iran

INTRODUCTION

Odontogenic Tumors (OTS) consist of a complex group of lesions of diverse histopathologic types and clinical behaviors, arising from tooth-producing apparatus (epithelial and/or ectomesenchymal tissues) (Neville *et al*., 2009). In humans, tumors of the odontogenic tissues are comparatively rare, comprising of about 1% of all jaw tumors and several histologic classification schemes have been devised for OTS. The most common classification is based on the origin of tumors (Regezi *et al*., 2008). In 1971, the World Health Organization published the first edition of the histological typing of odontogenic tumors and in 1992 a revised second edition appeared (Ochsenius *et al*., 2002).

Several reports on odontogenic tumors were published from various parts of the world based on these two classifications. However, due to many controversial issues about subtypings, terminology and diagnosis, the WHO published the latest update edition of histological typing of OTS with some significant changes. The tumors are reclassified based on some information such as etiology, epidemiological information, clinical and radiographic features, tumor macroscopy, tumor genetics and prognosis of the lesions besides the histologic feature.

Knowledge of relative frequency and typical basic features such as age, location and gender of different OTS can be useful in developing a clinically differential diagnosis.

Available literature about the relative frequency of OTS is mostly obtained from American and African populations (Sriram and Shetty, 2008). Very few studies are reported among Asians especially from the Iranian subcontinent. The aim of this study was to determine the relative frequency and clinicopathologic presentation of OTS using the database available in the Department and the Department of Pathology of two main hospitals in Shiraz which received most of the head and neck lesions from Fars province over a period of 10 years (1997-2007) and to compare these data with previous reports.

MATERIALS AND METHODS

The subject of this study was all cases of odontogenic tumors that were biopsied and had undergone pathologic examination at the department of pathology of Shiraz Dental School, Khalili and Namazi hospitals between March 1997 and March 2007. Microscopic slides of all samples were reviewed by a second oral pathologist to confirm the histologic diagnosis. The information gathered from the clinical charts included the patients' age and sex, site of the

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lesion and its clinical history. A total number of 118 lesions were reclassified as intraosseous odontogenic tumors according to the criteria of WHO.

The original and recurrent tumors were considered as an individual case. The classification of site distribution was performed according to the radiographic extension as used by Sriram and Shetty (2008) with the addition of an excess category named class 5 to the mandibular lesions. Class 1 consisted of lesions in the anterior segment of the maxilla (from the distal aspect of 3⌋ to the distal aspect of ⌊3), Class 2 consisted of lesions in the posterior segment of the maxilla (from the mesial aspect of the first premolar to the end of the dental arch) Class 3 consisted of lesions that involved all segments of the maxilla. The lesions of the mandible were also divided into 5 categories. Class 1 consisted of lesions in the anterior segment of the mandible (from the distal aspect of 3⌋ to the distal aspect of ⌊3), Class 2 consisted of lesions in the posterior segment of the maxilla (from the mesial aspect of first premolar to the end of the dental arch).

Class 3 consisted of lesions that involved the ramus and angle of the mandible (from the distal aspect of second molar to the condyle). Class 4 consisted of lesions that involved all segments of the mandible. Class 5 consisted of lesions from the ramus of one side to another.

We used the statistical package for the social science, Version 11.5 (for windows) (SPSS, Chicago) and the Chi-square test for analyzing the results with the significance set at 95% ($\alpha = 0.05$).

RESULTS AND DISCUSSION

In this study, we found 118 cases of OTS which were all benign. Keratocystic odontogenic tumor (Kcot) was the most frequent tumor (42.3%) followed in descending order by Ameloblastoma (30.5%), Odontoma (9.3%), calcifying epithelial odontogenic cyst (6.7%), Myxoma and Adenomatoid odontogenic tumor (2.5%) (Table 1).

Table 2 shows the incidence of each tumor by age and sex. Of 118 OTS, the gender distribution was 60 females and 58 males with an overall male to female ratio of 0.9:1. There was no significant difference among men and women when comparing kcot and ameloblastoma ($\chi^2 = 0.0191$ p = 0.662).

The lesions occurred in patients from 7-70 years of age with a mean age of 27.2 years; 76.2% of cases were between the ages of 10 and 39 with a peak incidence in the third decade (32.2%). Keratocystic odontogenic tumor showed a peak occurrence in the second and third decades (56%) and the peak occurrence of ameloblastoma was seen in the forth decade (38.8%). Table 3 shows the location of each type of OTS in the maxilla and mandible. There were 91 cases in the mandible and 30 cases in the maxilla. The mandible was obviously more affected than the maxilla with a ratio of 3.04:1.

Table 1: Distribution of 118 odontogenic tumors according to sex

Lesion	No. of patients	Sex M	Sex F	Ratio
Ameloblastoma	36	17	19	0.8
Keratocystic odontogenic tumor	50	26	24	1.0
Odontoma	11	8	3	2.6
Ameloblastic fibroma	2	2	-	-
Ameloblastic fibro-odontoma	1	-	1	-
Myxoma	3	-	3	-
Adenomatoid odontogenic tumor	3	-	3	-
Calcifying odontogenic tumor	2	-	2	-
Cementoblastoma	2	1	1	-
Calcifying epithelial odontogenic tumor	8	4	4	-
Total	118	58	60	-

Ameloblastoma and Kcot had a strong predilection for the mandible; 94% of cases of ameloblastoma and 76% of cases of Kcot were found in the mandible. Calcifying odontogenic tumors and calcifying odontogenic cysts had a predilection for the maxilla (100 and 62%, respectively). In general, the most common location of OTS was the posterior part of the jaws, except for adenomatoid odontogenic tumor which was more common in the anterior part of both jaws.

In 1971, WHO published the first edition of the histological typing of odontogenic tumors and revised it in 1992 as the second edition which was used widely in recent studies. There were still many controversial issues over classification and terminology of OTS so, in 2005 the WHO published the latest edition for the definition and classification of these tumors. Some of significant differences in the new classification are mentioned below:

- The term keratocystic odontogenic tumor is now used instead of odentogenic keratocyst as a benign epithelium derived odontogenic tumor (Reichart *et al*., 2006)
- Adenomatoid odontogenic tumor which was formerly in the mixed odontogenic tumor group has now been classified as a benign tumor with odontogenic epithelium and mature stroma without odontogenic ectomesenchyme (Reichart *et al*., 2006)
- The new term clear cell odontogenic carcimoma has now been used for clear cell odontogenic tumor and it has been reclassified as a malignant odontogenic carcimoma (Reichart *et al*., 2006)
- There are also a few changes in the group of mesenchymal odontogenic tumors with or without odontogenic epithelium

Table 2: Age distribution of different odontogenic tumors

Lesion	Unknown	0-9	10-19	20-29	30-39	40-49	50-59	60-69	+70
Ameloblastoma	1	-	5	10	14	3	1	1	1
Keratocystic odontogenic tumor	-	3	14	13	8	4	4	4	-
Odontoma	1	1	3	5	1	-	-	-	-
Ameloblastic fibroma	-	-	-	2	-	-	-	-	-
Ameloblastic fibro-odontoma	1	-	-	-	-	-	-	-	-
Myxoma	-	-	-	3	-	-	-	-	-
Adenomatoid odontogenic tumor	-	-	3	-	-	-	-	-	-
Calcifying odontogenic tumor	-	-	-	-	-	-	2	-	-
Cementoblastoma	-	-	2	-	-	-	-	-	-
Calcifying epithelial odontogenic tumor	-	-	-	5	1	2	-	-	-

Table 3: Sites of different odontogenic tumors

	Mandible							Maxilla				
Lesion	Unknown	Total	Cl (1)	Cl (2)	Cl (3)	Cl (4)	Cl (5)	Unknown	Total	Cl (1)	Cl (2)	Cl (3)
Ameloblastoma	9	34	2	8	5	9	1	-	2	-	2	-
Keratocystic odontogenic tumor	13	38	4	10	10	1	-	5	15	3	7	-
Odontoma	2	6	-	4	-	-	-	3	5	1	1	-
Ameloblastic fibroma	-	2	-	-	1	1	-	-	-	-	-	-
Ameloblastic fibro-odontoma	-	1	-	1	-	-	-	-	-	-	-	-
Myxoma	1	2	-	-	1	-	-	-	1	-	1	-
Adenomatoid odontogenic tumor	1	3	2	-	-	-	-	-	-	-	-	
Calcifying odontogenic tumor	-	-	-	-	-	-	-	-	2	-	2	-
Cementoblastoma	-	2	-	2	-	-	-	-	-	-	-	-
Calcifying epithelial odontogenic tumor	-	3	1	1	-	1	-	4	5	-	1	-

Simple and WHO type of central odontogenic fibroma are now named as epithelial-poor and epithelial-rich odontogenic fibroma, respectively (Reichart *et al*., 2006). Since most of the previous studies on the frequency of various types of OTS such as (Odukoya, 1995; Lu *et al*., 1998; Ochsenius *et al*., 2002; Fernandes *et al*., 2005) are based on the 1992 WHO classification, we compared the results of this study with previous results according to the 1992 WHO edition. There are also few published studies about the frequency of OTS after the 1992 WHO classification. Therefore, further studies are necessary to obtain the real frequencies of OTS according to the new edition of the WHO classification.

In the present study, no malignant tumor was seen and this could be a referral bias, meaning that according to the aggressiveness of these lesions they were referred to more facilitated centers in the capital. Kcot was the most common tumor (42.3%) followed by ameloblastoma (30.5%) and odontoma (9.3%). The frequency of ameloblastma and odontoma in this study was similar to the data reported from Nigerian (Ladeinde *et al*., 2005) and Chinese people (Jing *et al*., 2007) whereas in most studies in Chili (Ochsenius *et al*., 2002) and Mexico (Mosqueda-Taylor *et al*., 1997), odontoma was the most common lesion (44.7 and 34.6%, respectively). These discrepancies probably result from geographic variations but it should be noted that the incidence of odontoma in some countries was likely underestimated because the clinical and radiographic features of this tumor are diagnostic and are seldom confused with any other lesion, so hospital management is not always undertaken.

The male-female ratio in this study was 0.9:1 which is in agreement with the data reported from Brazil (Fernandes *et al*., 2005), Chile, Mexico and China. However, what we found conflicts with the results of previous reports in the Nigerian population. Overall, these tumors generally affect men and women with similar predilection.

The mandible was affected more than the maxilla in this series of OTS (3.04:1). This finding confirms those reported in the Nigerian, Chinese and Brazilian series (3.1:2, 5.7:1, 2:1, respectively) which can be explained by the greater prevalence of ameloblastoma in these series.

However Mosqueda-Taylor *et al*. (1997) and Ochsenius *et al*. (2002) showed a slight predilection for the mandible. According to the location of the lesions they had a great tendency for the mandible although, the incidence of maxillary ameloblastomas varies considerably among different reports. In the present series, 6% of the ameloblastomas were found in maxilla. It was similar to the corresponding data from Asia, Africa and Brazil (2-8%).

In contrast, 16-22% of ameloblastomas in the American series were found in the maxilla suggesting another conflict due to geographic variations. The predilection of ameloblastoma for the posterior region of the mandible in this study is also consistent with previous reports. Malignant tumors were not found in this study. The frequency of malignant tumors in (Fernandes *et al*., 2005) series was 0.6% of all OTS. Other American series also showed very low incidence of these (<1.6%), contrasting to African and Chinese series which had a frequency of 5.2, 3.4 and 6.1%, respectively.

CONCLUSION

In the study, the features of OTS among the series are similar to those reported in China, Nigeria and Brazil rather than in Chili and Mexico. This may be explained by a true geographic difference between South America, Asia and Africa.

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