ORIGINAL RESEARCH

A 10-year retrospective study on odontogenic tumors in Iran

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Received : 23-11-11
Review completed : 02-03-12
Accepted : 04-07-12

ABSTRACT

Objective: The aim of this study was to review cases of odontogenic tumors diagnosed in two pathology centers in Tehran, Iran, during a 10-year period.
Study Design: Patients' records were seen at two teaching pathology Centre's of Shahid Beheshti University between the months of March 2000 to 2010 with histologic diagnosis of any type of odontogenic tumors. The records were analyzed for frequency, age, sex, site, as well as clinical, radiographic and histopathologic findings.
Results: Of 30706 biopsies, 4767 (15.5%) cases were diagnosed as oral and maxillofacial lesions. Among these, 720 cases were tumoral with 188 (26.1%) cases of odontogenic tumors. Tumors with odontogenic epithelium origin formed 70.2% of total numbers of odontogenic tumors. Mixed odontogenic tumors and tumors of odontogenic ectomesenchyme comprised 12.2% and 17.5% of the cases respectively. Ameloblastoma, with a frequency of 62.2% was the most common tumor in this review which was followed by odontoma and odontogenic myxoma.
Conclusion: Although there are few studies on odontogenic tumors in literature, the comparison of our results with existing data shows significant differences in the distribution of tumors and age of patients, which may be due to ethnic features and geographic distribution of patients. Future studies on other ethnic groups are essential for further clarification of the findings in this research.

Key words: Frequency, odontogenic tumors, Iran

Odontogenic tumors (OTs) comprise a complex group of lesions of diverse histopathologic types and clinical behavior. Some of these lesions are true neoplasms and may rarely exhibit malignant transformation and behavior. Others may represent tumor like malformations (hamartoma). These tumors, like normal odontogenesis, demonstrate varying inductive interactions between odontogenic epithelium and odontogenic ectomesenchyme. So these tumors derived from one of epithelial or ectomesenchymal odontogenic tissue or both.[1]

The first classification for these groups of lesions was published in 1971 by World Health Organization (WHO). In 1992 a revised edition of WHO classification was proposed by Philipsen and Reichart. In 2005, WHO published the latest edition of OTs classification with some changes, according to which odontogenic keratocyst was classified as a new entity of OTs. Several studies on OTs have been published from many parts of the world with different prevalence. Few studies have been reported among Asians especially about the frequency and characteristics of these tumors in the Middle East.

The aim of this study was to determine the epidemiologic characteristics of odontogenic tumors during a 10-year period in Iran according to categories of odontogenic tumors based on the 2005 WHO classification.[1] The data were then compared with previous reports in the literature.

MATERIALS AND METHODS

The data in this retrospective study included records of 2 referral pathology centers in Tehran: (a) Oral and Maxillofacial Pathology Department and (b) Taleghani Hospital of Shahid Beheshti University. Patients' records with biopsy-examined odontogenic tumors over a time span of 10 years from March 2000 to March 2010 were reviewed.

Patients' age, sex, tumor location and frequency, as well as clinical, radiographic and histopathologic features were the

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Access this article online

Quick Response Code:

Website: www.ijdr.in

DOI: 10.4103/0970-9290.116688

main analysis outcome measures. Data were analyzed using SPSS software (version 11.5).

RESULTS

Out of 30706 biopsies in 10 years, 4767 (15.5%) cases were oral and maxillofacial lesions with 720 tumoral diagnoses.

Of 720 tumoral cases in the oral and maxillofacial region, 188 (26.11%) cases were odontogenic tumors. Among odontogenic tumors, 181 (96.3%) cases were benign while only 7 (3.72%) cases were malignant. 132 (70.2%) cases were epithelial in origin, 33 (17.5%) cases were mixed tumors and 23 (12.2%) cases were odontogenic ectomesenchym tumors [Chart 1].

The most frequent tumor was ameloblastoma (62.2%), followed by odontoma (14.3%), Odontogenic myxoma (10.64%), ameloblastic carcinoma (3.72%) and adenomatoid odontogenic tumor (2.6%) [Table 1 and Chart 2].

Of the 188 OTs, in terms of gender, 104 cases were males and 84 cases were females. The male to female ratio was 1.2 in benign tumors and 2.5 in ameloblastic carcinoma [Table 2]. The peak age of incidence was the second and third decades of life [Table 3]. Regarding location [Table 4], 138 (73.4%) cases were in the mandible while 50 (26.6%) cases seen in the maxilla indicating an overall mandible-maxilla ratio of 2.7.

In this study, 117 cases of ameloblastoma were seen of which 98 cases were solid type and 19 cases were unicystic amelobelastoma. No peripheral type was seen. In solid type, the cases were more frequent in female and third decade of life. Almost all the lesions were located in the mandible (88.8%). Histologically, follicular pattern, with 28.57% occurrence, was the most common type.

In contrast, unicystic ameloblastoma was seen more in males with a contraction age relatively similar to solid ameloblastoma.

The present study identified 27 cases of odontoma which 8 (29.7%) cases belonged to females and 19 (70.3%) cases to males. The most frequently affected area was the anterior part of the maxilla (59.2%). There were 17 compound and 10 complex odontomas. The highest incidence was observed in 2nd decade (37%) of life. The most significant sign was expansion (22.2%) and interference with tooth eruption.

Table 1: Classification and distribution of odontogenic tumors

Diagnosis	Abbreviation	No.	%
Tumors of odontogenic epithelium			
Ameloblastoma	AME	117	62.2
Malignant ameloblastoma	MAME	-	-
Ameloblastic carcinoma	AC	7	3.74
Clear cell odontogenic carcinoma	CCOC	-	-
Adenomatoid odontogenic tumor	AOT	5	2.66
Calcifying epithelial odontogenic tumor	CEOT	3	1.60
Squamous odontogenic tumor	SOT	-	-
Total		132	
Mixed odontogenic tumors			
Ameloblastic fibroma	AF	2	1.06
Ameloblastic fibro-odontoma	AFO	3	1.60
Ameloblastic fibrosarcoma	AFS	-	-
Odonto-ameloblastoma	OA	1	0.53
Compound odontoma	OCp	17	9.06
Complex odontoma	OCx	10	5.31
Total		33	
Tumors of odontogenic ectomesenchyme			
Odontogenic fibbroma	OF	-	-
Granular cell odontogenic tumor	GCO	-	-
Odontogenic myxoma	OM	20	10.64
Cementoblastoma	CB	3	
Total		23	1.60

Table 2: Gender distribution of odontogenic tumors

Tumor	Male	Female	Male-female ratio
AME	61	56	1.09
CEOT	2	1	2
AOT	2	3	0.66
AF	1	1	1
AFO	2	1	1
OCp	12	5	2.4
OCx	7	3	2.3
OM	9	11	0.81
AC	5	2	2.5
CB	2	1	2
OA	1	0	1
Total	104	84	1.23

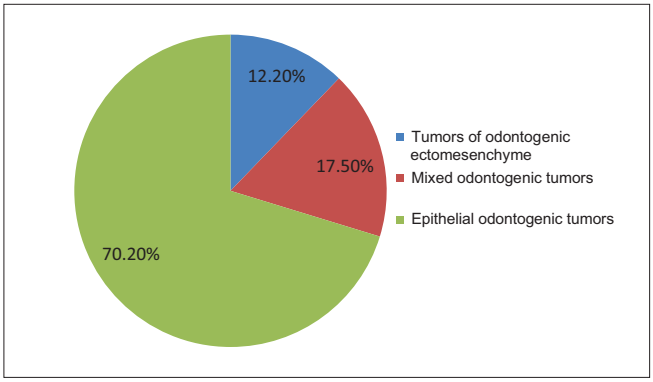

Chart 1: Frequency of odontogenic tumors according to origin

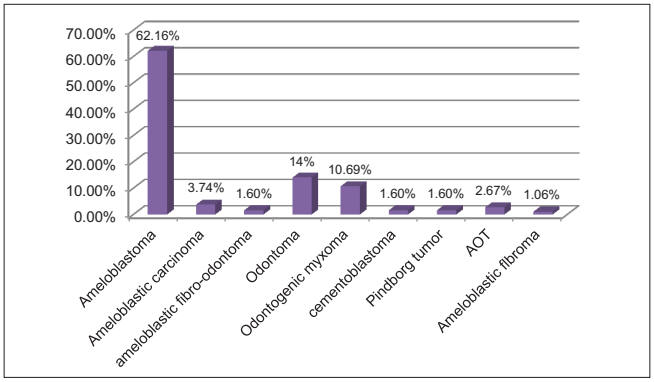

Chart 2: Frequency of odontogenic tumors

Table 3: Age distribution (years) of odontogenic tumors

Tumor	0-10	11-20	21-30	31-40	41-50	51-60	61-70	71-80	Mean±SD
AME	7	25	34	22	13	9	3	4	32.75±16.73
CEOT		1	1				1		33±0.27.87
AOT		5							15±1
AF		2							15±1.41
AFO	1	2							10.67±1.52
OCp	1	13	2	1					25.37±14.96
OCx	1	8	1						26.81±10.12
OM	2	8	4	3	2	1			24.45±12
AC			1	4		1	1		40.29±13.88
CB	1	1	1						16±8.78
OA			1						25
Total	13	65	45	30	15	11	5	4	

Table 4: Site distribution of odontogenic tumors

Tumor	Maxilla	Mandible	Mandible to maxilla ratio
AME	13	104	8
OM	11	9	0.8
AFO	1	2	2
CEOT	0	3	3
AOT	3	2	0.6
OA	0	1	1
AF	1	1	1
CB	1	2	2
AC	4	3	0.7
OCp	13	4	0.3
OCx	3	7	2.3
Total	50	138	

Odontogenic myxoma amounted to 20 (10.6%) cases. This tumor affected females more than males (i.e., 55% versus 45%). Patient's age ranged from 9 to 51 (mean 24.9%). There was a relatively significant predilection for maxilla (55%), with expansion as most frequent sign.

Ameloblastic carcinoma, diagnosed in these series, was limited to 7 cases. Because of the malignancy of this tumor it was separated from the other kinds of ameloblastoma. This tumor was more common in males (71.4%). The mean age of occurrence was 40.2 (10%). The tumor was seen in the maxilla (57.1%) rather than mandible (42.9%). The most frequent sign turned out to be expansion. Additionally, multilocular radiolucency was the most common radiographic feature.

Another tumor with limited occurrence, i.e., just 5 cases, was Adenomatoid odontogenic tumor (AOT). The mean age of the patients was 15. The tumor showed a predilection for females (60%) and the maxilla (60%). The most common type of AOT was follicular associated with impacted canine.

DISCUSSION

Odontogenic tumors are lesions derived from each of epithelial or ectomesenchymal tissues or both which serve as tooth forming apparatus. They comprise about 1% of all jaw tumors. Available literature on the relative frequency of OTs is mostly performed in America, Europe and Africa. Very few studies have so far been carried out in Asian countries.

This study presents the profile of odontogenic tumors in Iran according to category of OTs, based on the 2005 WHO classification. Such studies are still in high demand as the nomen culture has not been well established yet; the argument to support changes in nomen culture, particularly in relation to keratocystic odontogenic tumor and calcifying cystic odontogenic tumor, is not over yet.

In this research, out of a total of 4767 orofacial biopsies, 188 (3.94%) cases of OTs were observed which comprised 26.1% of tumoral lesions in the orofacial region.

This study confirmed previous studies reports that benign tumors (96.3%) were the most frequently seen OTs.[2,3] Malignant OTs were shown to be very rare. They were limited to 7 (3.7%) cases in our series.

Most of the available information about malignant OTs is case reports or small series of cases. This rarity was also reported by Daley *et al.*[4] The incidence was higher in Nigerian (5.1%) and Chinese (6.1%) population.[5-7] Male patients were affected more in recent study which was similar to W. Jing *et al.* study.[8] Also in line with Sriram's study, These tumors were more common in the maxilla (57.1%).[9]

In most of the previous studies, in terms of gender, OTs had a rather similar distribution for males and females. Nonetheless, there was a female preponderance in studies done by Regezi,[10] Wu and Chan[11] and Santos *et al.*,[12] and a male predominance by Odukoya.[6]

In the present study, there was a slight sex difference (55.3%) in favor of men. Additionally in our series, OTs was 2.72 times more frequent in the mandible than in the maxilla which is in line with the results in most of the previous studies.[8,9,12-15]

OTs showed a peak incidence in 2nd decade of life. This is different from Jing study with greatest incidence in third decade of patients' life.[8]

The most frequent tumor in this study was Ameloblastoma (62.2%) with an incidence comparable to the reports of Wu and Chan (59.4%),[11] Komori (57%)[16] and Odukoya (58.5%).[6]

This contrasts the rates of series in American, Canadian, Mexican and Chilean populations with odontoma as the most frequent tumor.[4,14,17,18]

Altogether, it seems that ameloblastoma is less frequent in North America than in Africa and Asia. The tumor is quoted as comprising 1% of all tumors in Caucasians.[9]

Reichart *et al.*[19] in an extensive review of ameloblastoma found marked predilection for the mandible with the ratio of 5.4:1, which was similar to Avelar's study.[20] On the other hand, some researchers like Sriram *et al.*[9] reported a very high mandible to maxilla ratio (18:1). In this series ameloblastoma was frequently encountered in the molar-ramus region (30.65%) similar to some previous studies like Olgac V.[21]

The mean age of involvement in the recent study was 26.2%; this is similar to Sriram *et al.*,[9] Avelar *et al.*[20] Ochsenius *et al.*,[14] Fernandes *et al.*,[15] and Okada[3] studies. Reichart *et al.*,[19] in a review of 3677 cases of ameloblastoma, reported the average age of 39.1 years and 27.7 years for the initial diagnosis in industrialized and developing countries respectively. Based on these observations, they hypothesized that ameloblastoma develops 10 to 15 years earlier in developing countries than industrialized countries.[9,19]

According to Dodge *et al.*, this may be due to the accelerating aging process in developing countries owing to poor nutrition and health care.[22] In this study the most predominant clinical sign was expansion reported in 94.4% of cases which was in accordance with previous study.[22] Furthermore, multilocular radiolucency was the radiographic feature in 77.6% of patients. There was a male predominance (55%) in our results similar to some previous studies; the predominance however, contradicted other studies[2,7,13,20,23] and one research found equal distribution between genders.[24]

Odontoma was the second most common odontogenic tumor (14.3%) in this series similar to Olgac V' study in Istanbul,[21] Fernandes AM's[15] report in the Brazil, but different from W. Jing *et al.*'s[8] study according to which odontoma was the 3rd most common odontogenic tumor after ameloblastoma and keratocystic odontogenic tumor respectively. Buchner[2] also reported the largest series of this tumor (826 cases).

The sex distribution in the present study was 59.3% for female and 40.7% for male which was similar to W Jing *et al.* study.[8] Odontoma tended to occur more in the maxilla (59.3%) although most studies showed an almost equal distribution in the maxilla and mandible.[2,20,25] This tumor was observed mainly in young people with a mean age of 25.3 years which is consistent with reports from elsewhere.[14,15,25]

It is difficult to perform a comparative analysis of odontoma frequency because these tumors are interpreted as developmental malformation or hamartoma rather than true neoplasms. The lesion is usually discovered on routine radiographic examination without any clinical sign.

After odontoma, odontogenic myxoma was the third most common tumor (10.6%) in this series just like those of Fernandes AM.[15] The frequency was in agreement with Ochsenius *et al.*,[14] and Tumme *et al.*,[25] studies, but contrasted with Buchner report indicating a very low incidence.[2] Elison NM[26] and Adebayo EO[23] reported myxoma as the second most common tumor.

In this study, patients ranged in age from 9 to 51(mean 24.9), which is almost similar to Elison NM *et al.* Tumors in this study similar to Sriram G *et al.*,[9] W Jing *et al.*,[8] and Elison NM *et al.*[26] showed a greater predilection for females (55%); this contrasts with Buchner *et al.*,[2] work. Besides, in this study similar to Santos JN[12] and Olgac V *et al.*,[21] there was a significant predilection for maxilla (55%).

Adenomatoid odontogenic tumor (AOT) constitutes 2.6% of all OTs in this study which is comparable to some other countries like Hong Kong where Swasdison *et al.*,[27] noted a 5.3% frequency in the population. In China however, with (8.3%)[5] and in Srilanka with 8.6%[3] the frequency was greater but it was lower in Malaysia.[28]

AOT was seen more in maxilla (60%) and in females which is different from the findings of Siar[28] and Fernandes *et al.*[15] Most of the present cases were follicular type with a well-defined unilocular radiolucency surrounding un-erupted tooth and the most common clinical presentation was swelling (80%), like other previous reports.[1,23,29-31]

Cases of calcifying epithelial odontogenic tumor (CEOT) were rare with a frequency of 1.6% in this series which shows a lower rate of frequency compared with the Egyptian people (3.7%)[13] and a similar rate with the Nijerian series (1.8%).[5] These findings confirm the rarity of the lesion. Similar to the previous reports, in this study all of the 3 cases were seen in the mandible.

The rarity of cementoblastoma (1.6%) as an ectomesenchymal tumor in this study was comparable to the findings reported in Israel[32] and other countries.[4,6,11,14,15,18,21,23,24,31] The mean age of 3 cases was 16 with a slight predilection for mandible and males.

Ameloblastic carcinoma was also rare, constituting 1.01% of all OTs Which agrees with internationally published studies from Egypt,[13] Hong Kong[4] and Nigeria[5] (0.6 to 5%) but differs from the results on the Estonian population with 16% frequency.[21] Both cases of this tumor were seen in the second decade of life with a slight predilection for the maxilla.

3 cases of ameloblastic fibro-odontoma were seen in this series with a frequency of 1.6%. The mean age of cases was 10.6 years with a slight predilection in male and mandible.

There was also one case of ameloblastic odontoma (0.53%) in the mandible of 25-year old male with pain and expansion.

CONCLUSION

According to this 10-year retrospective study on OTs in Iran, the most common tumor was ameloblastoma followed by odontoma and odontogenic myxoma. Retrospective analysis of the epidemiologic characteristic of the OTs in different population and countries are necessary to provide better understanding of these lesions. The results can be extremely useful for both oral and maxillofacial surgeons and pathologists.

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How to cite this article: Taghavi N, Rajabi M, Mehrdad L, Sajjadi S. A 10-year retrospective study on odontogenic tumors in Iran. Indian J Dent Res 2013;24:220-4.

Source of Support: Nil, **Conflict of Interest:** None declared.