

# Odontogenic Tumors: A 5 Years Retrospective Epidemiological and Pathological Study

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## ABSTRACT

**Background:** This is a 5 years retrospective study in a referral centre to evaluate the epidemiological and pathological features of odontogenic tumors and to compare it with those of other studies in the literature.

**Methods and Material:** This is a 5 year retrospective study from December, 2011 to November 2016 in a referral centre . A total 75 cases of odontogenic tumors have been retrieved from the archives. Their epidemiological and pathological profiles are noted and compared to other studies.

**Result:** It is found that odontogenic tumors are mostly seen in 2nd to 5th decade and with average age of presentation of 28.7 years. Males are affected 1.14 times more frequently than females and mandible is involved 3 times more frequently than maxilla. Only 1 (1.3%) malignant odontogenic tumor is found. Solid multicystic ameloblastoma is the most common (34.7%) histological type followed by ossifying fibroma (20%) and Keratocystic odontogenic tumor (13.3%). Ameloblastomas are most commonly seen in 2nd to 4th decade with a peak in 3rd decade and average age of 27.3 years. They show slight male predominance (male to female ratio M:F ratio of 1.3:1) and marked predilection for mandible (9 times). Average tumor size is 5.3 cm . Ossifying fibromas show a wide age of distribution of 6 year - 60 year with mean age of 32.8 year. It show slight female predominance with M:F ratio of 0.87:1 and predilection for mandible. Average tumor size is 3.8cm. Keratocystic odontogenic tumors also show a wide age of distribution from 2nd to 6th decade with average age of 35 year. Male are affected 2.3 times more frequently than female and mandible is involved 3.5 times more commonly than maxilla. Average size of the tumors is 3.36 cm.

**Conclusion:** Most of the findings in the present study are consistent with those of many other studies conducted in India, other Asian countries and Africa. However, the major difference of the present study with all other studies is the high frequency of ossifying fibromas in the present study which is not seen in any other previous study.

**Keywords:** *Odontogenic, Ameloblastoma, Ossifying, Fibroma, Keratocystic.*

## Introduction

Odontogenic tumors are those tumors that arises from and so resemble tooth forming tissues. Their spectrum ranges from tumor like hamartomas and cystic tumors resembling non neoplastic cysts to locally aggressive benign neoplasm and occasional malignant neoplasm<sup>(1)</sup>. They follow a wide geographic and racial variation. There are many studies from different part of world demonstrating their epidemiological and clinicopathological profiles<sup>(2-17)</sup>. Several studies from India, mainly from southern (Kerala and Andhra Pradesh) and western Part (Maharashtra) have illustrated their features in these region<sup>(2,3,4,5,6,8)</sup>. However, there are very limited data of these neoplasms from the rest of India. Here, we have presented the demographical and pathological features of 75 odontogenic tumors and have compared these with other studies from India and different part of world.

## Methods and Material

This is a 5 year retrospective study from December, 2011 to November, 2016 in a referral pathology centre. A total

75 cases of odontogenic tumors have been retrieved from the archives. The corresponding case record sheets, slides and blocks are retrieved. The old sections (H & E stained) as well as new sections cut from the block and stained by H & E stain are reviewed by two pathologists. There are 30 hemimandibulectomy, segmental mandibulectomy and segmental maxillectomy specimens and 45 small biopsies. Only tumors with confirmed odontogenic origin are included in the study. In case of ossifying fibroma radiologically confirmed intraosseous tumors are included and peripheral ossifying fibromas are excluded. The tumors are analyzed for histological types, age, sex, site distribution and size. Recall bias of the patients cannot be excluded in recording of age. Only whole excised specimens are considered for estimation of tumor size. The tumors are classified on the basis of WHO classification of odontogenic tumors, 2005.<sup>(1)</sup> The data are presented in tabulated format for descriptive analysis. The results are compared to other similar studies from other parts of India, Asian countries outside India, Africa, North and South America.

### Result

Table 1 shows the histological types of the tumors. Only 1 case (1.3%) of malignant odontogenic tumor, primary intraosseous squamous cell carcinoma is found. There are 26 cases (34.7%) of solid multicystic type of ameloblastoma which is the most common type. Ossifying fibroma (15 cases, 20%) is the next most common one followed by Keratocystic odontogenic tumor (10 cases, 13.3%). Of the 26 solid multicystic ameloblastomas 20 were follicular, and 3 each were plexiform and acanthomatous ameloblastomas.

Table 2 shows that although odontogenic tumors are prevalent in all the age groups upto 60 years, it is more common in 2nd to 5th decade with a peak in the 2nd and 3rd decade. Average age of presentation is 28.7 years and range is 6 years – 60 years.

Table 3 shows the sex distribution of the cases. It is seen that male are slightly more affected than female with a male to female ratio of 1.14:1.

Table 4 shows that 56 odontogenic tumors have involved mandible, 18 have involved maxilla and 1 tumor has

involved both mandible and maxilla. So, mandible is 3.1 times more frequently involved than maxilla.

Information about size of the tumors are available in 16 of 30 ameloblastomas, in 8 of 9 Keratocystic odontogenic tumors and in only a few of others tumors. It is found that ameloblastomas show marked variation of size ranging from 3cm to 10cm with average size of 5.3cm. Keratocystic Odontogenic tumors show less variation in tumor size ranging from 2.5cm to 4cm with smaller average tumor size of 3.3cm

### Discussion

There are many studies of odontogenic tumors found in literature. Some of these are presented along with the present study in the table 5.

The figures in brackets indicate total number of cases in that study.

**Age Distribution:** Odontogenic tumors are the most frequent in 2<sup>nd</sup> and 3<sup>rd</sup> decades of age.<sup>[2-7,10,12,13,17]</sup> In the present study, the most common age group affected are 2<sup>nd</sup> to 5<sup>th</sup> decade of age with a peak in 2<sup>nd</sup> and 3<sup>rd</sup> decade

**Table 1: Distribution of the cases according to histological types [according to WHO classification of odontogenic tumors, 2005<sup>(1)</sup>].**

Histological types	Frequency	% of all
Primary Intraosseous squamous cell carcinoma (PIOSCC)	1	1.3%
Ameloblastoma, solid and multicystic type (AMEL-S)	26	34.7%
Ameloblastoma, desmoplastic type (AMRL-D)	1	1.3%
Ameloblastoma, unicystic type (AMEL-U)	3	4%
Calcifying epithelial odontogenic tumor(CEOT)	4	5.3%
Adenomatoid odontogenic tumor (AOT)	4	5.3%
Keratocystic odontogenic tumor (KCOT)	10	13.3%
Ameloblastic fibroma (AF)	2	2.7%
Odontoma (OD)	4	5.3%
Calcifying cystic odontogenic tumor (CCOT)	5	6.7%
Ossifying fibroma (OSF)	15	20%

**Table 2: Age distribution of the cases.**

Histological type	Age in years						Average age
	10	>10-20	>20-30	>30-40	>40-50	>50-60	
AMEL	0	8	12	7	2	1	27
PIOSCC	0	0	1	0	0	0	
CEOT	0	2	0	0	2	0	32
AOT	0	3	1	0	0	0	15
KCOT	0	4	0	3	1	2	35
AF	0	0	0	0	2	0	50
OD	2	1	1	0	0	0	16.5
CCOT	1	2	1	0	1	0	22.6
OSF	1	2	4	3	3	2	32
<b>Total</b>	<b>4</b>	<b>22</b>	<b>20</b>	<b>13</b>	<b>11</b>	<b>5</b>	<b>28.7</b>

**Table 3: Sex distribution of the cases**

Histological types	Male	Female	Male : Female
AMEL	17	13	1.3:1
PIOSCC	0	1	-
CEOT	4	0	-
AOT	0	4	-
KCOT	7	3	2.3:1
AF	2	0	-
OD	2	2	1:1
CCOT	1	4	0.25:1
OSF	7	8	0.87:1
<b>Total</b>	<b>40</b>	<b>35</b>	<b>1.14:1</b>

**Table 4: Site distribution of the tumors.**

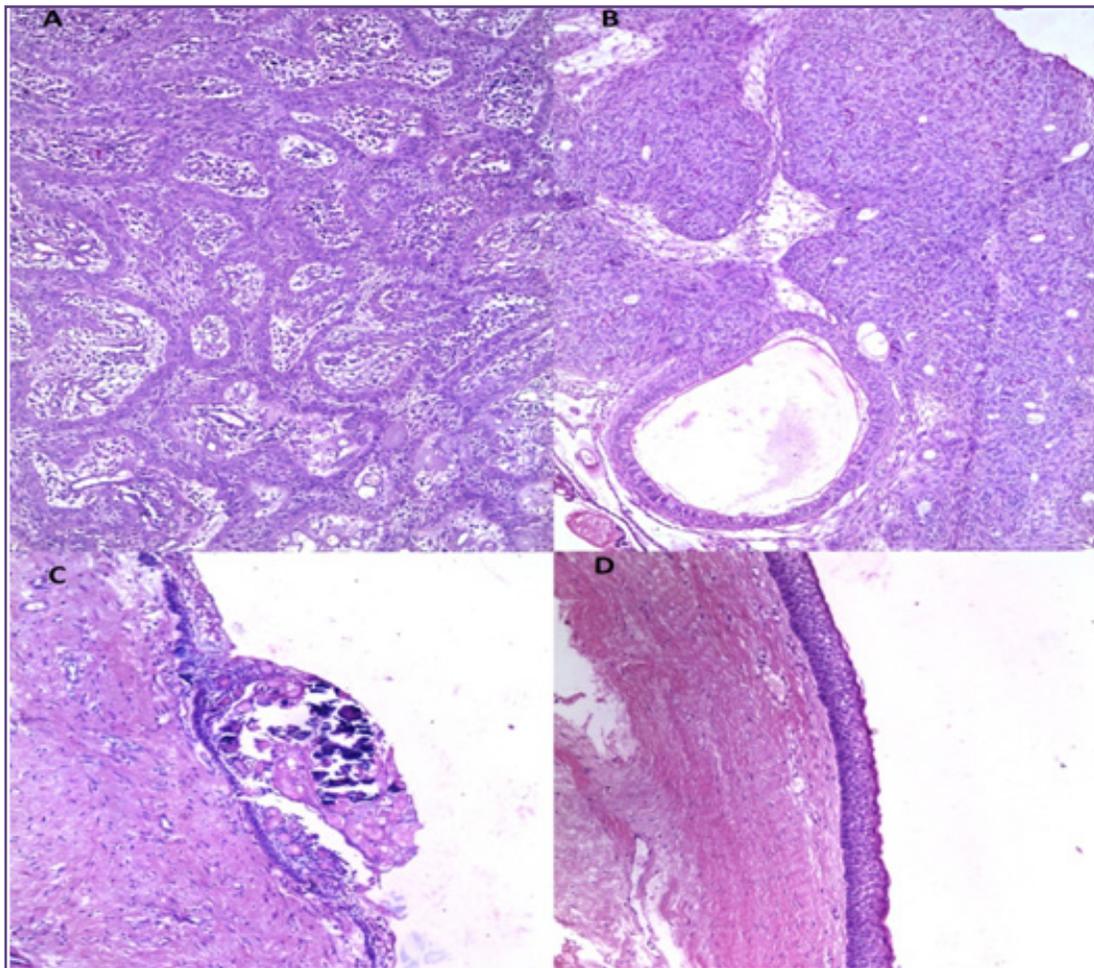
Histological type	mandible	Maxilla	Both	Mandible:Maxilla
AMEL	27	3	0	9:1
PIOSCC	1	0	0	-
CEOT	2	2	0	1:1
AOT	3	1	0	3:1
KCOT	7	2	1	3.5:1
AF	0	2	0	-
OD	3	1	0	3:1
CCOT	2	3	0	0.67:1
OSF	11	4	0	2.75:1
<b>Total</b>	<b>56</b>	<b>18</b>	<b>1</b>	<b>3.1:1</b>

**Table 5: Comparison of the present study with other studies available in literature.**

Studies	Average age (years)	Sex ratio (M:F)	Mandible: maxilla ratio	Most common histological type (%)
Pandiar D et al(395) (Kerala, India) <sup>[2]</sup>	32.69	1.4:1	2.43:1	KCOT(35.9), AMEL(25.9), CCOT(10.6)
Deepthi P V et al (305) (Southern Kerala, India) <sup>[3]</sup>	33.7	1.19:1	3.12:1	AMEL(50.2), KCOT(24.3)
Mullapudi S V et al (77) (Hyderabad, India) <sup>[4]</sup>	25	1.14:1	4.5:1	AMEL(71.4), AOT(8.5), CEOT(7.1)
Ramchandra S et al (76) (Andhra Pradesh, India) <sup>[5]</sup>	39.5	1.36:1	1.78:1	AMEL(68.42), OD(7.89)
Sriram G et al(250) (Mumbai, India) <sup>[6]</sup>	-	-	-	AMEL(61.5), AOT(12.5), Myxoma(6)
Kadashetti V et al (102) (Wardha, Maharashtra, India) <sup>[7]</sup>	-	4:1	4.7:1	KCOT(47.05), AMEL(36.2), OD(6.86),
Varkhede A et al (60) (Mumbai, India) <sup>[8]</sup>	-	1:1	2.53:1	AMEL(60.7), OD(20), AOT(10)
Guerrisi et al (153)† (Argentina) <sup>[9]</sup>	12.7	2:1	-	OD(50.9), AMEL(18.3), Myxoma(18.5)
Goteti S H et al (85) (Libya) <sup>[10]</sup>	29	1.2:1	2:1	AMEL(28.2), KCOT(25.2), OD(19.9)

Studies	Average age (years)	Sex ratio (M:F)	Mandible: maxilla ratio	Most common histological type (%)
Buchner A et al (1088) (California, USA) <sup>[11]</sup>	-	-	-	OD(75.9), AMEL(11.7), Myxoma(2.2)
Santos J N et al (127) (Brazil) <sup>[12]</sup>	26.6	0.57:1	1.35:1	OD(50.4), AMEL(30.7), AOT(8.67)
Taghavi N et al (188) (Iran) <sup>[13]</sup>	-	1.2:1	2.7:1	AMEL(62.2), OD(14.3), Myxoma(10.64)
Ajayi O F et a (92)† (Nigeria) <sup>[14]</sup>	14.9	1:1	2.7:1	AMEL(48.9), AOT(19.6), Myxoma(8.7)
Worawongvasu R et al (590) (Thailand) <sup>[15]</sup>	-	-	-	AMEL(35.0), KCOT(24.2), OD(15.2)
Mosqueda-Taylor A et al (349) (Mexico) <sup>[16]</sup>	-	-	-	OD(34.6), AMEL(23.7), Myxoma(17.7)
Saghravanian N et al (165) (Iran) <sup>[17]</sup>	26.3	0.85:1	2.44:1	AMEL(42.4), OD(26.7), AOT & Myxoma(9.1%) each
Present study (75)	28.7	1.14:1	3.1:1	AMEL(40.0), OSF(20), KCOT(13.3)

†Conducted in children and adolescents only



**Fig. 1: Photomicrograph of odontogenic tumors. A. Plexiform ameloblastoma (10X,H & E) B. Adenomatoid odontogenic tumor (10X, H & E) C.Calcifying cystic odontogenic tumor (10X,H & E) D. Keratocystic odontogenic tumor (10X,H & E).**

and average age of 28.7 years. This is consistent with the studies by Mullapudi SV et al, Goteti SH et al, Santos JN et al and Saghravanian N et al.<sup>[4,10,12,17]</sup> However some other Indian study showed a slightly older age (Average age 4-11 years older) than the present study.<sup>[2,3,5]</sup>

**Sex Distribution:** Most of the studies till now have shown that odontogenic tumors are more common in male than female.<sup>[2,3,4,10,13]</sup> However, a few studies have shown either equal prevalence or slight female predominance.<sup>[8,12,14,17]</sup> We have found a male to female ratio of 1.14:1 which is comparable to studies by Deepthi P V et al, Mullapudi SV et al, Goteti SH et al and Taghavi N et al.<sup>[3,4,10,13]</sup>

**Site Distribution:** All the studies till now have demonstrated mandible as the predominant site of odontogenic tumors. Preferred mandibular location is also noted in the present study with a mandible to maxilla ratio of 3.1:1. Several Indian studies and studies done outside India have also shown a mandible to maxilla ratio akin to our study.<sup>[2,8,13,14,17]</sup>

**Histological Types:** Malignant odontogenic tumors are rare.<sup>[2,6,12,14-19,25]</sup> The present study shows only 1 malignant tumors (1.3% of all), i. e. primary intraosseous squamous cell carcinoma.

We have found ameloblastoma as the most common histological type followed by ossifying fibroma and keratocystic odontogenic tumor. Calcifying cystic odontogenic tumor, calcifying epithelial odontogenic tumor, adenomatoid odontogenic tumor and odontoma show low and almost equal prevalence. Ameloblastic fibroma is the least common type.

It is seen that the frequency of different histological types of odontogenic tumor show a considerable geographic variation. Most of the western studies show odontomas as the most frequent type.<sup>[9,11,12,16]</sup> But most of the Indian studies, Asian studies outside India and African studies have found ameloblastoma as the most frequent type.<sup>[3-5,8,10,13-15,17]</sup> A few Indian studies, occasional Asian studies outside India and occasional western studies have found keratocystic odontogenic tumor as the most frequent type.<sup>[2,7,18,19]</sup> Like most other Indian studies we have also found ameloblastoma as the most common histological type. Solid multicystic ameloblastoma constitute 34.7% (26 of 75 cases) of all odontogenic tumors, being the most common type. They mostly show follicular pattern (20 of 26 cases) and a few show plexiform and acanthomatous pattern (3 of each type). Unicystic ameloblastoma are known for earlier age of onset and better clinical outcomes.<sup>[20,21]</sup> Unicystic ameloblastoma comprises of 4% (3 of 75 cases) of all odontogenic tumors in our study, which is comparable to

Ajayi OF et al.<sup>[14]</sup> 5.4% (5 of 92) cases in their study were unicystic ameloblastoma. Only 1 case of desmoplastic ameloblastoma is found in the present study.

Ameloblastomas in our study have mostly affected people in the 2<sup>nd</sup> to 4<sup>th</sup> decades with a peak in 3<sup>rd</sup> decade and a mean age of 27.3 year. This is consistent with studies by Deepthi PV et al, Goteti SH et al, Taghavi N et al and Mullapudi SV et al.<sup>[3,4,10,13]</sup> We have found slight male predominance (male to female ratio of 1.3:1) for ameloblastoma as like many other studies.<sup>[2,4,10]</sup> A marked predilection of ameloblastoma for mandible is shown by many studies and our study have proved it again.<sup>[2,5,12,13]</sup> Average size of ameloblastoma is found to be 5.3cm. Ossifying fibroma is classified as a bone-related lesion by World Health Organization classification of odontogenic tumor, 2005.<sup>[1]</sup> It arises from the stem cell in the periodontal ligament that can give rise to both fibrous tissue and bone. Radiologically, it present as demarcated lesion in jaw bone with radio-lucency or mixed radio-lucency and radio-opacity. An unusually high proportion (20%, 15 of 75 cases) of ossifying fibroma is found in the present study, making it the 2<sup>nd</sup> most common histological type, which was not found in any other previous studies. Ossifying fibromas are distributed over a wide age range of 6 year - 60 years with a mean age of 32.8 year. Similar age distribution was noted by Y Liu et al (age range 6 year to 57 year and mean age of 27.8 years) and De Andrade M et al (mean age of 33.7 year).<sup>[22,23]</sup> There is slight female predominance with male to female ratio of 0.87:1 in our study almost similar to Y liu et al (M:F=0.82:1) and only slightly different from De Andrade M et al (M:F=0.60). Majority of our cases have involved the posterior aspect of jaw with a mandible to maxilla ratio of 2.75:1. Marked mandibular preference was also observed in the other two above mentioned studies (mandible to maxilla ratio of 4:1 and 7:1) The average size of the tumor in the present study (3.8cm) is also comparable to that reported by de Andrade M et al (3.1cm)

There was a longstanding confusion whether keratocystic odontogenic tumor should be considered as a cystic lesion or neoplasm until WHO has included it in the list of odontogenic tumors in 2005 considering its local aggressive behaviour, histology and abnormal PTCH gene function.<sup>[24]</sup> Since then a few studies have found it as the most common histological type and many others as the second or third most common type.<sup>[2,3,7,10,15,18,19,25]</sup> We have found it as the third most common histological type comprising of 13.3% (10 of 75 cases). It has shown a wide age of distribution of second to sixth decade with average age of presentation of 35 years. Pandiar D et al have also found a similar age distribution.<sup>[2]</sup> In the present study male are affected 2.3 times more frequently than females.

Some studies in literature show male predominance while others show the reverse.<sup>[2,10,26]</sup> We have found Keratocystic odontogenic tumors to be more common in mandible than maxilla as like some other studies.<sup>[10,26]</sup>

The numbers of cases of other histological types are quite few and So comparison to other studies does not seem to be relevant.

## Conclusion

The demographic profile of the patients in the present study resembles to that of many other Indian studies and studies outside India. In case of histological types, ameloblastomas are the most common in the present study as like many other Indian studies, studies in other Asian countries or in Africa, but unlike studies in most of the western countries which show odontoma as the most frequent type. A major difference of our study with all other studies is the high frequency of ossifying fibroma in our study being the second most common histological type which is not found in any other previous studies.

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