Odontogenic tumors in Western India (Gujarat): Analysis of 209 cases

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Abstract

Objective: Odontogenic tumors show a distinct geographic variation. In 2005 a new WHO classification was published which included odontogenic keratocyst as one of the odontogenic tumors, renaming it as a keratocystic odontogenic tumor. To our knowledge there are only few studies based on 2005 classification in Asian subcontinent. This study was done to determine the relative frequency of odontogenic tumors in Gujarat and compare it with reports from other parts of the world.

Study Design: A retrospective study was designed. Necessary information was obtained from the records of the Oral Pathology Department, GDCH Ahmedabad. The histopathological diagnosis were re-evaluated according to the criteria of WHO histological classification 2005.

Results: A total of 209 cases were reported in just a short span of 5-years. The most frequent histologic type was ameloblastoma 47.4%, followed by Keratocystic odontogenic tumor (KCOT) 23.4%. Odontomas which are the most frequent odontogenic tumor in European and American subcontinent, accounted only 5.3% in this study.

Conclusion: In India ameloblastoma and KCOT are the most frequent odontogenic tumors, thus supporting the distinct geographic variation of these rare tumors.

Key Words: Odontogenic tumors, ameloblastoma, geographic variation.
Introduction
Odontogenic tumors are derived from tooth forming apparatus, either the epithelial or the ectomesenchymal or both. These tumors have a specific histological structure that reflects various stages of odontogenesis. They are rare, comprising only about 1% of all tumors in the jaw (1). They share two major characteristics, namely they arise from the tissue with the potential for differentiation into tooth or periodontal structures, and therefore found exclusively in the mandible and maxilla and, on rare occasions, the gingiva. Another variable but distinctive feature includes formation of tooth-related extracellular substances some of which may calcify and be visible on radiographs like odontome, ameloblastic fibrodentinoma, ameloblastic fibro-odontoma; they are a product of epithelial–mesenchymal interactions. The biological behaviour of these lesions ranges from hamartoma-like lesions and benign neoplasms to rare, aggressive, malignant tumors.

Although many retrospective studies have been conducted in Africa (2), Asia (3), Europe (4) and America (5) but they are based on 1992 WHO classification. A new classification was proposed in 2005, which included odontogenic keratocyst as a benign odontogenic tumor. To our knowledge, there are no reports on the frequency of these tumors from Gujarat. Therefore present study was planned to analyze retrospectively the demographic data of odontogenic tumors in Gujarat based on new WHO classification 2005 and to compare it with other datas.

Material and Methods
The study material was obtained from the Department of Oral pathology Government Dental College and Hospital Ahmedabad. A retrospective study of odontogenic tumors obtained over a period of 5 years from 1st January 2004 to 31st December 2008 was designed. Information including age, sex, site of tumor and frequency were obtained. Slides stained with haematoxylin and eosin were reviewed. The diagnoses were re-evaluated according to the criteria of WHO histological classification 2005. For tumor location the following scheme was used. The maxilla was divided into 6 anatomical regions, 3 on either side: anterior (from the midline to the distal surface of the canine), premolar (from the mesial aspect of the first premolar to the distal side of the second premolar), and molar (from the mesial aspect of the first molar distally). The mandible was divided into 3 anatomical regions on each side: anterior and premolar as described above, and molar (from the mesial aspect of the first molar) to ramus (upper portion of ramus above the occlusal plane).

Results
During the 5 year period from January 2004 to December 2008 a total of 209 cases of odontogenic tumors were diagnosed. All the odontogenic tumors were benign. The distribution of histological types and frequency of odontogenic tumors is presented in Table 1. Some of the odontogenic tumors were divided into further subtypes. The ameloblastoma were divided into 2 histologic subtypes: solid/multicystic ameloblastoma (SMA) and unicystic ameloblastoma(UA), odontomes were divided as compound and complex type whereas the peripheral odontogenic tumors were divided as peripheral calcifying odontogenic tumor (peripheral CCOT) and peripheral odontogenic fibroma as these were the only two peripheral variants noted.

<table>
<thead>
<tr>
<th>Histologic types</th>
<th>Frequency</th>
<th>Percent (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Ameloblastoma</td>
<td>99</td>
<td>47.4</td>
</tr>
<tr>
<td>a. Unicysic (UA)</td>
<td>70</td>
<td>33.5</td>
</tr>
<tr>
<td>b. Solid/Multicystic(SMA)</td>
<td>29</td>
<td>13.9</td>
</tr>
<tr>
<td>2. Keratocystic odontogenic tumor (KCOT)</td>
<td>49</td>
<td>23.4</td>
</tr>
<tr>
<td>3. Adenomatoid odontogenic tumor (AOT)</td>
<td>16</td>
<td>7.7</td>
</tr>
<tr>
<td>4. Calcifying cystic odontogenic tumor (CCOT)</td>
<td>13</td>
<td>6.2</td>
</tr>
<tr>
<td>5. Odontome</td>
<td>11</td>
<td>5.3</td>
</tr>
<tr>
<td>a. Complex</td>
<td>2</td>
<td>1.0</td>
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<tr>
<td>b. Compound</td>
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<td>4.3</td>
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<tr>
<td>6. Odontogenic Myxoma</td>
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</tr>
<tr>
<td>7. Cementoblastoma</td>
<td>6</td>
<td>2.9</td>
</tr>
<tr>
<td>8. Ameloblastic fibroma</td>
<td>2</td>
<td>1.0</td>
</tr>
<tr>
<td>9. Calcifying epithelial odontogenic tumor (CEOT)</td>
<td>3</td>
<td>1.4</td>
</tr>
<tr>
<td>10. Peripheral odontogenic tumors</td>
<td>3</td>
<td>1.4</td>
</tr>
<tr>
<td>Peripheral Calcifying odontogenic cyst</td>
<td>1</td>
<td>0.4</td>
</tr>
<tr>
<td>a. Peripheral calcifying cystic odontogenic tumor (Peripheral CCOT)</td>
<td>2</td>
<td>1.0</td>
</tr>
<tr>
<td>Total</td>
<td>209</td>
<td>100.0</td>
</tr>
</tbody>
</table>

Table 1. Showing histologic types and frequency of odontogenic tumors
In this study the other less common types were calcifying epithelial odontogenic tumour (CEOT) and ameloblastic fibroma (AF) accounting for 3 cases (1.4%) and 2 cases (1%) each. 3 cases (1.4%) of peripheral odontogenic tumor were also noted.

Out of total 209 cases, 176 cases (84.2%) were found during second, third and fourth decades, 14 cases (6.7%) in fifth decade, 13 cases (6.2%) in the sixth and seventh decade of life and only 6 cases (2.9%) in first decade. The high prevalence of odontogenic tumors was observed in young age while rare in children below 10 years of age as in Table 2. Among all the odontogenic tumors, 118 (56.5%) were in males and 91 (43.5%) in females, with an overall male:female ratio of 1.3:1 which shows male predominance but unicystic ameloblastoma appeared as

| Age (years) | 0 to 4 | 5-9 | 10-14 | 15-19 | 20-24 | 25-29 | 30-34 | 35-39 | 40-44 | 45-49 | 50-54 | 55-59 | 60-64 | 65-69 | Total | Mean age | SD |
|------------|-------|-----|-------|-------|-------|-------|-------|-------|-------|-------|-------|-------|-------|-------|-------|-------|--------|------|
| UA         | -     | 4   | 6     | 11    | 16    | 12    | 6     | 7     | 4     | -     | 2     | -     | 1     | 1     | 70    | 25.9   | 12.1 |
| SMA        | -     | -   | 2     | 2     | 9     | 6     | 4     | 1     | 2     | 1     | 2     | -     | -     | -     | 29    | 33.2   | 10.4 |
| Kcot       | -     | 2   | 7     | 12    | 10    | 8     | 3     | 4     | -     | 2     | -     | -     | -     | 1     | 49    | 28.2   | 10.7 |
| Aot        | -     | 1   | 4     | 9     | 2     | -     | -     | -     | -     | -     | -     | -     | -     | -     | 16    | 15.8   | 3.8  |
| Ccot       | -     | -   | -     | 2     | -     | 3     | 4     | 1     | 1     | -     | 1     | 1     | -     | -     | 13    | 33.2   | 11.3 |
| Compound   | -     | -   | -     | 3     | 1     | 1     | -     | -     | 1     | -     | -     | -     | -     | -     | 9     | 25.3   | 10.5 |
| Complex    | -     | -   | -     | 1     | -     | -     | -     | -     | -     | -     | -     | -     | -     | -     | 2     | 19.5   | 2.5   |
| Odontogenic myxoma | -   | 1   | -   | -     | 2     | 3     | 1     | -     | -     | -     | -     | -     | -     | -     | 7     | 23.4   | 7.4   |
| Cementoblastoma | -   | -   | 2   | -     | 3     | -     | -     | -     | 1     | -     | -     | -     | -     | -     | 6     | 22.0   | 10.0  |
| Ameloblastic fibroma | -     | 1     | 1     | -     | -     | -     | -     | -     | -     | -     | -     | -     | -     | -     | 2     | 14.5   | 2.5   |
| Ceot       | -     | -   | -     | -     | 3     | -     | -     | -     | -     | -     | -     | -     | -     | -     | 3     | 27.0   | 0.00  |
| Peripheral Ccot | -   | -   | 1   | -     | -     | -     | -     | -     | -     | -     | -     | -     | -     | -     | 1     | 17.0   | 0.00  |
| Peripheral Odontogenic Fibroma | -   | -   | -     | 1     | -     | -     | -     | -     | -     | -     | -     | -     | -     | -     | 2     | 29.5   | 12.5  |
| Total      | 0     | 6   | 15    | 38    | 41    | 15    | 41    | 26    | 21    | 12    | 2    | 3     | 1     | 2     | 209   |        |        |

Table 2. Showing age wise distribution of odontogenic tumors

(3.3%) and 6 cases (2.9%) respectively. In this study the other less common types were calcifying epithelial odontogenic tumour (CEOT) and ameloblastic fibroma (AF) accounting for 3 cases (1.4 %) and 2 cases (1%) each. 3 cases (1.4%) of peripheral odontogenic tumor were also noted.

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<table>
<thead>
<tr>
<th>DIAGNOSIS</th>
<th>Male</th>
<th>Female</th>
<th>Total</th>
<th>TOTAL</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.Ameloblastoma</td>
<td>51</td>
<td>48</td>
<td>99</td>
<td>47.4</td>
</tr>
<tr>
<td>a. Unicystic</td>
<td>34</td>
<td>36</td>
<td>70</td>
<td>33.5</td>
</tr>
<tr>
<td>b. Solid/Multicystic</td>
<td>17</td>
<td>12</td>
<td>29</td>
<td>13.9</td>
</tr>
<tr>
<td>2.Keratocystic odontogenic tumor</td>
<td>31</td>
<td>18</td>
<td>49</td>
<td>23.4</td>
</tr>
<tr>
<td>3.Adenomatoid odontogenic tumor</td>
<td>8</td>
<td>8</td>
<td>16</td>
<td>7.7</td>
</tr>
<tr>
<td>4.Calcifying cystic odontogenic tumor</td>
<td>10</td>
<td>3</td>
<td>13</td>
<td>6.2</td>
</tr>
<tr>
<td>5.Odontome</td>
<td>8</td>
<td>3</td>
<td>11</td>
<td>5.3</td>
</tr>
<tr>
<td>a. Complex</td>
<td>1</td>
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<tr>
<td>9.Calcifying epithelial odontogenic tumor</td>
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<td>0</td>
<td>3</td>
<td>1.4</td>
</tr>
<tr>
<td>10.Peripheral odontogenic tumor</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>1.4</td>
</tr>
<tr>
<td>a. Peripheral calcifying cystic odontogenic tumor</td>
<td>1</td>
<td>0</td>
<td>1</td>
<td>0.5</td>
</tr>
<tr>
<td>b. Peripheral odontogenic fibroma</td>
<td>0</td>
<td>2</td>
<td>2</td>
<td>1.0</td>
</tr>
</tbody>
</table>

Table 3. Showing gender wise distribution of odontogenic tumors
commonest tumor in females as shown in Table 3. Out of total cases of odontogenic tumors, 145 (69%) tumors were encountered in the mandible and 64 (31%) in the maxilla which showed predilection for mandible and mandible to maxillary ratio was 2.3:1. From them, odontogenic myxoma, AOT, CCOT and odontome showed more predilection for maxilla. The most frequently affected area in the mandible was molar region (73%) while in the maxilla it was anterior region (50%). Commonest tumors in mandibular molar area were ameloblastoma (both subtypes UA and SMA) and KCOT whereas most common tumor in the maxillary anterior region were AOT, CCOT and Odontome. The site distribution is summarized in Table 4.

**Discussion**

The literature on the relative frequency of odontogenic tumors from India is very less reported. The present study represents a large number of cases of odontogenic tumors over a short span of 5 years from India specifically in Gujarat state. In this study some interesting differences regarding their prevalence was noted. Amongst all the odontogenic tumors, ameloblastoma 47.4% was the most commonly encountered tumor followed by KCOT 23.4%. This finding was quite contrasting with the findings of Avelar et al. (6) who showed KCOT 30% as the most prevalent odontogenic tumor followed by ameloblastoma 23.7%. Most of the odontogenic tumors (84.2%) were found during 2nd to 4th decade which showed high prevalence during young age whereas only 3% cases were seen below 10 years of age. This might be because most of odontogenic tumors are commonly associated with permanent teeth and crown formation of most of the permanent teeth is completed by the age of 4 to 5 years. In our study up to the age of 4 years no odontogenic tumor was seen which indicate that odontogenic tumors probably develop after crown formation is complete. Odontogenic tumors were more common in males with male: female ratio of 1.3:1 which is also reported by Oduyoka (7), and also showed mandibular predilection with mandibular: maxillary ratio of 2.3:1 which is in agreement with Arotiba et al. (8).

Ameloblastoma being the most frequent tumor in the present study is similar to the report of Lu et al. (9) from China, Oduyoka (7) from Africa and Varkhede et al. (10) from India. In contrast, in Chili (11), Mexico (12) and Canada (13) ameloblastomas accounted for 20%, 24% and 18% respectively where the most common tumor encountered was odontoma, with rates of 44.7%, 34.6%, 46.0% respectively. This also strengthens the
belief that ameloblastomas are more common in Asians
and Africans compared with Caucasians. India to be a
developing country, the age distribution of ameloblas-
toma was less with mean age of 28 years which was
consistent with the results of Reichart et al. (14) who
reported the average age of initial diagnosis in indus-
trialized countries to be 39.1 years compared with 27.7
years from developing countries. Based on these obser-
vations, they hypothesized that persons from developing
countries develop ameloblastomas 10-15 years earlier
than in industrialized countries. Dodge (2) proposed that
this variation among countries may be due to the acce-
lerated aging process in developing countries owing to
poor nutrition and health care. When comparing the age
distribution among the two subtypes of ameloblastomas
in this study, an obvious contrast between SMA and UA
was found. The mean age of the patients with unicystic
ameloblastoma (25.9 years) was much lower than that of
the patients with classical solid/multicystic ameloblasto-
as (33.2 years) which was similar to that reported by
Ackermann et al. (15) in UA i.e 23.8 years. Ameloblasto-
toma showed male predilection with male female ratio
of 1.1:1 whereas unicystic ameloblastoma showed fema-
le predilection (51.4%). 73 % of ameloblastomas tended
to occur in the posterior mandible, which is consistent
with previous reports (11) but ameloblastomas are
seen more frequently in the mandibular anterior region
among Blacks (21.6%) compared to Caucasians 12.6%
and Asians 11.9% (14). Adekeye and Lavery (16) repor-
ted predilection for mandibular anterior region among
blacks. In our study, it was seen in only 6% of cases.
KCOT appeared the second most prevalent tumor in this
study (23.4%) with the peak age of occurrence in second
and third decades of life with male predominance (63%)
and mandibular molar area (55%) was the most affec-
ted site. Because of the recent reclassification in 2005, it
was not possible to compare it with that of other studies
in literature except with the findings of Avelar et al. (6)
who showed KCOT (30%) as the most prevalent odon-
togenic tumor followed by ameloblastoma (23.7%).
In this study, the incidence was higher in males (63%)
which was similar to the findings of Ahlfors et al. (17)
but contrasting to study of Avelar et al. (6) who showed
female predilection (56.6%).
Adenomatoid odontogenic tumor made up 7.8% of all
odontogenic tumors in the present study which was si-
milar to the report of Lu et al. (9), Mosqueda-Taylor et
al. (12) and Oduyoka (7) but higher than the findings
of Daley et al. (13). It occurred in much younger age
(mean age 15.8 years) as compared to other odontogenic
tumors. Younger age of occurrence for AOT may be due
to the fact that this tumor is more frequent in anterior
region which might alert the individual at an earlier age.
Secondly, as most AOT are associated with unerupted
tooth so patient may seek consultation concerning fa-
ilure of the associated anterior teeth to erupt. Our series
presented equal sex predilection with male:female ratio
of 1:1 in contrast to Okada et al. (3) who showed fema-
le predominance. The most common site was anterior
maxilla (50%) followed by mandibular anterior region
(38%) with maxilla mandible ratio of 1.3:1.
The calcifying cystic odontogenic tumor (CCOT) was
seen with the frequency of 6.2%. There was higher in-
cidence in males (77%) and 62% of cases were in the
maxilla. These findings are similar to the findings of
Okada et al. (3)
Odontogenic myxoma was an uncommon tumor, with
a low relative frequency of 3.8%. This low relative fre-
quency is also reported by Kaffe et al.(18) The mean
age of occurrence of odontogenic myxoma (23.4 years)
was significantly earlier than that of SMA (almost 10
years earlier) in the present study. This may be due to
the fact that odontogenic myxomas are more aggres-
svie compared to ameloblastomas. In the present study,
odontogenic myxoma was more common in females
(57%) and showed maxillary predilection (71%) which
was quite contrasting to the findings of Ladeinde et al.
(19) who reported mandibular predilection.
We found an incidence of odontome as 5.3 %. This is
much lower than the rates in series from the USA (5),
Canada (13), and Germany (20) where odontome was
the most common odontogenic tumor. In present study,
compound odontome was more common (4.3%) than
complex type (1%) and there was predilection for maxi-
illary anterior region with maxilla to mandible ratio of
10:1.
The incidence of cementoblastoma was 2.9 % with equal
sex predilection and occurring most commonly in the
mandibular molar area (50%) whereas Lu et al. (9) re-
ported female predilection and most affected site being
similar to our study.
The incidence of the ameloblastic fibroma (AF) and that
of the calcifying epithelial odontogenic tumor (CEOT)
was 1% and 1.4 % respectively in the present study.
Only 2 cases of AF were seen confirming their rarity and
both in females, whereas all the 3 cases of CEOT were
seen in males. Both the lesions were in the mandible but
Santos et al. (21) have reported a higher frequency of
these tumors in the maxilla.
Peripheral odontogenic tumors accounted 1.4 % of the
odontogenic tumors in the present study. The low fre-
cquency of these neoplasms was also reported by Ide et
al. (22), which underlines their rarity.
Based on this collaborative retrospective study it was
observed that there is marked geographic differences in
relative incidences of various odontogenic tumors. Mo-
moreover as seen in the present study the incidence of odon-
togenic tumor is quite high in Gujarat. Unicystic amelo-
blastoma is the most common odontogenic tumor in this
environment whereas odontome is relatively rare.
References