A study of 1177 odontogenic lesions in a South Kerala population

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INTRODUCTION

Odontogenic tumors are derived from epithelial, ectomesenchymal and/or mesenchymal elements of the tooth-forming apparatus. These lesions range from hamartomatous to benign neoplasms to malignant tumors with metastatic potential. They are found exclusively within the maxillofacial skeleton (intraosseous) or in the gingiva overlying tooth-bearing areas or alveolar mucosa in edentulous regions. Odontogenic cysts are derived from the epithelium of the dental apparatus.

World Health Organization (WHO) published the classification of odontogenic tumors in 1971, and later revised it in 1992 and in 2005. In the 1992 classification,
calcifying odontogenic cyst was introduced as an odontogenic tumor. In 2005 WHO classification, odontogenic keratocyst was reclassified as keratocystic odontogenic tumor (KCOT). This has increased the frequency and distribution of odontogenic tumors. There seems to be regional variations in the distribution of odontogenic cysts and tumors in the literature. Very few studies have been reported among Asians, especially from India. Epidemiological data on odontogenic cysts and tumors are lacking from Kerala, the southern part of India. This study was undertaken to address the distribution of odontogenic cysts and tumors in a tertiary dental health-care center in southern part of Kerala.

SUBJECTS AND METHODS

Pathology archival records of the Department of Oral Pathology and Microbiology were reviewed and all the cases of odontogenic cysts and tumors from 1998 to 2012 were retrieved. The histopathologic diagnoses were confirmed by reviewing the hematoxylin and eosin stained slides and were reclassified according to the 2005 WHO classification. If any recurrence was noticed, it was considered as a single case. Cases with incomplete records were excluded from the study. The study variables were age, gender, site of the lesion and histopathology of odontogenic cysts and tumors. Descriptive statistical analysis was performed using the computer software, Statistical Package for Social Sciences (SPSS) version 16. IBM SPSS Software version 16.

RESULTS

Of the 7117 oral biopsies retrieved from the archives during1998-2012, 1177 were odontogenic lesions. About 305 odontogenic tumors (4.29% of total specimens) were found. Benign odontogenic tumors comprised 99.0% of odontogenic tumors and 1.0% were malignant odontogenic tumors. The most common tumor was ameloblastoma which constituted 50.2% of odontogenic tumors. Nearly 80.4% of ameloblastomas were solid, while unicystic and peripheral types formed 17.6% and 2.0%, respectively. KCOT formed the second most common tumor, comprising 24.3% of odontogenic tumors. Odontoma formed 13.1% of all cases. The frequency of odontogenic tumors is shown in Table 1.

Odontogenic tumors occurred in the age range of 4–74 years with a mean age of 33.7 ± 16.8 years. Of 153 ameloblastomas, 103 cases (67.32%) occurred in the third-fifth decades of life. 62.16% of KCOT occurred in the second to fourth decades. Odontoma (60%) showed a higher frequency in the second decade. A significant association (P = 0.01) was found between age and the histopathological diagnosis. The distribution of odontogenic tumors according to age is shown in Table 2.

Table 1: Distribution of the odontogenic tumors according to histopathologic types and gender

| Odontogenic Tumors | Count | Percentage | Male | Female | Male/Female |
|--------------------|-------|------------|------|--------|-------------|
| Ameloblastoma      | 153   | 50.2       | 72   | 81     | 1:1.13      |
| SOT                | 2     | 0.66       | 54   | 20     | 2:1         |
| CEOT               | 3     | 0.98       | 2    | 1      | 2:1         |
| AOT                | 12    | 3.93       | 6    | 6      | 1:1         |
| KCOT               | 74    | 24.3       | 0    | 1      | -           |
| AF                 | 1     | 0.33       | 1    | 1      | 1:1         |
| AFO                | 2     | 0.66       | 0    | 2      | -           |
| Odontoma           | 40    | 13.1       | 4    | 3      | 1.33:1      |
| CCOT               | 7     | 2.30       | 22   | 18     | 1.22:1      |
| OM                 | 2     | 0.66       | 1    | 1      | 1:1         |
| OF                 | 4     | 1.31       | 2    | 2      | 1:1         |
| Cementoblastoma    | 2     | 0.66       | 0    | 2      | -           |
| AC                 | 1     | 0.33       | 1    | 0      | -           |
| PIOSCC             | 2     | 0.66       | 1    | 1      | 1:1         |

SOT: Squamous odontogenic tumor, CEOT: Calcifying epithelial odontogenic tumor, AOT: Adenomatoid odontogenic tumor, KCOT: Keratocystic odontogenic tumor, AF: Ameloblastic fibroma, AFO: Ameloblastic fibro-odontoma, CCOT: Calcifying cystic odontogenic tumor, OM: Odontogenic myxoma, OF: Odontogenic fibroma, AC: Ameloblastic fibro-odontoma, PIOSCC: Primary intra osseous squamous cell carcinoma

Odontogenic tumors showed a slight male predilection. About 54.4% of cases were males and 45.6% were females. Ameloblastoma showed an almost equal distribution in males and females with a male to female ratio of 1.1.13. KCOT showed a male predominance with a male to female ratio of 2:1. Odontoma showed a slight male predilection.

Mandible was the most common jaw involved (76.07%). In 57.7% of odontogenic tumors, mandibular posterior region was involved. The distribution based on different anatomic sites is shown in Table 3.

Odontogenic cysts constituted 12.25% (872 cases) of oral biopsies over a period of 1998-2012. Radicular cyst was the most common odontogenic cyst and accounted for 75.11% of odontogenic cysts. Dentigerous cyst was the second most common cyst forming 17.2% of odontogenic cysts. Other cysts accounted for 7.67% of cases.

Overall, a slight male predilection was noticed as shown in Table 4. Nearly 55.2% of odontogenic cyst occurred in males and 44.8% in females. Radicular cyst showed almost equal gender distribution. Dentigerous cyst showed a female predilection.

Radicular cyst showed a peak incidence in third and fourth decades with a mean age of occurrence of 35.7 ± 16.7 years. The dentigerous cyst was more common in the second decade. The distribution of odontogenic cyst according to age is given in Table 5.

The maxilla was the most common jaw affected by the odontogenic cysts. In 46.1% of cases maxillary anterior region was involved. The next common site was a mandibular posterior.
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Radicular and dentigerous cyst occurred mostly in the maxillary anterior region.

**DISCUSSION**

Different odontogenic tumors show variation in clinical presentation and aggressiveness. In this study, odontogenic cysts and tumors constituted 16.54% of total oral biopsies. The frequency of odontogenic tumors was 4.29%. This is comparable to other studies from Asia,[6] higher than those from America,[7,8] but lower than those from Africa.[9] A racial difference could be the most possible attributable reason for these variations in the prevalence. Table 6 shows a comparison of various retrospective studies on odontogenic tumors reported from different regions.

The most common odontogenic tumor in this study is ameloblastoma (50.2%), similar to other studies from Asian[6,10] and African[9,11] countries. Clinicopathological studies from American[7,12] countries presented odontoma as the most common odontogenic tumor followed by ameloblastoma in the second position. In this study, odontoma is in the third position following ameloblastoma and KCOT, with a frequency of 13.1%. The lower prevalence of odontoma in the Asian and African population may be due to the lack of routine dental care as these tumors remain unnoticed for years. Moreover, after surgical removal these tumours might not been sent for histopathological examination.

| Odontogenic tumor | Age in decades |
|-------------------|----------------|
|                   | 0-9 | 10-19 | 20-29 | 30-39 | 40-49 | 50-59 | 60-69 | 70-79 | Mean  | SD    |
| Ameloblastoma     | 1   | 14    | 40    | 29    | 34    | 16    | 15    | 4     | 37.79 | 15.45 |
| SOT               | 0   | 0     | 0     | 1     | 0     | 0     | 0     | 1     | 0.00  | 14.14 |
| CEOT              | 0   | 1     | 0     | 1     | 1     | 0     | 0     | 0     | 1.00  | 11.53 |
| AOT               | 0   | 5     | 4     | 0     | 1     | 2     | 0     | 0     | 2.50  | 15.54 |
| KCOT              | 1   | 14    | 16    | 16    | 8     | 13    | 3     | 3     | 35.51 | 16.84 |
| AF                | 0   | 1     | 0     | 0     | 0     | 0     | 0     | 0     | 0.00  | 11.00 |
| AFO               | 1   | 1     | 0     | 0     | 0     | 0     | 0     | 0     | 0.00  | 11.31 |
| CCOT              | 0   | 1     | 2     | 3     | 0     | 0     | 1     | 0     | 34.00 | 19.05 |
| Odontoma          | 3   | 24    | 7     | 2     | 2     | 2     | 0     | 0     | 17.55 | 10.07 |
| OM                | 0   | 1     | 0     | 0     | 1     | 0     | 0     | 0     | 35.00 | 24.04 |
| OF                | 0   | 1     | 0     | 0     | 2     | 1     | 0     | 0     | 39.00 | 18.78 |
| Cementoblastoma   | 0   | 1     | 0     | 0     | 1     | 0     | 0     | 0     | 28.00 | 19.80 |
| AC                | 0   | 0     | 0     | 0     | 0     | 1     | 0     | 0     | 52.00 | -     |
| PIOSCC            | 0   | 0     | 0     | 0     | 1     | 1     | 0     | 0     | 49.50 | 7.78  |

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| Odontogenic tumor | Maxillary anterior | Maxillary posterior | Mandibular anterior | Mandibular posterior |
|-------------------|--------------------|---------------------|--------------------|---------------------|
| Ameloblastoma     | 4                  | 10                  | 31                 | 105                 | 3                   |
| SOT               | 1                  | 1                   | 0                  | 0                   | 0                   |
| CEOT              | 0                  | 1                   | 1                  | 1                   | 0                   |
| AOT               | 7                  | 3                   | 2                  | 0                   | 0                   |
| KCOT              | 6                  | 11                  | 13                 | 44                  | 0                   |
| AF                | 0                  | 0                   | 0                  | 1                   | 0                   |
| AFO               | 0                  | 1                   | 0                  | 1                   | 0                   |
| CCOT              | 0                  | 1                   | 3                  | 4                   | 0                   |
| Odontoma          | 18                 | 8                   | 3                  | 11                  | 0                   |
| OM                | 0                  | 0                   | 1                  | 1                   | 0                   |
| OF                | 0                  | 0                   | 1                  | 3                   | 0                   |
| Cementoblastoma   | 0                  | 0                   | 0                  | 2                   | 0                   |
| AC                | 0                  | 0                   | 0                  | 1                   | 0                   |
| PIOSCC            | 0                  | 1                   | 0                  | 1                   | 0                   |

SOT: Squamous odontogenic tumour, CEOT: Calcifying epithelial odontogenic tumour, AOT: Adenomatoid odontogenic tumour, KCOT: Keratocystic odontogenic tumour, AF: Ameloblastic fibroma, AFO: Ameloblastic fibro-odontoma, CCOT: Calcifying cystic odontogenic tumour, OM: Odontogenic myxoma, OF: Odontogenic fibroma, AC: Ameloblastic carcinoma, PIOSCC: Primary intra osseous squamous cell carcinoma

| Odontogenic cyst | Frequency | Percentage | Male | Female | Male/ Female |
|-----------------|-----------|------------|------|--------|--------------|
| RC              | 655       | 75.11      | 343  | 312    | 1.09:1       |
| DC              | 175       | 17.2       | 105  | 45     | 2.33:1       |
| Residual cyst   | 44        | 5.02       | 20   | 24     | 1.2:1        |
| LPC             | 9         | 1.03       | 5    | 4      | 1.25:1       |
| Unclassified    | 14        | 1.61       | 8    | 6      | 1.33:1       |

RC: Radicular cyst, DC: Dentigerous cyst, LPC: Lateral periodontal cyst

Region (24.54%). Radicular and dentigerous cyst occurred mostly in the maxillary anterior region.
After the inclusion of odontogenic keratocyst as KCOT in 2005 WHO classification, the prevalence of odontogenic tumors has increased. In this study, KCOT accounts for 24.3% of odontogenic tumors, forming the second most common odontogenic tumor. The frequency of adenomatoid odontogenic tumor (AOT) was 3.93% which is comparable to studies from China.6,10 Epidemiological studies from other parts of India13 showed a higher frequency of 9% for AOT. Further follow-up studies on different geographical location could prove whether a racial difference is present for the occurrence of certain types of odontogenic tumor in a given population.

The frequency of odontogenic myxoma in this study was 0.66%, which is a lower value compared to previous studies. Retrospective studies from Nigeria,9 Brazil6 and Mexico14 showed a frequency of 6.5–17.7% related to odontogenic myxoma. The frequency of odontogenic myxoma in Sri Lanka15 and China6 was in the range of 2.6–4.9%. Other studies from India13 also showed a higher frequency than this study. The reason for this low incidence needs further investigation.

This study showed the mean age of odontogenic tumors as 33.7 years with peak occurrence in the third decade, similar to other studies from India,16 China10 and Nigeria.9 However, studies from Chile7 and Brazil37 showed the mean age less than a decade. This may be because odontoma constituted the most common odontogenic tumor in those populations or may be due to a racial difference in incidence.

Overall a slight male predilection (male:female = 1.19:1) of odontogenic tumors was noticed in this study. This is in concordance with studies from China,6,10 Australia18 and another study from India.16 Ameloblastoma showed a male to female ratio of 1:1.13. This result was different from studies in China,6 Nigeria9 and another study from India16 where a male predilection was noticed. However, a study from Brazil8 showed a female predilection. In this study, KCOT showed a definite male predilection which is in agreement with many other studies,6,20 but this is different from a study from Brazil.8 This possibly discloses a gender difference among different populations.

Mandible was the most common jaw affected with maxilla to mandible ratio of 1:3.2, which is similar to other studies from Asia6,13 and Africa.9 However, studies from American continent7,34,39 showed an almost equal predilection for both the jaws. This might be due to the lower frequency of ameloblastoma in that population.

In this study, 18% of ameloblastoma showed recurrence. No significant association was found between recurrence and age, gender or site. 10.7% of KCOT also showed recurrence; and no statistical association was found between recurrence and age, gender or site.
Odontogenic cysts comprised 12.25% of all oral biopsies during 1998–2012, which was 2.86 times the frequency of odontogenic tumors. This frequency of odontogenic cysts is similar to other studies in the UK[23] and Saudi Arabia.[22] The most common odontogenic cyst in the present study was radicular cyst, similar to other studies.[23‑25] Radicular cysts comprised 75.11% of odontogenic cysts, which is higher than most of the other studies.[23,24] However, a study from Sicily[25] showed a higher frequency of 84.5%. The higher frequency in our study may be due to the exclusion of odontogenic keratocyst from the category of the cyst.

Dentigerous cyst constituted 17.2% of odontogenic cysts. This result is in concordance with a systematic review by Johnson et al.[26] However, the incidence is lower than studies from Brazil[23] and Saudi Arabia.[22] This study showed a definite male predilection with a male to female ratio of 2.33:1. Daley et al.[12] suggested that this might be due to a smaller jaw size and a greater trend for prophylactic extraction of third molars in females. In the present study, dentigerous cyst showed an almost equal predilection for maxillary anterior and mandibular posterior regions. This is in contrast to other studies[23,24] showing a definite predilection for the mandibular posterior region. Esthetic concerns of the patients regarding non-eruption of anterior teeth, especially canine might have resulted in an increased reporting of dentigerous cyst in that region. Any geographical difference in the population regarding the site of occurrence is yet to be ruled out. Residual cyst constituted 5.02% of odontogenic cyst and showed a slight female predilection. These cysts showed high frequency in fifth to seventh decades and most commonly occurred in the mandibular posterior region. The remaining cysts constituted only 2.65% of cases.

Clinically, most of these cysts and tumors (60.6%) presented with a swelling, though some were asymptomatic (20.4%). Complete clinical data could not be retrieved for the remaining cases. We also recorded the radiographic features of ameloblastoma and KCOT, even though this was not included in our objectives. Sixty-nine (45.4%) cases of ameloblastoma showed a multilocular radiolucency and 24 (15.8) were unilocular. Radiographic details were not available for 59 cases. About 37.8% of KCOT showed unilocular radiolucency whereas 25.7% were multilocular. Radiographic details were not available for the remaining cases of KCOT.

The present study reviewed the epidemiological profile of odontogenic cysts and tumors in a tertiary dental health-care setting in southern Kerala. There are only three such institutions in Kerala, one each in the northern, central and southern part. Hence, people with tumor and tumor-like lesions of the jaw reporting to this institution could be taken as fairly representative sample of South Kerala population. We do not know whether there is a real population difference regarding the incidence and distribution of odontogenic cysts and tumors. However, the demographic details in the literature and the result of our review point to that direction. Further studies across different populations may be beneficial.

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