

Odontogenic Tumors: A Review of 675 Cases in Eastern Libya

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ABSTRACT

Aims: The aim of this study was to determine the relative frequency of odontogenic tumors (OTs) in an Eastern Libyan population based on the 2005 World Health Organization (WHO) classification, and also to compare the actual data with previous studies. **Materials and Methods:** We retrieved and analyzed 85 OTs from a total of 675 tumors and tumor-like lesions of the oral and perioral structures, for gender, age, tumor site, and frequency. The diagnosis was based on the most recent WHO (2005) classification of OTs. **Results:** OTs constituted 12.6% of all oral/jaw tumors and tumor-like lesions. Ameloblastoma (28.2%) was the most common type, followed by keratocystic odontogenic tumor (25.2%) and odontoma (19.9%). The male: female ratio was 1.2:1, and maxilla: mandible ratio 1:2. The mean age of occurrence of tumors was 29 years with a peak incidence between 10 and 40 years. **Conclusions:** OTs are relatively common lesion in this Libyan Population, but the incidence of tumors is neither similar to Caucasians nor Sub-Saharan population.

KEYWORDS: Cysts, odontogenic, tumors

INTRODUCTION

The relative frequency of odontogenic tumors (OTs) has been disputed due to controversies and confusion in the tumor taxonomy, subtyping and pleomorphism in its presentation.^[1-4]

In the past, many studies based on 1971 and 1992, World Health Organization (WHO) classifications studies were carried out in various parts of the world.^[2-18] To our knowledge, few studies based on latest edition (2005) of WHO classification have been reported.^[9,19,20]

In this study histopathologic reports (both odontogenic and nonodontogenic) of biopsy specimens from 1997 to 2007 were retrieved and rediagnosed based on 2005 classification.^[1]

MATERIALS AND METHODS

We retrieved histopathology reports of the biopsy specimens from the university teaching hospital, Benghazi, which is the only referral center in the entire eastern part of Libya serving

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1/3rd (1.5 million) of the entire Libyan population to analyze all oral tumors. The relative frequency of OTs in Libyan population was analyzed and compared with previous reports.

The OTs were analyzed for frequency, sex, and site distribution. With regards to site distribution, the maxilla and mandible were divided into three anatomic regions: Anterior, premolar, posterior.

The data obtained was compared with the reports from various parts of the world, based on 1992 classification

RESULTS

In the present study, 85 OTs were retrieved, which constituted 12.6% of the 675 tumors and tumor-like lesions involving the oral and maxillofacial region. Of the 85 OTs, 84 (98.8%) were benign and 1 (1.1%) was malignant.

The most common tumor found was ameloblastoma, which constituted 28.2% followed by keratocystic odontogenic tumor (KCOT) (25.8%) and odontoma (19.9%) [Table 1].

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Table 2 shows age, gender, and site distribution for all OTs. OTs were present in the age range between 6 and 75 years with a mean age of 29.4 years. The peak incidence of OTs was in the third decade (31.7%), with 74.1% of the patients in between 10 and 39 years. The male to female ratio in the present study was 1.2:1. There were 28 (32.9%) tumors in the maxilla and 57 (67%) in the mandible; 42.3% of the OTs were located in the posterior region, ameloblastoma comprising 21.1% of those, followed by KCOT (8.2%). Except ameloblastic fibroma, rest of the tumors showed a mandibular predilection.

In Table 3, the frequency of the OTs was compared with the previous studies, according to 1992 WHO classification.

DISCUSSION

The WHO published the third and updated edition of OTs in 2005.^[1] There were six major changes compared to the 1992 WHO classification: (1) Parakeratinised odontogenic keratocyst is classified as benign tumor derived from odontogenic

Table 1: Frequency of odontogenic tumors (2005 World Health Organization classification)

| | Total | Percentage |
|---|-------|------------|
| Benign (84 cases, 98.8%) | | |
| Odontogenic epithelium with mature fibrous stroma, without odontogenic ectomesenchyme | | |
| Ameloblastoma | 24 | 28.2 |
| Squamous odontogenic tumor | - | - |
| Calcifying epithelial odontogenic tumor | 1 | 1.1 |
| Adenomatoid odontogenic tumor | 2 | 2.3 |
| Keratocystic odontogenic tumor | 22 | 25.8 |
| Odontogenic epithelium with odontogenic ectomesenchyme, with or without hard tissue formation | | |
| Ameloblastic fibroma/fibro-dentinoma | 2 | 2.3 |
| Ameloblastic fibro-odontoma | - | - |
| Odontoma, complex type | 9 | 10.5 |
| Odontoma, compound type | 8 | 9.4 |
| Odontoameloblastoma | - | - |
| Calcifying cystic odontogenic | 6 | 7.0 |
| Dentinogenic ghost cell tumor | - | - |
| Mesenchyme and/or odontogenic ectomesenchyme, with or without odontogenic epithelium | | |
| Odontogenic fibroma | 1 | 1.1 |
| Odontogenic myxoma/fibromyxoma | 6 | 7.0 |
| Cementoblastoma | 3 | 3.5 |
| Malignant (1 case, 1.17%) | | |
| Odontogenic carcinoma | 1 | 1.1 |
| Primary intraosseous squamous | | |
| Cell carcinoma | - | - |
| Clear cell odontogenic carcinoma | - | - |
| Ghost cell odontogenic carcinoma | - | - |
| Total | 85 | 100 |

epithelium and termed KCOT. (2) Adenomatoid odontogenic tumor (AOT) originates from odontogenic epithelium but not from ectomesenchyme. (3) Calcifying odontogenic cyst (COC) is divided into two benign and malignant groups. (4) Clear cell odontogenic tumor (CEOT) is considered malignant lesion and termed clear cell odontogenic carcinoma (CCOC). (5) Odontogenic carcinoma is not included. (6) Changes related to terminology and typing.

The incidence of OTs in the Sub-Saharan African Population is reported to be between 19 and 32%^[4-7] and in Caucasians 1%.^[13] The relative incidence of OTs of all the tumors and tumor-like lesions in this study was 12.5%, which suggests that OTs are not uncommon in Libyan population.

In the present study, ameloblastoma was the most common tumor (28.2%), which is consistent with the Nigerian^[4-6] European,^[11] and Chinese studies^[9,10] KCOT constituted the second (25.8%) most common tumor followed by odontoma (19.9%). The frequency of KCOT concurs with the report by Jing *et al.*^[9] The frequency of Odontomas was relatively high compared to other African studies,^[4-8] but low in comparison to American reports.^[13] The frequencies of ameloblastoma and odontoma are consistent with the report by Olgac *et al.*

There was an almost equal sex predilection in this study (M:F = 1.2:1), which was similar to Nigerian studies,^[4,5] but in contrast to female preponderance from Hong Kong, Chile, Mexico, Brazil, and Estonia.^[14-17]

OTs showed a marked preference for mandible in this study (2:1) and Nigerian series^[4-7] however, American and European studies showed a predilection for maxilla.^[11-13] The marked prevalence to mandible is believed to be due to the high incidence of Ameloblastoma.

The frequency of ameloblastoma was between 11 and 24% in Caucasians^[13,14] while among black Africans, it accounts for 66–99%.^[2,6] When 1992 classification was used, ameloblastoma constituted 38% of the OTs in the present study.

The peak incidence of ameloblastomas was in the third decade, and the mean age was 29 years, which concurs with other African findings.^[4-6] The male predilection observed in this series is in line with the Nigerian study,^[7] but differs from studies from Turkey^[11] and South America,^[15] in which female predominance was reported.^[9]

The 2005 WHO classification considered parakeratinised odontogenic keratocyst as benign OT tumor derived from odontogenic epithelium due to its aggressive nature and recurrence rate, and it is now termed KCOT.^[1,18] Of the 32 OKCs retrieved in total, 22 (68.8%) were parakeratinised. The study by Jing *et al.*^[9] reported 91.1% of the odontogenic keratocysts comprised parakeratinised epithelium (KCOT). KCOT was the second (22.5%) most common tumor, with a male– female ratio 1.2:1. KCOT

Table 2: Age (years), sex and site distribution of patients with odontogenic tumors

| | Total | Male | Female | Mean age±SD | Maxilla | Mandible | Maxilla:mandible ratio |
|--------|-------|------|--------|-------------|---------|----------|------------------------|
| AME | 24 | 15 | 9 | 30.4±14 | 3 | 21 | 1:7 |
| SOT | - | - | - | - | - | - | - |
| CEOT | 1 | 1 | - | 50 | - | 1 | - |
| AOT | 2 | 2 | - | 19.0±5 | 2 | - | - |
| KCOT | 22 | 14 | 8 | 31.5±15 | 8 | 13 | 1:1.6 |
| AF/AFD | 2 | 1 | 1 | 34.0±28 | 2 | 1 | 2:1 |
| AFO | - | - | - | - | - | - | - |
| OC | 9 | 3 | 6 | 17.0±6 | 3 | 6 | 1:2 |
| OCp | 8 | 2 | 6 | 23.0±5 | 3 | 5 | 1:1.6 |
| CCOT | 6 | 4 | 2 | 28.0±7 | 3 | 3 | 1:1 |
| DGOT | - | - | - | - | - | - | - |
| OF | 1 | 1 | - | 50 | 1 | - | - |
| OM | 6 | 5 | 1 | 32.3±13 | 2 | 4 | 1:2 |
| CB | 3 | 1 | 2 | 39.3±6 | 1 | 2 | 1:2 |
| AC | 1 | 1 | - | 43 | - | 1 | - |
| PIOSCC | - | - | - | - | - | - | - |
| CCOC | - | - | - | - | - | - | - |
| GCOC | - | - | - | - | - | - | - |
| AFS | - | - | - | - | - | - | - |
| Total | 85 | 48 | 37 | 29.0±14 | 28 | 57 | 1:2 |

AME: Ameloblastoma, SOT: Squamous odontogenic tumor, CEOT: Calcifying epithelial odontogenic tumor, AOT: Adenomatoid odontogenic tumor, KCOT: Keratocystic odontogenic tumor, AF/AFD: Ameloblastic fibroma/fibrodentinoma, AFO: Ameloblastic fibro-odontoma, OC: Odontoma, complex type, OCp: Odontoma, compound type, OA: Odontoameloblastoma, CCOT: Calcifying cystic odontogenic, DGCT: Dentinogenic ghost cell tumor, OF: Odontogenic fibroma, OM: Odontogenic myxoma/fibromyxoma, CB: Cementoblastoma, AC: Ameloblastic carcinoma, PIOSCC: Primary intraosseous squamous cell carcinoma, CCOC: Clear cell odontogenic carcinoma, GCOC: Ghost cell odontogenic carcinoma, SD: Standard deviation

Table 3: Percentage incidence of odontogenic tumors in various studies, based on 1992 World Health Organization classification

| | Present study | Europe | Africa | | Americas | | Asia | |
|------------|---------------|------------------------|------------------------|-------------------------|------------------------|------------------------|-----------------------|--------------------|
| | | Turkey ^[11] | Nigeria ^[7] | Tanzania ^[9] | Brazil ^[15] | Mexico ^[14] | China ^[10] | HK ^[17] |
| AME | 38 | 25.2 | 58.5 | 80.1 | 45.3 | 23.7 | 58.6 | 62 |
| SOT | - | 2.1 | - | - | 1.5 | - | 0.4 | - |
| CEOT | 1.5 | 0.9 | 0.4 | 1.7 | 1.2 | 0.8 | 0.3 | - |
| AOT | 3.1 | 2.1 | 6.2 | 0.9 | 3.8 | 7.1 | 8.3 | 7 |
| KCOT | - | - | - | - | - | - | - | - |
| AF/AFD | 3.1 | 1.5 | 4.5 | 1.7 | 1.8 | 1.4 | 1.8 | - |
| AFO | - | - | - | - | 0.3 | 0.8 | 0.3 | 1 |
| OC and OCp | 26.9 | 20.7 | 4.2 | 2.6 | 25.0 | 34.6 | 6.7 | 6 |
| OA | - | - | - | - | 1.8 | - | 0.2 | - |
| COC | 9.5 | 5.5 | 2.4 | 1.7 | 3.5 | 6.8 | 4.6 | 2 |
| OF | 1.5 | 9.9 | 4.5 | 1.7 | 3.2 | 4.5 | 0.7 | - |
| OM | 9.5 | 15.7 | 11.8 | 7.0 | 9.1 | 17.7 | 0.7 | 1 |
| CB | 4.7 | 1.9 | 0.7 | 1.7 | 2.3 | 0.8 | 2.6 | - |
| Malignant | 1.5 | 1.1 | 5.2 | - | 0.3 | 1.1 | 4.8 | - |
| Cases | 63 | 527 | 289 | 116 | 340 | 349 | 759 | 82 |

KCOT was not included as the comparison is based on 1992 WHO classification. AME: Ameloblastoma, SOT: Squamous odontogenic tumor, CEOT: Calcifying epithelial odontogenic tumor, AOT: Adenomatoid odontogenic tumor, KCOT: Keratocystic odontogenic tumor, AF/AFD: Ameloblastic fibroma/fibrodentinoma, AFO: Ameloblastic fibro-odontoma, OC: Odontoma, complex type, OCp: Odontoma, compound type, OA: Odontoameloblastoma, OF: Odontogenic fibroma, OM: Odontogenic myxoma/fibromyxoma, CB: Cementoblastoma, COC: Cell odontogenic carcinoma, WHO: World Health Organization

showed a predilection for mandible with a maxilla-mandible ratio of 1:1.6. The results were consistent with the Chinese study,^[9] which was also based on the latest classification.

The mean age of compound odontoma was 23.3 years and that of complex odontoma 17.7 years which are not in agreement with other reports, where the mean age of compound odontoma was lesser than complex odontoma.^[9,11,12,15]

Two cases of AOT were reported, both in the maxilla and before the age of 30. The occurrence of tumor in a younger age group was also shown in other reports.^[4,5,8,12,15,16]

In 2005, calcifying odontogenic cyst was reclassified into categories: Calcifying cystic odontogenic tumor (CCOT), a benign cystic neoplasm characterized by an ameloblastoma-like epithelium with ghost cells that may calcify. Dentinogenic ghost

cells tumor (DGCT) histologically constituting ameloblastic epithelium with mature connective tissue and ghost cells, in association with dysplastic dentin; and ghost cell carcinoma which is a malignant variant of CEOT and DGCT. Clear cell OT is now termed clear CCOC.

In this study, there were six cases of CCOT with a mean age of 28 years and the maxilla–mandible ratio was equal. The male preponderance of the tumor was similar to series by Jing *et al.*^[9]

Odontogenic myxoma was the second most common tumor in the sub-Saharan population.^[4,6,8] In our study, odontogenic myxoma comprised 7% of all tumors and were similar to the frequency seen in previous reports.^[5,8,9,15] The tumor shows a predilection for mandible similar to the reports by Nigerian series,^[5,7] but other reports have recorded predilection for maxilla.^[11,14,16] Although most reports suggest a female predilection of the tumor, it is interesting to note that in our study odontogenic myxoma is seen predominantly in males (5:1).

Ameloblastic fibroma constituted 2.3% of all OTs, which was consistent with reports from other series,^[4-9,11,13-15] but not with that of Estonian study^[2] (16%).

Other tumors seen in this study with low frequency are CEOT, cementoblastoma, and central odontogenic fibroma.

The incidence of malignant OTs was significantly high in Sub-Saharan African series^[5-7] and Chinese studies.^[9,10] Our study reported one case of malignant tumor (1.1%), and when 1992 classification was used for comparison, the frequency was 1.5%. The rarity of the malignant tumors was in agreement with American^[13-16] and European series (0.1–1.1%).^[11]

The incidence of OTs in this Libyan population is similar to studies from Turkey^[11] and Brazil,^[15] rather than aligning either to Sub-Saharan or Caucasian populations. Due to an altered pattern of incidence in select populations, further studies are encouraged to ascertain the similarities in the genetic pattern of different geographic locations and racial populations.

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Conflicts of interest

There are no conflicts of interest.

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