

## Odontogenic tumors in children and adolescents. A 15-year retrospective study in Argentina

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Received: 30-10-2006

Accepted: 25-03-2007

Guerrisi M, Piloni MJ, Keszler A. Odontogenic tumors in children and adolescents. A 15-year retrospective study in Argentina. Med Oral Patol Oral Cir Bucal 2007;12:E180-5.

© Medicina Oral S. L. C.I.F. B 96689336 - ISSN 1698-6946

**Indexed in:**

-Index Medicus / MEDLINE / PubMed  
-EMBASE, Excerpta Medica  
-SCOPUS  
-Índice Médico Español  
-IBECS

### ABSTRACT

There are few studies on bucco-maxillary lesions in children and adolescents, and reports on odontogenic tumors (OT) are scanty. Comparison among the available data is difficult due to differences in the criteria used in each study. The aim of the present work was to perform a retrospective study of OT in patients aged up to 20 years diagnosed at a center specialized in histopathologic diagnosis of oral diseases. All cases diagnosed with OT between 1990 and 2004 were retrieved from the Service archives. The 153 retrieved cases accounted for 7% of total biopsies performed during that period, and to 78.4% of tumors of the jaws. Mean age of the whole population was 12.7 years and the male to female ratio was 2:1. The most frequent tumor types were Odontoma, (50.9%), Ameloblastoma (18.3%) and Myxoma (8.5%), and the most infrequent was Calcifying Epithelial Odontogenic Tumor (1.3%). According to our results, OT cannot be considered infrequent. They are the prevalent tumors of the jaws in the age group studied herein.

**Key words:** *Odontogenic tumors, odontopediatric pathology, maxillary pathology.*

### RESUMEN

Los estudios sobre lesiones buco maxilares en niños y adolescentes son relativamente poco comunes y escasos los reportes sobre tumores odontogénicos (TO) en la literatura internacional actualizada. Los datos disponibles son difíciles de comparar debido a los diferentes criterios de análisis utilizados. El objetivo del presente trabajo fue realizar un estudio retrospectivo de TO en pacientes de hasta 20 años de edad diagnosticados en un centro especializado en diagnóstico anatomopatológico bucal. Se seleccionaron de los archivos del Servicio los casos con diagnósticos de TO correspondientes al período 1990-2004. Los 153 casos hallados representaron el 7% del total de biopsias realizadas y el 78.4 % de los tumores maxilares. La edad media general fue de 12.7 años y la relación hombre-mujer 2:1. Los tipos tumorales más frecuentes fueron el Odontoma (50.9%), Ameloblastoma (18.3%) y Mixoma (8.5%). El más infrecuente fue el Tumor Odontogénico Epitelial Calcificante (1.3%). Según los datos obtenidos los TO no pueden ser considerados infrecuentes. En los maxilares son los prevalentes en el grupo etario estudiado.

**Palabras clave:** *Tumores odontogénicos, patología odontopediátrica, patología maxilares.*

## INTRODUCTION

Buccomaxillary neoplasms affecting children and adolescents are most frequently benign (1, 2). Those occurring in the maxillae include bone tumors and OT, which only present in the jaws. OT comprise a large heterogeneous group of lesions originating from the epithelium and/or odontogenic ectomesenchyme and remnants (3). OT include entities of a hamartomatous nature, such as odontoma, benign neoplasms, some of which are aggressive as is the case of ameloblastoma and myxoma, and malign neoplasms capable of metastasis (4-6). Odontogenic keratocysts have recently been included in the WHO classification of OT as keratocystic odontogenic tumors on account of their clinical behavior and genetic and molecular features (7). There are relatively few studies on buccal and maxillofacial lesions in children and adolescents, and reports on OT are scanty. Data reported in the literature on the frequency of OT differ greatly. According to some authors, OT account for merely 1% of oral lesions, whereas to other authors they account for 28% (8). Establishing comparisons among the reported data is difficult due to the differences in the classification and criteria applied to diagnose and group the lesions, the upper age limit, and racial-ethnic origin of the population (9-13). Whereas some studies comprise the full range of oral and maxillofacial diseases (1, 2, 9, 10), others only focus on oral and maxillofacial tumors (12-15). There are also studies that include tumor-like lesions (8,16), and/or cysts (17), and others that evaluate odontogenic tumors (11, 18, 19). In addition, there are reports that address a specific type of odontogenic neoplasm (20, 21). Reports also vary greatly in their findings regarding the predominant OT in this age group. Asamoah et al (22) showed adenomatoid odontogenic tumors to be the most frequent, whereas Adebayo et al (11) and Arotiba et al (18) on one hand, and Ulmansky et al (8) on the other, found ameloblastoma and myxoma to be prevalent respectively.

The aim of the present study was to perform a retrospective study of OT in child and adolescent patients diagnosed at a service specialized in oral histopathologic diagnosis, and to compare results with data previously published in the literature in order to detect potential population differences.

## MATERIALS AND METHODS

The study material was obtained from the archives of the Laboratory of Surgical Pathology of the School of Dentistry, University of Buenos Aires. All cases of odontogenic tumors in patients aged up to 20 years diagnosed between 1990 and 2004 were retrieved; the histopathologic diagnoses were in keeping with the WHO classification of tumors. Odontogenic keratocysts (keratocystic odontogenic tumor) were not included in the study in order to render results comparable with those reported in the literature. Clinical data regarding age, gender, and tumor location, were retrieved from the biopsy protocols. The frequency of each tumor type was established. Mean age, distribution according to gender, and the prevalent location of each tumor, were determined. In order to establish tumor distribution according to the

decade of life, the cases were divided into the following age groups: 0 to 10, and 11 to 20 years of age.

## RESULTS

The 153 cases of OT accounted for 7% of total diagnosed cases corresponding to children and adolescents (n= 2250) throughout the studied 15-year period. Considering lesions located specifically in the jaws (n: 1228), frequency of OT reached 12%, accounting for 78.4% of exclusively tumor lesions (n: 195) (Figure 1).

### - Types of lesion

Results obtained in this series showed that 50.9% of OT were odontoma; 18.3% were ameloblastoma, and 8.5% were myxoma. Adenomatoid odontogenic tumor and ameloblastic fibro odontoma showed a similar frequency, reaching 5.2 and 4.6% respectively. Calcifying cystic odontogenic tumor (formerly termed calcifying odontogenic cyst) accounted for 3.9% of cases and odontogenic fibromas for 3.2%. The least frequent tumors were cementoblastoma and ameloblastic fibroma, each of which accounted for 1.9% of tumors, and calcifying epithelial odontogenic tumor, which accounted for 1.3 % (Table 1).

### - Localization

Ameloblastoma, myxoma, ameloblastic fibro odontoma, cementoblastoma, ameloblastic fibroma, and calcifying epithelial odontogenic tumor were most frequently located in the posterior sector of the mandible. Odontoma and adenomatoid odontogenic tumor were most frequently located in the anterior sector of the maxilla, whereas calcifying cystic odontogenic tumor was most frequently encountered in the posterior sector of the maxilla. Distribution of odontogenic fibroma in both maxillae was similar (Table 1).

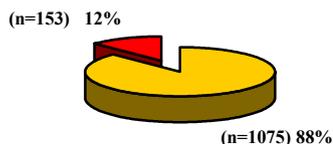
### - Gender and Age

Odontoma, myxoma, adenomatoid odontogenic tumor, calcifying cystic odontogenic tumor, and calcifying epithelial odontogenic tumor, showed equal or similar distribution according to gender. Ameloblastoma, cementoblastoma, odontogenic fibroma and ameloblastic fibro odontoma were between 2 and 6 fold more frequent in males, whereas ameloblastic fibroma was more frequent in females. The mean age of the whole population was 12.7 years (3-20 years). Considering tumor type, the lowest and highest mean ages corresponded to ameloblastic fibrous odontoma (mean: 11 years) and ameloblastoma (mean: 17.7 years) respectively. All tumor types were more frequent in the second decade of life, and some types, such as cementoblastoma and calcifying epithelial odontogenic tumor, only presented in the second half of this decade (Table 1).

## DISCUSSION

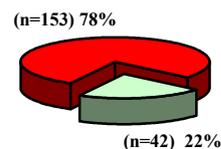
Bone tumors involving other skeletal sites are rarely encountered in the maxillae. OT are considered highly infrequent since they account for a very low percentage of the diseases that affect both hard and soft tissues (1, 2, 5). However, their frequency is relevant when considering tumors of the jaws (9, 12, 13).

**Frequency of OT considering general pathology of the jaws**



■ Other pathologies of the jaws ■ Odontogenic Tumors

**Frequency of OT considering tumors of the jaws**



■ Non-odontogenic Tumors ■ Odontogenic Tumors

**Fig. 1.** Frequency of Odontogenic Tumors within general pathology and tumors of the jaws.

**Table 1.** Histologic types. Relative incidence, distribution according to gender, age, and localization of odontogenic tumors in the present series of cases.

| Tumor Type                         | Cases      |            | Gender<br>M:F | Age (years) |            | Prevalent<br>Localization |
|------------------------------------|------------|------------|---------------|-------------|------------|---------------------------|
|                                    | n          | %          |               | 0-10        | 11-20      |                           |
| Odontoma                           | 78         | 50.9       | 1:1.2         | 15          | 63         | Maxilla. AS               |
| Ameloblastoma                      | 28         | 18.3       | 2:1           | 3           | 25         | Mandible. PS              |
| Myxoma                             | 13         | 8.5        | 1:1.4         | 1           | 12         | Mandible. PS              |
| Adenomatoid odontogenic tumor      | 8          | 5.2        | 1:1           | 0           | 8          | Maxilla. AS               |
| Ameloblastic fibro-odontoma        | 7          | 4.6        | 6:1           | 3           | 4          | Mandible. PS              |
| Calcifying cystic odont. tumor     | 6          | 3.9        | 1.5:1         | 1           | 5          | Maxilla. PS               |
| Odontogenic fibroma                | 5          | 3.2        | 4:1           | 0           | 5          | Mandible ~ Maxilla        |
| Cementoblastoma                    | 3          | 1.9        | 2:1           | 0           | 3          | Mandible. PS              |
| Ameloblastic fibroma               | 3          | 1.9        | 1:2           | 0           | 3          | Mandible. PS              |
| Calcifying epithelial odont. tumor | 2          | 1.3        | 1:1           | 0           | 2          | Mandible. PS              |
| <b>TOTAL</b>                       | <b>153</b> | <b>100</b> |               | <b>23</b>   | <b>130</b> |                           |

AS: Anterior sector  
PS: Posterior sector

**Table 2.** Frequency of OT in total biopsies and buccomaxillary tumors in different series.

| Author                | Year | Population     | Age  | Odontogenic Tumors | % Total Biopsies | % Total Tumors | % Tumors of the Jaws |
|-----------------------|------|----------------|------|--------------------|------------------|----------------|----------------------|
| Sato et al (12)       | 1997 | Japan          | 0-15 | 79                 | N/D              | 31.6           | 79.8                 |
| Chen et al (9)        | 1998 | Taiwan         | 0-15 | 71                 | 13.7             | 42.7 (*)       | 83.5                 |
| Ulmansky et al (8)    | 1999 | Israel         | 0-15 | 18 (**)            | 1.8              | 15.9           | N/D                  |
| Tanaka et al (13)     | 1999 | Japan          | 0-15 | 28                 | N/D              | 26.6           | 84.8                 |
| Adebayo et al (11)    | 2002 | Nigeria        | 0-20 | 78                 | N/D              | 30.9 (*)       | N/D                  |
| Al-Khateeb et al (16) | 2003 | Jordan         | 0-19 | 25                 | N/D              | 9.7            | 58.1                 |
| Ajayi et al (19)      | 2004 | Nigeria        | 0-19 | 92                 | N/D              | 19.3           | N/D                  |
| Tanrikulu et al (15)  | 2004 | Turkey         | 0-15 | 17                 | N/D              | 18.8           | 21.5                 |
| Jones et al (2)       | 2006 | United Kingdom | 0-16 | 43 (**)            | 0.9              | 22.8           | N/D                  |
| Present series        |      | Argentina      | 0-20 | 153                | 6.8              | 56.8           | 78.4                 |

(\*) Includes tumor-like lesions  
 (\*\*) Does not include odontoma  
 N/D data not found

**Table 3.** Most prevalent odontogenic tumors. Comparative Data.

| Author                | % Odontoma | % Ameloblastoma | % Myxoma |
|-----------------------|------------|-----------------|----------|
| Sato et al (12)       | 59.4       | 34.1            | 1.0      |
| Chen et al (9)        | 56.3       | 29.6            | 2.8      |
| Ulmansky et al (8)    | N/D        | 11.1            | 38.8     |
| Tanaka et al (13)     | 50.0       | 39.2            | N/D      |
| Adebayo et al (11)    | 7.7        | 53.8            | 19.2     |
| Al Khateeb et al (16) | 56.0       | 24.0            | N/D      |
| Ajayi et al (19)      | 4.3        | 48.9            | 8.7      |
| Tanrikulu et al (15)  | 47.0       | 11.7            | N/D      |
| Jones et al (2)       | 73.0       | 4.9             | 2.7      |
| Present series        | 50.9       | 18.3            | 8.5      |

The vast majority of OT are benign, although some follow an aggressive clinical course due to their recurrence and to the bone destruction caused by their substantial growth. Malign OT are rare and are usually highly aggressive (1, 6, 19).

Reports vary as to the frequency of OT, partly because of differences in the parameters used by the authors. Some authors consider these tumors to be infrequent in children and adolescents, but base their estimate on the full range of diseases known to affect this population, or on oral and facial tumors and tumor-like lesions (2, 8-10). Adebayo et al (11) attribute the high percentage of OT they observed in patients aged up to 18 years, and which accounted for 31% of buccomaxillary tumors and tumor like lesions, to the ethnic characteristics of the study population. However, Sato et al (12) and Tanaka et al (13), observed a similar frequency in their respective series of Japanese patients when applying the same criteria to analyze their results. In our series of Caucasian patients, 78.4% of tumors located in the jaws were OT. Our results are in agreement with those reported by Chen et al (9), and Sato et al (12), but substantially higher than findings reported by Ulmansky et al (8) (Table 2). Reports also vary as regards prevalence and frequency of each type of OT. Our results regarding the prevalence of odontoma are in keeping with those reported by several authors (9, 12, 13, 16, 23), showing frequency to range between 50 and 59.4%.

According to Jones et al (2), however, odontoma accounts for 73%. Conversely, Adebayo et al (11) and Ajayi et al (19) found odontoma to account for 7.7% and 4.3% of OT respectively. This marked difference could be attributed to the fact that this lesion is usually asymptomatic and therefore remains undetected. The number of cases of odontoma reported in the present work is substantially higher than that observed in the Argentine population in an earlier period (1). This may be due to the currently widespread use of orthopantomography in dental treatments, which has allowed detection and enucleation of these tumors. Excluding odontoma, considered a hamartomatous rather than a neoplastic lesion, ameloblastoma is the most prevalent tumor type according to most publications. Several authors (9, 11-13, 16) have found frequency of ameloblastoma within total cases of OT to range between 24 and 53.8%, which is markedly higher than the 18.3% frequency observed in our series of cases. Jones et al (2), Tanrikulu et al (15), and Ulmansky et al (8) observed significantly lower percentages, which ranged between 4.9 and 11%, and found this tumor type to be the second most frequent. It is noteworthy that Ulmansky et al (8) found myxoma to be the most prevalent tumor type, reaching 38.8%. Their result is considerably higher than the 8.5% prevalence observed in our series, and also differs from reports by Sato et al (12) and Abedayo et al (11) who found myxoma to account for 1.3 and 19.2% respectively. (Table 3).

The remaining OT are rare, although some predominantly affect young patients, as is the case of adenomatoid odontogenic tumor, ameloblastic fibro odontoma, and ameloblastic fibroma. Few or no cases of these tumors were encountered in the series of cases analyzed in this study, except for the series evaluated by Asamoá et al (22) in which adenomatoid odontogenic tumor was the most frequent. This tumor accounted for 5.2% of OT in our series of cases, in agreement with findings reported by Jones et al (2). However, it must be pointed out that according to the latter authors adenomatoid odontogenic tumor is the most frequent true odontogenic neoplasm, and ameloblastoma is the second most frequent. The radiographic appearance of the intra-osseous follicular variant of adenomatoid odontogenic tumor may resemble a dentigerous cyst, which is a frequently encountered lesion in the age group evaluated in this study. It is therefore essential to examine imaging studies thoroughly when establishing presumptive differential diagnosis. Detailed observation of radiographic images allows detecting the extent of the radiolucent unilocular image beyond the cemento-enamel junction, and sometimes allows detecting the presence of small radiopaque foci which evidence the mineralized areas that these tumors frequently present.

## CONCLUSIONS

Tumors presenting in the jaws are predominantly benign and OT are the most frequent. The differences among reports regarding frequency cannot always be attributed to population or ethnic differences, since, as shown in our comparison among the reported series, the authors applied

different criteria to analyze their data. Variations among reports regarding frequency of the different tumor types must also be evaluated. The scant number of odontoma cases reported by some authors (11) may be due to the asymptomatic nature of this lesion, which is frequently an incidental finding in radiographic studies performed due to other presenting complaints. Conversely, the high frequency of myxoma in one of the series (8) compared herein suggests that cases of hyperplastic pericoronal follicles may have been included as myxoma, since their histologic features are misleading. OT cannot be considered infrequent, as shown by the results presented in this study. Radiographic images are not usually pathognomonic for each tumor type, and some frequently appear as osteolytic lesions associated to retained teeth, resembling cystic lesions. This must be taken into account when establishing presumptive clinical diagnosis. The data obtained in this study can be considered representative of our population of children and adolescents, given that our series comprises cases referred from different parts of the country for histopathologic diagnosis at our center, and may serve to establish presumptive diagnosis prior to biopsy. The variation in frequency observed among series reported in the literature does not seem to be due to ethnic or racial differences, but rather to the criteria applied in each study and to the lack of resources to detect unsuspected asymptomatic lesions.

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