



Clinicopathologic features of ameloblastoma in southern Brazil: A 55-year single-center analysis

Ana Luisa Homem de Carvalho^a, Manoela Domingues Martins^b, Francinne Miranda da Rosa^b, Annemarie Saudades^b, Manoel Sant'Ana Filho^b

ABSTRACT

Objective: Ameloblastoma is a benign odontogenic tumor (OT) originating from the proliferation of odontogenic epithelium without involvement of the mesenchymal tissue. The aims of the present study were to perform a retrospective analysis of ameloblastoma, compare the findings with those described in previous studies and outline the epidemiological profile of this type of tumor in southern Brazilian population.

Methods: Cases of ameloblastoma were retrieved between 1954 and 2009 from a single center and analyzed for age, gender, ethnic, primary site and histological type. Cases were classified according to the 2005 WHO classification of OTs.

Results: Descriptive statistical analysis of the data was performed, with the determination of the frequency of the categorical variables. Eighty-nine cases of ameloblastoma were found: 78 cases of solid tumor (88.6%), two cases of desmoplastic tumor (2.2%) and nine cases of the unicystic tumor (10.2%). There was a predominance of the female gender (58.4%) and Caucasian ethnicity (62.9%). Most ameloblastomas were located in the mandible (78.6%) and 40.2% occurred prior to the third decade of life.

Conclusion: The present findings are in agreement with those reported in previous studies carried out in other states of Brazil and support that notion that there may be a geographical influence on the profile of patients affected by ameloblastoma.

Key words: Odontogenic tumors; Ameloblastoma; Epidemiology

^a Oral Pathology, School of Dentistry, Pontifical Catholic University of Rio Grande do Sul (PUCRS), Porto Alegre, Brazil

^b Oral Pathology Department, School of Dentistry, Universidade Federal do Rio Grande do Sul, Porto Alegre, Brazil

Aspectos clínico-patológicos de ameloblastomas no sul do Brasil: 55 anos de uma única Instituição

RESUMO

Introdução: Ameloblastoma é uma neoplasia odontogênica benigna (NOB) que se origina da proliferação do epitélio odontogênico sem envolvimento do mesênquima. Os objetivos do presente estudo foram realizar uma análise retrospectiva do ameloblastoma, comparar os resultados com os descritos em estudos anteriores e traçar o perfil epidemiológico deste tipo de tumor na população do sul do Brasil.

Metodologia: Casos de ameloblastoma diagnosticados entre 1954 e 2009 em um único centro foram analisados quanto à idade, sexo, etnia, localização e tipo histológico. Os casos foram classificados de acordo com a classificação da OMS de 2005 de NOBs.

Resultados: Foi realizada análise estatística descritiva dos dados com a determinação da frequência das variáveis categóricas. Oitenta e nove casos de ameloblastomas foram diagnosticados: 78 casos corresponderam ao tipo sólido (88.6%), dois casos ao tipo desmoplásico (2.2%) e nove casos ao tipo unicístico (10.2%). Houve predomínio pelo gênero feminino (58.4%) e de etnia branca (62.9%). A maioria localizou-se na mandíbula (78.6%) e 40.2% ocorreram preferencialmente na terceira década de vida.

Conclusão: Nossos achados estão de acordo com os achados realizados em outros estados do Brasil. Isto mostra que pode ocorrer uma influência geográfica sobre o perfil dos pacientes acometidos por ameloblastoma.

Palavras-chave: Tumores odontogênicos; Ameloblastoma; Epidemiologia

Correspondence:
Manoel Sant'Ana Filho
manoel@ufrgs.br

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INTRODUCTION

According to the World Health Organization [1], ameloblastoma is a benign odontogenic tumor originating from the proliferation of odontogenic epithelium without involvement of the mesenchymal tissue. It accounts for 1% of all tumors of the maxilla as well as 11% of all odontogenic tumors. The slow, painless growth of this tumor often results in a late diagnosis [2]. Although benign, ameloblastoma is locally invasive and therefore surgery with a margin of safety is the treatment of choice, which results in considerable mutilation in many cases [3,4]. However, the mechanism for this invasive behavior remains unknown [1,5,6].

From the clinical-radiological standpoint, ameloblastoma is classified as solid or multicystic (70 to 80% of cases), unicystic (13%), peripheral or extrasosseous (2 to 10%) [1]. Clinically, it is most commonly found in the posterior region of the mandible (70% of cases) [1,4,7,8]. Histologically, ameloblastoma is classified as solid, desmoplastic or unicystic [1]. The solid types are subdivided into follicular, plexiform (which are the most common), acanthomatous, granular cell or basal cell [1]. More than one histological types can be found in the same tumor [1,9]. However, there is little evidence that these histological types have any significance regarding clinical behavior and prognosis. The unicystic variant generally affects younger patients (second decade of life). This form is considered less invasive and consequently associated to more conservative treatment, a more favorable prognosis and a lesser incidence of recurrence [4,7,8].

There is no consensus in the literature on the most appropriate form of treatment for the different types of ameloblastoma. Conservative treatment consists of enucleation and curettage, followed by cyrotherapy, whereas more radical treatment involves marginal resection of the affected segment or mandibulectomy/maxillectomy. Due to its less aggressive behavior, conservative treatment is recommended for a unicystic ameloblastoma [2].

Studies have been published on the profile of patients with ameloblastoma and it is reported that geographic variations may exert an influence on epidemiological data regarding this tumor in different populations [5,10-18] (Table 1). The aims of the present study were to perform a retrospective analysis of cases of ameloblastoma, compare the findings with those described in previous studies and outline the epidemiological profile of this type of tumor in the Brazilian population.

METHODS

The present retrospective study received approval from the Human Research Ethics Committee of the Universidade Federal do Rio Grande do Sul (UFRGS, Brazil).

All records of histopathological findings of the Histopathology Laboratory of the UFRGS School of Dentistry (southern Brazil) between 1954 and 2009 were reviewed for the identification of cases with a diagnosis of ameloblastoma. Slides stained with hematoxylin & eosin were examined under an optical microscope for the confirmation of the diagnosis and the findings were

Table 1. Summary of papers with analysis of clinical-demographic data on ameloblastoma.

Authors/year of publication	Study location	Sample size	Period analyzed	Gender	Anatomic site	Ethnic group	Age
Chidzonga et al., 1996	Zimbabwe	117 cases	1982 to 1991	Men (53%)	Mandible (95.7%)	–	Mean age: 30 years
Mosqueda-Taylor et al., 1997	Mexico	80 cases	1960 to 1996	Women (55.4%)	Mandible (82.4%)	–	11 to 79 years
Junquera et al., 2003	Spain	22 cases	1975 to 2000	Women (59%)	Mandible (100%)	–	22 to 72 years
Fernandes et al., 2005	Brazil	154 cases	1954 to 2004	Women (54.5%)	Mandible (85%)	–	Mean age: 27.7 years
Ledesma-Montes et al., 2007	Mexico and Guatemala	163 cases	1959 to 2000	Men (52.8%)	Mandible (86.4%)	–	Mean age: 31.7 years
Adeline et al., 2008	Kenya	184 cases	1995 to 2005	Women (55.4%)	Mandible (93.5%)	–	Mean age: 30.2 years
Tawfik et al., 2010	Egypt	34 cases	1993 to 2008	Men (67.65%)	Mandible (94.4%)	–	Mean age: 34.5 years
Fregnani et al., 2010	Brazil	121 cases	1953 to 2003	Women (53%)	Mandible (80%)	Caucasian (72%)	18 to 32 years (41.3%)
Fulco et al., 2010	Brazil	54 cases	Not informed	Women (50%) Men (50%)	Mandible (98.1%)	–	Mean age: 38.3 years
Krishnapillai, Angadi, 2010	India	73 cases	1989 to 2007	Men (56.1%)	Mandible (91.8%)	–	10 to 78 years
Present study	Brazil	89 cases	1954 to 2009	Women (58.4%)	Mandible (78.6%)	Caucasian (62.9%)	10 to 29 years (40.2%)

classified based on the criteria defined by the WHO [1] as solid ameloblastoma (subtypes: follicular, plexiform, acanthomatous, granular cell and basal cell), desmoplastic ameloblastoma or unicystic ameloblastoma. The following variables were also analyzed: gender, age, ethnic background, anatomic site and histological type. Among the 96 cases with a histopathological diagnosis of ameloblastoma, 89 contained information on these variables and were included in the present study.

A database was built using the Excel® program, followed by statistical analysis using the Statistical Package for Social Sciences® (version 13.0). The level of significance was set to 5%. Categorical variables were tabulated as follows: gender (male and female), ethnic background (Caucasian, African descent, other ethnicity and not informed), anatomic site (mandible, maxilla and not informed) and histological type (follicular, plexiform, acanthomatous, granular cell and basal cell). The quantitative variable age was categorized in decades of life to facilitate the analysis and comprehension of the data. Descriptive statistical analysis was performed for the determination of the frequencies of the categorical variables studied.

RESULTS

A total of 32,035 tumors were diagnosed at the UFRGS Oral Histopathology Laboratory between 1954 and 2009, 89

of which were cases of ameloblastoma, accounting for 0.27% of the diagnoses. Descriptive analysis was performed based on the histopathological diagnosis, patient characteristics and anatomical location (Table 2). Among the 89 cases of ameloblastoma, 78 were solid tumors (88.6%), two were desmoplastic tumors (2.2%) and nine were unicystic tumors (10.2%).

Among the solid tumors, there was a predominance of the female gender (56.0%) and Caucasian ethnicity (66.7%); most were located in the mandible (76.9%) and 40.2% occurred prior to the third decade of life. Regarding the histological findings, the follicular form was the most prevalent (34 cases; 43.68%), followed by the plexiform (29 cases; 37.2%), acanthomatous (8 cases; 10.3%) and basal cell (5 cases; 6.3%) types. No cases of granular cell ameloblastoma were diagnosed. Two cases were classified as “not identified” (2.6%) and corresponded to cases with more than one histological pattern on the same slide. The cases of desmoplastic ameloblastoma occurred in the mandible of Caucasian women in the 5th decade of life (one of these cases had no information on the ethnic background or age of the patient). Unicystic ameloblastoma was more prevalent in the female gender (66.7%), affected individuals of Caucasian and African descent equally (33.3%), was located predominantly in the mandible (88.9%) and occurred mostly between the third and sixth decades of life (77.7%) (Table 2).

Table 2. Distribution of ameloblastomas according to gender, ethnic background, anatomic site and age group; Porto Alegre, Rio Grande do Sul, Brazil, 2010.

Variable		Solid ameloblastoma (N=78) 88.6%	Desmoplastic ameloblastoma (n=02) 2.2%	Unicystic ameloblastoma (N=09) 10.2%	Total (N=89) 100%
Gender	Male	31 (39.7%)	0 (0%)	03 (33.3%)	34 (38.2%)
	Female	44 (56.4%)	02 (100%)	06 (66.7%)	52 (58.4%)
	Not informed	03 (3.8%)	0 (%)	0 (0%)	03 (3.4%)
Age	10 to 19 years	10 (12.8%)	0 (0%)	03 (33.3%)	13 (14.6%)
	20 to 29 years	22 (28.2%)	0 (0%)	01 (11.1%)	23 (25.6%)
	30 to 39 years	15 (19.2%)	0 (0%)	0 (0%)	15 (16.8%)
	40 to 49 years	10 (12.8%)	01 (50%)	0 (0%)	11 (12.3%)
	50 to 59 years	02 (2.6%)	0 (0%)	03 (33.3%)	05 (5.6%)
	60 to 69 years	03 (3.8%)	0 (0%)	0 (0%)	03 (3.8%)
	70 to 79 years	05 (6.4%)	0 (0%)	0 (0%)	05 (5.6%)
	80 to 89 years	01 (1.3%)	0 (0%)	0 (0%)	01 (1.1%)
	Not informed	10 (12.8%)	01 (50%)	2 (22.2%)	13 (14.6%)
Ethnicity	Caucasian	52 (66.7%)	01 (50%)	03 (33.3%)	56 (62.9%)
	African descent	10 (12.8%)	0 (0%)	03 (33.3%)	13 (14.6%)
	Other ethnicity	03 (3.8%)	0 (0%)	01 (11.1%)	04 (4.5%)
	Not informed	13 (16.7%)	01 (50%)	02 (22.2%)	16 (18.0%)
Site	Mandible	60 (76.9%)	02 (100%)	08 (88.9%)	70 (78.6%)
	Maxilla	07 (9%)	0 (0%)	0 (0%)	07 (7.8%)
	Extraosseous	02 (2.6%)	0 (0%)	0 (0%)	02 (2.2%)
	Not informed	09 (11.6%)	0 (0%)	01 (11.1%)	10 (11.23%)

DISCUSSION

The incidence of odontogenic tumors exhibits considerable variation in epidemiological studies carried out in different countries [5,10-18]. Such variations may be explained by geographic, ethnic and socioeconomic factors [16] and the large number of reactive/inflammatory tumors submitted to histopathological analysis in North American countries, which does not occur in Latin American countries [11].

Ameloblastoma is the most prevalent odontogenic tumor in studies carried out in Brazil (45.2% to 67% of cases) [12,16], China (58.6 to 59.4%) [20,21], Nigeria (58.5%) [22] and Egypt (41.5%) [14]. However, a number of investigations report ameloblastoma to be the second most prevalent form of odontogenic tumor, such as studies carried out in Canada (17.8% of cases) [22], Mexico (23.7%) [10] and Brazil [11]. This difference may be explained by the fact that some studies do not include odontoma, which increases the relative frequency of ameloblastoma [16].

In the present study, ameloblastoma accounted for 0.27% of all tumors analyzed at the UFRGS Oral Histopathology Laboratory between 1954 and 2009. This frequency is smaller than rates reported in a Latin American population [13] and a previous Brazilian investigation [12], which describe a frequency of 0.8% and 0.5%, respectively. It should be stressed that these greater frequencies were due to the fact that the studies cited used both laboratory findings and data from hospitals that received patients sent for treatment with a previous diagnosis of ameloblastoma.

Among the cases of ameloblastoma analyzed, 88.6% were classified as solid, which is in agreement with findings described in the literature [14,16,17], with the exception of a multicenter study carried out in Latin America, which found that solid ameloblastoma only accounted for 33.7% of cases [13]. In the evaluation of the histological subtypes of solid ameloblastomas, the follicular and plexiform patterns were the most prevalent, which is in agreement with findings described in previous studies [3,14,17]. A number of authors state that there is little evidence that these histological types have any significance regarding clinical behavior and prognosis. However, such concepts remain controversial and further studies are needed to clarify this issue.

Desmoplastic ameloblastoma accounted for 2.2% of cases in the present study. This is in agreement with findings reported in previous investigations [3,13,14,17], which report prevalence rates ranging from 0.5% to 5.6%. Moreover, Fulco et al. [17] classified 3.7% of cases as hybrid tumors formed by masses of desmoplastic ameloblastoma with significant areas of solid ameloblastoma.

The prevalence of unicystic ameloblastoma was 10.2% in the present investigation. Most studies report a lower prevalence rate of this type of tumor, ranging from 4% to 6% [3,14,16]. The only study to report a greater prevalence rate of unicystic ameloblastoma in comparison to solid ameloblastoma was carried out by Ledesma-Montes et al. [13],

in which 63.7% of cases were classified as unicystic ameloblastoma. The authors explain this high prevalence rate by the technical rigor employed in the differentiation of unicystic ameloblastoma from cases diagnosed as solid ameloblastoma.

Regarding gender, women were more affected by ameloblastoma in the present study (58.4%). Data on gender differences vary in the literature. Some studies report an equal frequency between men and women [17]; others report a predominance of the male gender [10,13,15,18] and still others report a predominance of the female gender [5,11,12,14,16]. These discrepancies suggest that the findings are directly proportional to the profile of the patients who seek medical-dental care in each population.

In the present study, 40.2% of the cases of ameloblastoma occurred prior to the third decade of life. The literature reports considerable variation regarding this variable. However, in a meta-analysis on ameloblastoma, Reichart et al. [3] report a median age of 34 and 24 years upon the initial diagnosis in industrialized and developing countries, respectively.

Studies addressing clinical-epidemiological data on ameloblastoma generally do not assess ethnic background, as this characteristic is strongly associated with cultural and socioeconomic factors. In the present study, ameloblastomas occurred with greater frequency among patients of Caucasian descent (62.9%), which is in agreement with the 72% described by Fregnani et al. [16] in a study on this type of tumor in a sample of Brazilians. However, these data cannot be extrapolated to the national population, as the population of the state of Rio Grande do Sul is 82.3% Caucasian and 5.9% African descent; moreover, metropolitan Porto Alegre is 80.7% Caucasian and 8.0% African descent (IBGE, 2008) [23]. Therefore, a greater number of biopsies from Caucasian individuals are to be expected.

The mandible was the most affected anatomic site (78.6% of cases). This finding is in agreement those reported in all epidemiological studies on ameloblastoma reviewed [3,5,10-13,15,16,18], which demonstrates that the anatomic site of ameloblastomas is not associated with ethnic, culture or socioeconomic issues. However, we know of no explanation for the predominance of the mandible in cases of ameloblastoma.

Due to its retrospective nature, the present study cannot provide data on the current prevalence of ameloblastoma. However, one may affirm that the tumors studied herein were predominantly solid, located in the mandible and affected Caucasian women up to 29 years of age more. The only characteristic unanimously reported in all populations is the preferential location of ameloblastoma in the mandible, whereas the prevalence of the other characteristics varies in accordance with the demographic characteristics of each country. The present findings are in agreement with those reported in previous studies carried out in other states of Brazil and support that notion of a geographical influence on the profile of patients affected by ameloblastoma.



CONCLUSION

The present findings are in agreement with those reported in previous studies carried out in other states of Brazil and support that notion that there may be a geographical influence on the profile of patients affected by ameloblastoma.

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