



Odontogenic tumors: retrospective study of 32 cases diagnosed in a stomatology center in Maringá, Paraná, Brazil

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ABSTRACT. The knowledge of the epidemiological characteristics such as anatomic location, age, and gender of the patients with odontogenic tumors is extremely valuable in the development of differential diagnoses. In the present observational and retrospective study, the objective was to check the occurrence of these tumors in patients attended at the project of Oral Lesions: 'Diagnosis, treatment and epidemiology of diseases of the oral cavity' of the State University of Maringá, between 1995 and 2015. Data were obtained from the project charts, in what concerns the variables: gender, age, race, diagnosis, lesion site, time of evolution, symptomatology and radiographic characteristics. Tumors were classified in agreement with the World Health Organization (2005). A total of 2581 records were analyzed. Among them 32 (1.45%) showed the diagnosis of odontogenic tumor. There was no statistically significant difference among genres. The age group with greater incidence was 23-33 years old. Among the patients, 55.3% stated they did not know which the time span of evolution of the tumor. There was greater incidence in people of Caucasian origin and the most affected site was the left posterior region of the mandible. The keratocystic odontogenic tumor, followed by ameloblastoma and odontoma were shown to be the most common neoplasms found in the patients who participated in the project.

Keywords: oral lesions, prevalence, early diagnosis.

Tumores odontogênicos: estudo retrospectivo de 32 casos diagnosticados em um centro de estomatologia em Maringá, Paraná, Brasil

RESUMO. O conhecimento das características epidemiológicas como localização anatômica, idade e gênero dos pacientes portadores de tumores odontogênicos é extremamente valioso no desenvolvimento de diagnósticos diferenciais. No presente estudo observacional e retrospectivo, o objetivo foi checar a ocorrência destes tumores em pacientes atendidos no projeto de Lesões Bucais: 'Diagnóstico, tratamento e epidemiologia de doenças da cavidade oral' da Universidade Estadual de Maringá, entre 1995 e 2015. Foram utilizados dados contidos nos prontuários, no que diz respeito às variáveis: gênero, idade, etnia, diagnóstico, localização da lesão, tempo de evolução, sintomatologia e características radiográficas. Os tumores foram classificados de acordo com a Organização Mundial de Saúde (2005). Foram analisados 2.581 prontuários, em que 32 (1,45%) mostraram o diagnóstico de tumor odontogênico. Não houve diferença estatisticamente significativa entre gêneros, sendo a faixa etária mais acometida entre 23-33 anos; 55,3% dos pacientes relataram não saber qual o tempo de evolução do tumor; houve maior incidência em pessoas de ascendência caucasiana e o local mais afetado foi a região posterior esquerda de mandíbula. O tumor odontogênico queratocístico, seguido por ameloblastoma e odontoma foi a neoplasia mais comumente encontrada nos pacientes participantes do projeto.

Palavras-chave: lesões orais, prevalência, diagnóstico precoce.

Introduction

Odontogenic tumors (OTs) comprise a complex group of lesions with diverse clinical behaviors and histological types. Some of these lesions are true neoplasms and rarely present a malignant behavior. Others may represent malformations similar to tumors (hamartomas). Odontogenic tumors similar to normal odontogenesis present various inductive

interactions between the odontogenic epithelium and odontogenic ectomesenchyme. (Neville, 2005)

Some OTs are composed of odontogenic epithelium only, without participation of the odontogenic mesenchyme. Others, denominated mixed odontogenic tumors, are composed of odontogenic epithelium and elements of the ectomesenchyme. Calcified dental tissue may or

may not be formed in these lesions. A third group of odontogenic tumors is composed mainly of odontogenic ectomesenchyme (Tawfik & Zyada, 2010). The odontogenic epithelium may be included among these lesions, but does not play any essential role in their pathogenesis. Therefore, in 2005, the World Health Organization defined them as: epithelial odontogenic tumors, mixed odontogenic tumors and ectomesenchymal tumors, according to the tissue of origin (Reichart & Philipsen, 2004).

With the main purpose of early diagnosis and treatment of oral lesions, in 1995, the Dental Department of the State University of Maringá created the Extension Project: "Diagnosis, treatment and epidemiology of diseases of the oral cavity (*Diagnóstico, tratamento e epidemiologia das doenças da cavidade bucal - LEBU*"). At present this project is a reference in the area of oral lesions in the State of Paraná, and patients from Maringá and region are referred to the 15th Regional of Health to be treated by the academic staff and residents in surgery and bucco-maxillo-facial traumatology, under the supervision of professors. During appointment, the patients are evaluated with regard to the aspect of normality of oral tissues, and when there are alterations they undergo physical and complementary exams required to diagnose lesions. It is provided not only the best form of treatment, but the practice of a complete approach to health care. (Jing et al., 2007)

Therefore, the aim of this study was checking the occurrence of OTs in a Service of Oral Diagnosis of the Maringá State University (LEBU Project), which is reference in the management of patients with oral lesions, from 1995 to 2015.

Material and methods

A retrospective, observational type of survey was conducted of the biopsies performed in the LEBU Project, in order to quantify the number of cases of OTs diagnosed in the period from 1995 to 2013. From a total sample of 2581 record charts, the data of each patient were collected. From this selection, all the other forms of lesions found in the oral cavity were excluded, in addition to all the record charts to which the histopathological report was not attached, making it impossible to recognize the conclusive diagnosis, even if the diagnostic hypothesis were some type of odontogenic tumor.

Demographic data such as the patient's identification, gender, skin color and age, and clinical aspects such as time of evolution, symptomatology, radiographic aspect and location

were collected. As regards the location of the neoplasms, the mandibular or maxillary involvement was determined, divided into the anterior and posterior regions.

Lesions were considered OTs, according to the WHO classification in the year 2005. (Barnes, Eveson, Reichart, & Sidransky, 2005).

A database was organized with the variables, which served to support tabulation and statistics. The data obtained were digitized in a Microsoft Excel 2010 software program spreadsheet, and were statistically analyzed by means of the *Software Statistica* 8.0. program. Percentage and frequency tables and graphs were used followed by the Chi-square test to verify possible associations between the variables evaluated. The level of significance adopted was 5%; that is to say the associations with $p < 0.05$ were considered significant.

The present study was approved by the Ethics Committee on Research with Human Beings (Report N°. 303.660, 03/06/2013).

Results

There were 32 cases of OTs found, accounting for 1.47% of the 2581 oral lesions biopsied during the study period. Of these 32 cases, 100% were benign odontogenic tumors. As regards gender, 58% (19) of all the tumors affected women and 42% (13) affected men. With reference to race, 82% (26) were found in Caucasians, 5% (2) in mulattoes and 13% (4) black individuals. The incidence as regards the age of patients ranged from 12 to 72 years, with the most affected being those from 23-33 years of age, accounting for 34.2%. There was no statistically significant difference for any of the variables ($p > 0.05$).

The site most affected by the tumors was the mandible with 71% (23) of the cases, against 26.3% (9) in the maxilla, with posterior region of the mandible being the most frequently affected with 31.5% (12) of the total number of cases.

According to the symptomatology of the lesions, 14 (45%) were asymptomatic, 12 (37%) presented painful symptoms and in six cases (18%) this information was not stated on the record charts. The majority of the patients (55.3%) stated that they did not know for what length of time the neoplasms had been developing. There was no statistically significant difference for any of the variables ($p > 0.05$).

In Table 1, the odontogenic tumors found in the present study are listed, together with their respective prevalence rates. The Keratocystic Odontogenic Tumor (KOT) was the most frequently occurring type in this study, with 16 of

the cases (in one case it was found concomitantly with a unicystic ameloblastoma), followed by ameloblastoma (6) and odontoma (5).

Table 1. Prevalence of neoplasms diagnosed in patients referred to the LEBU Extension Project.

Tumors			N	%
KOT			15	46.87
Ameloblastoma	Solid	4	6	18.75
	Unicystic	2		
Odontoma	Composite	1	5	15.62
	Complex	4		
Mixoma			2	6.25
Odontogenic fibroma			1	3.12
Adenomatoid			1	3.12
odontogenic tumor				
Calcifying cystic			1	3.12
odontogenic tumor				
KOT + Unicystic			1	3.12
Ameloblastoma				
Total			32	100

Discussion

Historically, man has sought to define and live a healthy life, which depends on individual (behavioral actions, dependent on the individual) and group (more complex actions dependent on conditions of life and the medium) actions. At LEBU, the students' goal was to perform these two actions of health promotion, because in addition to invasive clinical intervention (when necessary), they instructed the patients on how to acquire healthy habits and organized epidemiological surveys, researches and studies that served as a basis for the development of prevention strategies (the best way of obtaining health). In addition to health promotion, the aim was the prevention of disease, recovery and rehabilitation of those who were sick, thereby performing all the actions responsible for providing individuals with a good quality of health. The epidemiologic surveys conducted at the LEBU Project are of great value. The care has been provided for almost 20 years, and is in high demand by patients once the project is a reference throughout the entire region of Maringá.

According to Neville (2009), OTs are uncommon lesions in which, even in specialized maxillofacial and oral pathology laboratories, less than 1% of all the samples received are composed of that type of lesion. In the literature this prevalence is variable, however, it does not exceed the value of 5%, as demonstrated by Gupta and Ponnyah (2010), Luo and Li (2009), Saghraevanian, Jafarzadeh, Bashardoost, Pohlavan, and Shirinbak (2010), Servato, Prieto-Oliveira, Faria, Loyola, and Cardoso (2013) and Osterne, Brito, Alves, Cavalcante, and Souza (2011) and the present study, in which the prevalence rates found were 4.13, 2.97, 1.9, 3, 3.92

and 1.45%, respectively. Among the 32 cases of OTs diagnosed in our survey, all were benign. Taghavi, Rajabi, Mehrdad, and Sajjadi (2013) in their study with 188 cases of OTs found that only 3.78% were malignant; the same was concluded by Gupta and Ponnyah (2010) and Jing et al. (2007) in whose studies the percentage of malignant OTs was 3%, confirming their uncommonness.

Concerning the gender of patients, we found the distribution of OTs to be similar between the two genders, with no statistically significant difference between them ($p=0.0786$). However, a slight more tendency on women (58.3%) was observed, corroborating the studies of Avelar, Antunes, Santos, Andrade, and Dourado (2008) and Osterne et al. (2011), both Brazilian. In Iran, Taghavi et al. (2013) found that in general, OTs had a slight preference for men. In our study, the odontoma followed by the KOT and ameloblastoma were the TOs most frequently found in women; in men the prevalence of KOT was (75%), corroborating some of the studies. (Tawfik & Zyada, 2012; Jing et al., 2007).

The white race was the onemostly affected the by the OTs, with 81.57% of the cases, followed by the black race 13.15% and mulatto 5.26% descendants. In none of the studies reviewed there was an approach to the variable race, thus not allowing us to make a comparison between researches. In the literature, we found no reliable demographic index with regard to ethnicity of the population of Maringá and region to justify the prevalence of the OTs. One only states that the population is composed of descendants of various ethnicities: Polish, Italian, German, Ukrainian, Dutch, Spanish, Japanese and Portuguese, and by the majority of immigrants coming from the States of Rio Grande do Sul, Santa Catarina, São Paulo and Minas Gerais. (Instituto Paranaense de Desenvolvimento Econômico e Social [IpardeS], 2013)

The age of the patients diagnosed varied widely (12-78 years), in agreement with the studies reviewed. (Osterne et al., 2011; Olgak, Koseoglu, & Aksakalli, 2006). The second and third decades of life were the ones affected the most by OTs, accounting for 63.15% of the total number of cases diagnosed, among which 45.11% occurred between 11-22 years, and 54.17% between 22-33 years of age, corroborating the majority of the studies reviewed. Only Osterne et al. (2011) related that 65.83% of the patients were in the second to fourth decades of life. According to Aadithya and Urs (2014) in pediatric patients (0-16 years), the rate of OTs is 26.3% (45/171 biopsies performed). In our study, no OT was found in patients from 0-11 years of age. About

10 OTs were present in individuals from 12-22 years old, of KOT and mixoma subtypes.

Only three types of OTs were diagnosed in the second decade of life in the present study: KOT (63.63%), odontoma (27.27%), and calcifying cystic odontogenic tumor (9.1%). The OTs found in the third decade of life were KOT (33.33%), complex odontoma (22.22%), adenomatoid odontogenic tumor (11.11%), unicystic ameloblastoma (11.11%), solid ameloblastoma (11.11%) and one case of KOT concomitant with unicystic ameloblastoma (2.63%).

Concerning the symptomatology of lesions, 45% were reported to be asymptomatic and 37% with painful symptomatology. However, in 18% of the cases this information was not stated on the record chart ($p=0.45941$). Due to the incomplete filling out of this datum there could be no confidence about any statement regarding the symptomatology of the OTs in the present research. Of all the studies reviewed, only three worked with symptomatology, about which there was a consensus between Avelar et al. (2008) and Jing et al. (2007) that the majority of OTs are asymptomatic, whereas Servato et al. (2013) affirmed that half of the cases found presented symptoms, and associated painful symptomatology of the lesions with the patient's delay in seeking care, because the lesions were already at an advanced stage.

In the present study an unprecedented variable was evaluated, not found in previous researches: the time of evolution of the lesions. Perhaps due to the fact that it is a subjective datum that depends 100% on the patient's perception, and that there is no exact date to state, but rather an approximate one. Therefore, according to our findings, 55.3% of the patients stated that they did not know for how long the lesions had been developing. This datum alerts us about the need for a persistent policy on the prevention of diseases of the oral cavity, which must be adopted by all means of health promotion, making the population aware of the importance of self-exam and regular visits to the dentist.

Regarding the distribution of the OTs between the mandible and maxilla, our findings were in agreement with many previously published studies (Jing et al., 2007; Taghavi et al., 2013; Avelar et al., 2008; Olgac et al., 2006), in which the mandible has been most affected. Only Gupta et al. 2010 related a higher incidence of OTs in the maxilla.

The lesion with the highest prevalence in this study, with 46.8% of cases was the KOT (Table 1), in agreement with previous researches (Servato et al., 2013). However, other studies (Jing et al., 2007; Gupta & Ponnyah, 2010; Osterne et al., 2011; Taghavi et al., 2013) found the solid ameloblastoma to be the lesion most commonly found. In the

present research, the ameloblastoma showed a prevalence of 18.7%, ranking it as the second most common tumor, followed by the odontoma with a prevalence of 15.6%. It is important to emphasize that the introduction of the KOT into the group of OTs was the most significant alteration performed by the WHO in 2005, since it numerically and significantly changed the prevalence of these lesions. This fact consequently limits the number of relevant studies available (Luo & Li, 2009; Osterne et al., 2011; Taghavi et al., 2012; Olgac et al., 2006) for this research, because some authors, such as Saghavanian et al. (2010) still hesitate to use it.

In the present study the KOT could be found in a large portion of the age-ranges, with greater prevalence in the white race and male gender, corroborating the findings of Luo & Li (2009) and in contrast with other surveys (Jing et al., 2007; Osterne et al., 2011; Avelar et al., 2008) in which there was greater incidence on the female gender. The majority of lesions were asymptomatic and the most affected site was the posterior region of the mandible (60%), in agreement with other studies (Jing et al., 2007; Luo & Li, 2009; Osterne et al., 2011). The radiographic findings of KOT were in agreement with those described by Neville (2009) of a "radiolucent area, with sclerotic margins that were frequently well defined, with larger lesions possibly being shown to be multiloculated, and an unerupted tooth being involved in the lesion in 25 to 40% of the cases". In our survey, only one of the 15 lesions presented involvement of an unerupted tooth.

Ameloblastoma was the second most frequently found OT, affecting the mandible in 75% of the cases (Osterne et al., 2011; Taghavi et al., 2013; Avelar et al., 2008; Olgac et al., 2006); with greater incidence in the posterior region. (Osterne et al., 2011; Taghavi et al., 2013; Avelar et al., 2008). Avelar et al. (2008) reported that the ameloblastoma did not present preference for gender. Its occurrence was restricted to the white race, contrary to the findings of Servato et al. (Servato et al., 2012) in which 60% of cases occurred in non white patients. The peak incidence was between the third and fourth decades of life, corroborating the findings of Osterne et al. (2011). Of the six cases of ameloblastoma diagnosed in our study, in three of them (50%) the patients related some type of symptomatology, in contrast with the findings of Avelar et al. (2008) in which 79% of the cases found were asymptomatic. A multilocular radiolucent area was the radiographic aspect most frequently found, corroborating the findings of other studies. (Neville, 2009; Taghavi et al., 2013).

Odontoma was found exclusively in the female gender, confirming the prevalence described by other studies (Jing et al., 2007; Osterne et al., 2011; Avelar

et al., 2008) however, in disagreement with Gupta et al. (2010) in whose study found a higher incidence in men. Its incidence is restricted to the second and third decades of life, as described by Jing et al. (2007) with predilection for the white race. Of the five cases found, four were complex odontomas. The most affected site was the maxilla, as was the case in the majority of the studies reviewed (Jing et al., 2007; Osterne et al., 2011; Taghavi et al., 2013; Avelar et al., 2008; Olgac et al., 2006). Taghavi et al. (2013) debated about the difficulty in making a comparative analysis of the frequency of odontoma, considering those tumors are interpreted as malformations and hamartomas instead of as true neoplasms.

The difficulty in locating some records, especially when the long period of 20 years is considered as one of the limitations of this study. Also, the lack of some data in the medical records of some patients also limited the conduction of the study. These are frequent limitations in epidemiological studies. Anyway, this study aimed to help health professionals who are frequently faced with cases of oral lesions, by providing them with epidemiological information obtained by means of a retrospective evaluation, enabling them to attempt to promote the health of patients.

Conclusion

According to the applied methodology, a prevalence of 1.45% of OTs diagnosed at the LEBU Project was observed during a period of 20 years. From them, the most common was the KOT, followed by ameloblastoma and odontoma. No malignant OT was found. The OTs showed a slight predilection for the female gender and white race, and the second and third decades of life were the most affected; nevertheless there was no statistically significant difference.

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