

Prevalence of tumours of the maxillomandibular complex diagnosed in a reference center in Brazil

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Tumors of the maxillomandibular complex are a heterogeneous group of lesions with a wide spectrum of clinical and histopathological characteristics. **Aim:** To evaluate the prevalence of odontogenic and non-odontogenic tumors associated with maxillary bones in a Reference Center for Oral Lesions. **Methods:** A cross-sectional study based on the medical records of a Reference Center for Oral Lesions at the State University of Feira de Santana, from 2006 to 2018. The data was initially analyzed in a descriptive manner. For bivariate analysis, Pearson's chi-square test was applied. The level of significance was set at 5%, where $p \leq 0.05$ is considered significant. **Results:** The prevalence of tumors was 2.27%. The average age of the individuals was 22.2 (± 15.1) years, the majority being up to 39 years (79.59%) and female (69.40%). A statistically significant difference was observed in relation to age ($p = 0.00$), as well as regarding the location of tumors in the anterior or posterior region ($p = 0.02$). Odontogenic tumors were benign, with odontoma being most frequent (46.90%), followed by ameloblastoma (16.30%). As for the non-odontogenic, neurofibroma (4.10%) and osteoma (4.10%) were the most common across the benign, while osteosarcoma accounted for 6.10% of cases. **Conclusion:** Odontogenic tumors were the most frequent in women, with age up to 39 years, odontoma being most common in the posterior region of the mandible. Among non-odontogenic tumors, central neurofibroma and osteoma were the most common. Osteosarcoma was more frequent in men over 40 years old and in the mandible region.

KEYWORDS: Diagnosis. Prevalence. Pathology, oral. Cross-sectional studies. Epidemiology.



Introduction

The maxillomandibular complex is subject to the development of several conditions¹. Intraosseous lesions of the jaws constitute a heterogeneous group of lesions that present a wide spectrum of clinical and histopathological characteristics, ranging from cysts, tumors and bone-associated lesions².

The first internationally accepted classification for maxillofacial lesions was published by the World Health Organization (WHO) in 1971, which has been modified over the years (1992 and 2005), in an attempt to better define its diagnostic criteria³. The most recent WHO edition, in 2017, introduced some changes in the 2005 classification, and, in addition to the return of odontogenic cysts, two new entities were included: sclerosing odontogenic carcinoma and primordial odontogenic tumor⁴. This new classification focuses on those that are biologically benign or malignant, signaling a simplification of the previous version⁵.

Odontogenic tumors form a complex group of lesions that range from hamartomatous or non-neoplastic proliferations to malignant neoplasms with metastatic capacity⁶, originating from the remnants of the tooth-forming apparatus⁷. Regarding non-odontogenic tumors, their classification is not yet well established, whether this would be according to the original tissue or according to its topography. According to the WHO, these tumors have a predilection for the mandibular bone, but some of the lesions, such as chondroma, chondrosarcoma and osteosarcoma, although occurring in the mandible, do not essentially show a greater predilection for this site^{8,9}.

In Brazil, epidemiological studies show that, among all diagnosed oral lesions, 1.3 to 4.8% are odontogenic tumours¹⁰, presenting results similar to other Latin American countries, such as Chile and Mexico¹¹. Regarding non-odontogenic tumors, a study conducted in Queensland, Australia, showed that benign non-odontogenic lesions were 6.8 times more likely to appear than malignant non-odontogenic lesions¹².

National epidemiological studies related to bone lesions are scarce¹³. These studies have an important relevance for the knowledge of the population profile, as well as the injuries that can occur in the stomatognathic system, helping in the early diagnosis and treatment. Thus, this study aimed to describe the prevalence of odontogenic and non-odontogenic tumors of the maxillomandibular complex in a Reference Center in Brazil.

Materials And Methods

Design, area of study and characterization of the sample

This is a retrospective cross-sectional study, based on secondary data from medical records and conclusive anatomopathological reports of individuals diagnosed at a Reference Center for Oral Lesions (CRLB) in the Department of Health Sciences, State University of Feira de Santana (UEFS). This research was registered and approved by Research Ethics Committee of the Institution where it was carried out (Protocol Number: 015/2008, CAAE 0015.0.059.000-08).

Cases of odontogenic and non-odontogenic tumors were selected between 2006 to 2018 and classified according to the current WHO classification (2017)⁵. The criteria

for exclusion were: medical records that had only descriptive reports; reports with the same registration number; and different diagnoses for the same patient.

Data collection and selected variables

This collection was performed by a single examiner trained to complete the collection worksheet. Data regarding gender, age, anatomical site (maxilla versus mandible; posterior versus anterior) and histopathological type were obtained from patients' records, which also contained the Informed Consent Form duly signed by the patient or guardian.

Data analyses

The data was initially analyzed descriptively. For the bivariate analysis, the normality of the data was verified with the Kolmogorov-Smirnov test. Then, Pearson's chi-square test was applied for categorical variables. The level of significance used was 5%, where $p \leq 0.05$ was considered significant. Analysis was carried out using the Statistical Package for the Social Sciences software, version 17.0. (SPSS Inc., Chicago, IL, USA).

Results

During the study period, all 2,156 histopathological reports of oral lesions, diagnosed at CRLB, were evaluated. Of these, 290 were diagnosed with some type of intraosseous lesion, and 49 were conclusive for tumors of the maxillofacial complex, representing a prevalence of 2.27%.

The mean age of the individuals was 22.2 (± 15.1) years. The majority, 39 (79.59%), were in the group of up to 39 years and were female (69.40%). Statistically significant differences were observed between tumors of the maxillomandibular complex and other intraosseous lesions in relation to age ($p = 0.00$), as well as regarding the anatomical site (anterior versus posterior) ($p = 0.02$) (Table 1).

Table 1. Bivariate analysis of tumors of the maxillomandibular complex and other intraosseous lesions, CRLB, UEFS, 2006-2018.

Variables		Tumors	Other intraosseous lesions	
n (%)	n (%)	n (%)	p	
Age range	Up to 39 years	39 (79.59%)	123 (51.10%)	0.00*
	From 40 years	10 (20.41%)	118 (48.90%)	
Gender	Female	34 (69.40%)	164 (68.00%)	0.85
	Male	15 (30.60%)	77 (32.00%)	
Anatomical site (Mandible vs Maxilla)	Mandible	27 (55.10%)	140 (58.10%)	0.70
	Maxilla	22 (44.90%)	101 (41.90%)	
Anatomical site (Posterior vs Anterior)	Anterior	27 (55.10%)	90 (37.30%)	0.02*
	Posterior	22 (44.90%)	151 (62.70%)	

* $p < 0.05$

Benign odontogenic tumors (39) were the most common. Odontoma was the most frequent 23 (46.90%), followed by ameloblastoma 08 (16.30%). Cementoblastoma 3 (6.10%) was the third most common. No malignant odontogenic tumor was diagnosed. As for non-odontogenic tumors (10), central neurofibroma 02 (4.10%) and osteoma 02 (4.10%) were the most common, while osteosarcoma was the malignant tumor present in 3 (6.10%) of the cases (Table 2).

Table 2. Tumors of the maxillomandibular complex according to frequency and percentage, CRLB, UEFS, 2006-2018.

Tumors of the maxillomandibular complex	n	%
Odontogenic tumors		
Odontoma	23	46.90
Ameloblastoma	08	16.30
Cementoblastoma	03	6.10
Myxoma	02	4.10
AOT*	02	4.10
CEOT**	01	2.05
Non-Odontogenic tumors		
Benign		
Central neurofibroma	02	4.10
Osteoma	02	4.10
Fibroblastoma	01	2.05
Hemangiopericitoma	01	2.05
Malignant		
Osteosarcoma	03	6.10
Small round cell sarcoma	01	2.05
Total	49	100

* Adenomatoid odontogenic tumor; ** Calcifying epithelial odontogenic tumor

Table 3 shows the distribution of odontogenic and non-odontogenic tumors by age, gender and location.

Table 3. Distribution of odontogenic and non-odontogenic tumors according to age, gender and location, CRLB, UEFS, 2006-2018.

Tumors	Age (years)		Gender		Location	
	1-39	≥ 40	Female	Male	Maxilla	Mandible
Odontogenic						
Odontoma	21	02	16	08	08	08
Ameloblastoma						
Unicystic	05	00	05	00	00	05
Ameloblastoma	02	01	01	02	00	03
Cementoblastoma	02	01	03	00	01	02
Mixoma	02	00	02	00	00	02
AOT*	01	01	01	01	01	01
CEOT**	01	01	01	01	01	01

Continue

Continuation

Tumors	Age (years)		Gender		Location	
	1-39	≥ 40	Female	Male	Maxilla	Mandible
Non-odontogenic						
Benign						
Centra Neurofibroma	01	01	01	01	00	02
Osteoma	00	02	01	01	02	00
Fibroblastoma	01	00	01	00	01	00
Hemangiopericitoma	01	00	01	00	00	01
Malignant						
Osteossarcoma	01	02	01	02	01	02
Small round cell sarcoma	01	00	00	01	00	01

* Adenomatoid odontogenic tumor; ** Calcifying epithelial odontogenic tumor

Discussion

In this study, intraosseous lesions of the jaws showed a prevalence of 13.45%, based on the 13-year analysis. A low prevalence (25%) was reported by Jaafari-Ashkavandi and Akbari² (2017) after 22-year of collecting data, which differs from the findings of Ali¹⁴ (2011), whose percentage was 31% in 5-year follow-up.

Most tumors were of odontogenic origin, corroborating with Parkins et al.¹⁵ (2007). However, a study conducted in Ghana found a greater number of non-odontogenic tumors. This can be justified by the fact that non-aggressive odontogenic lesions, such as odontomas, have not been diagnosed, since only symptomatic patients with facial edema were included in the sample¹⁶.

Odontogenic tumors corresponded to 1.80% of all cases, a result that is similar to other studies^{17,18} and these lesions mainly occurred in female patients. There are studies in the literature reporting similar occurrence between males and females¹⁹; however, some studies have reported a higher prevalence in males^{20,21}, while others also have shown females to be more affected²², corroborating with our result. There is no plausible explanation proven for these differences. Souto et al.²³ (2014) described that a higher prevalence in women can be explained through the fact that women are more likely to seek healthcare, making these lesions more detectable and raising the number of cases in the female gender.

The age group with the most cases was the fourth decade of life, as noted by Kebede, et al.²¹ (2017). In contrast, Pereira, et al.²⁴ (2010) described a higher frequency in individuals over 50 years old. The variation may be related to the different samples and populations analyzed.

Malignant odontogenic tumors were not diagnosed in this study. They are extremely rare lesions, with reported incidences of 1.1%²⁵ and 1.18%²⁶. In Silvera et al.²⁷ (2021) study, the malignant lesions were more common in males and in the mandible, affecting individuals of 55 ± 21.6 years.

Overall, odontomas were the most prevalent of the group, followed by unicystic ameloblastoma, similar to the data from previous publications¹⁹. However, AlSheddi et al.²⁸

(2015) found that odontogenic keratocysts were the most common, followed by ameloblastomas, which can be justified by the difference in the WHO classification, as they used the third edition (2005), which classifies keratocysts as tumors, whereas we used the fourth, which classifies them as a cyst.

In some studies, myxoma was the third most common odontogenic tumour^{27,29}. In our study, adenomatoid odontogenic tumor (AOT) and myxoma had a similar frequency (4.10% each) and were in the fourth position. Calcifying epithelial odontogenic tumor (CEOT) comprised a lower occurrence (2.05%). Ali¹⁴ (2011) observed that CEOT appears as the least frequent among odontogenic tumors.

The majority of the studies on odontogenic tumors demonstrated a strong predilection for the mandible, especially the posterior region^{20,30}. Jaw-specific genetic mechanisms that regulate the evolution and development of upper and lower dentitions appear to differ and this may provide a partial explanation to the difference in the incidence of odontogenic tumors in the mandible versus maxilla³¹. However, Açikgoz, et al.³² (2012) and Kambalimath, et al.³³ (2014), suggested involvement in the maxilla. Regarding odontoma, the highest prevalence was in the maxilla, being in line with other authors^{19,28}.

It is worth mentioning that among the group of non-odontogenic tumors, neurofibroma and osteoma were the most common among the benign, diverging from the findings of Rodrigues, et al.³⁴ (2010) and Johnson, et al.¹² (2013), in which the central giant cell granuloma was the most frequent. Osteosarcoma was the most frequent among the malignant. These are rare, aggressive, with a high mortality rate³⁵. Jaafari-Askavandi and Akbari² (2017) pointed out that this lesion corresponded to 28.1% of the neoplasms in their findings.

Due to this being a descriptive study, the variables analyzed do not allow for inferring causality, requiring further longitudinal studies. However, the results contribute to a better understanding of the clinical-epidemiological profile of individuals and in the development of strategies and actions destined towards diagnosis and treatment of such lesions.

The present study was carried out in a reference center, in which oral pathologists issued a histopathological diagnosis, enabling the evaluation of lesions that affect the jaws. New epidemiological studies on tumors of the maxillomandibular complex should be carried out in Brazil, in order to obtain greater knowledge surrounding their behavior, thus improving their diagnosis and treatment.

In conclusion, the prevalence of tumors of the maxillomandibular complex was 2.27%. Odontogenic tumors were the most frequent in women, aged up to 39 years, odontoma being the most common in the posterior region of the mandible. Among non-odontogenic tumors, central neurofibroma and osteoma were the most common. Osteosarcoma was more frequent in men over 40 years old and in the mandible region.

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