

Evaluation of cysts and odontogenic tumors over a period of 8 years in a reference medical hospital in Brazil

Avaliação de cistos e tumores odontogênicos ao longo de 8 anos em hospital médico de referência no Brasil

Evaluación de quistes y tumores odontogénicos durante un período de 8 años en un hospital médico de referencia en Brasil

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Abstract

Objective: This study aimed to evaluate the relative prevalence, demographic distribution and clinical-pathological characteristics of the maxillary odontogenic cysts and tumors of the jaws in a pathology laboratory located in a university hospital in Northeastern Brazil. **Methods:** An observational and retrospective study was carried out, in which information was collected on histopathological diagnosis, age, gender and location of the lesion of patients who had been diagnosed with odontogenic cysts or odontogenic tumors between 2013 and 2020. **Results:** Among the 546 specimens, 57 were odontogenic cysts and 41 were odontogenic tumors. The most frequent odontogenic cysts were the odontogenic keratocyst 21 (21.4%) and the root cyst 19 (19.4%). The most frequent odontogenic tumors were ameloblastoma 27 (27.6%) and odontoma 6 (6.1%). **Conclusion:** The data found in this study are possibly associated with the service assistance profile.

Keywords: Odontogenic cysts; Odontogenic tumors; Neoplasms of head and neck; Oral and Maxillofacial Pathology.

Resumo

Objetivo: Este estudo teve como objetivo avaliar a prevalência relativa, distribuição demográfica e características clínico-patológicas dos cistos e tumores odontogênicos maxilares em um laboratório de patologia localizado em um hospital universitário do Nordeste do Brasil. **Métodos:** Foi realizado um estudo observacional e retrospectivo, no qual

foram coletadas informações sobre diagnóstico histopatológico, idade, sexo e localização da lesão de pacientes com diagnóstico de cistos ou tumores odontogênicos entre 2013 e 2020. *Resultados:* Entre os 546 espécimes, 57 eram cistos odontogênicos e 41 eram tumores odontogênicos. Os cistos odontogênicos mais frequentes foram o ceratocisto odontogênico 21 (21,4%) e o cisto radicular 19 (19,4%). Os tumores odontogênicos mais frequentes foram ameloblastoma 27 (27,6%) e odontoma 6 (6,1%). *Conclusão:* Os dados encontrados neste estudo estão possivelmente associados ao perfil de atendimento do serviço.

Palavras-chave: Cistos odontogênicos; Tumores odontogênicos; Neoplasias de cabeça e pescoço; Patologia Oral e Maxilofacial.

Resumen

Objetivo: Este estudio tuvo como objetivo evaluar la prevalencia relativa, distribución demográfica y características clínico-patológicas de quistes y tumores odontogénicos maxilares en un laboratorio de patología ubicado en un hospital universitario del noreste de Brasil. *Métodos:* se realizó un estudio observacional y retrospectivo, en el que se recogió información sobre diagnóstico histopatológico, edad, sexo y localización de la lesión de pacientes diagnosticados de quistes o tumores odontogénicos entre 2013 y 2020. *Resultados:* de las 546 muestras, 57 fueron quistes tumores odontogénicos y 41 fueron tumores odontogénicos. Los quistes odontogénicos más frecuentes fueron el queratocisto odontogénico 21 (21,4%) y el quiste radicular 19 (19,4%). Los tumores odontogénicos más frecuentes fueron el ameloblastoma 27 (27,6%) y el odontoma 6 (6,1%). *Conclusión:* Los datos encontrados en este estudio posiblemente estén asociados con el perfil de servicio del servicio.

Palabras clave: Quistes odontogénicos; Tumores odontogénicos; Neoplasias de cabeza y cuello; Patología oral y maxilofacial.

1. Introduction

Many studies on oral health are based only on clinical data that, although important, do not inform the actual pathological nature of the lesion. Therefore, studies based on biopsies are essential to determine the prevalence of lesions that commonly affect a given population. Additionally, identifying the frequency of oral and maxillofacial lesions is essential for establishing the diagnosis and treatment of the patient. (Joseph et al., 2019); knowledge of the location, age, and gender predilections of different diseases of the oral cavity is useful with regard to determining their demographic data; and, finally, it is essential for the planning and execution of public health programs. (Alhindi et al., 2019).

Odontogenic cysts and tumors of the jaws represent one of the most prevalent groups of oral and maxillofacial lesions (Açikgöz et al., 2012). Maxillary cysts can be defined as pathological intraosseous cavities filled with fluid, semi-solid or gaseous material, partially or totally covered by epithelial tissue and delimited by a connective tissue capsule. When the cyst develops from remaining odontogenic epithelial tissues, it is called an odontogenic cyst. This category is then subdivided according to its inflammatory or developmental origin. (Philipsen et al., 2006; Nuñez-Urrutia et al., 2010; Demirkol et al., 2014). Odontogenic tumors were first described in the scientific literature in 1869, when Pierre Paul Broca introduced the term "odontoma" to classify a tumor lesion originating from tooth-forming tissues (Philipsen et al., 2006). They correspond to a heterogeneous group of lesions ranging from hamartomas to neoplastic or malignant lesions derived from ectodermal and/or mesenchymal odontogenic tissues (Aregbesola et al., 2018).

Given the different nature of odontogenic cysts and tumors, several consensus meetings have tried to produce a more systematic classification of these lesions, promoted by the World Health Organization (WHO) in 1971 (Philipsen et al., 2006). Since then, other WHO meetings have reviewed the classifications of these injuries in 1992, 2005 and 2017 (Bianco et al., 2020). Although several case series of odontogenic cysts and tumors have been reported worldwide, few have evaluated and compared the two groups of lesions together. (Daley et al., 1994; Gaitán-Cepeda et al., 2010; Jaeger F et al., 2017). However, the aim of our study was to evaluate the relative prevalence, demographic distribution and clinicopathological characteristics of odontogenic cysts and tumors of the jaws in a pathology laboratory located in a leading university hospital in northeastern Brazil.

2. Methodology

After approval of the University of Pernambuco Ethics Committee (35920620.7.0000.5192), an observational, descriptive and retrospective study was carried of patients (Lima-Costa & Barreto, 2003) who had been diagnosed with odontogenic cysts or odontogenic tumours between 2013 and 2020 from the Pathology Laboratory, University Hospital Oswaldo Cruz, University of Pernambuco, which is one of the most representative public services of health in Brazil. The study was performed in full accordance with the World Medical Association Declaration of Helsinki.

All lesions were revised by 2 oral pathologists and defined according to the criteria of classification of the World Health Organization published in 2017 (El-Naggar et al., 2017). Information regarding the gender, histopathological diagnosis, age and local of the lesion were collected.

Data were tabulated and statistically analyzed using the Statistical Package for Social Sciences version 20.0 software (SPSS, Chicago, IL). Frequencies and the P-value test were used to compare the distribution of odontogenic cysts and tumors associated with independent variables. They were considered significant when the P value was less than 0.05.

3. Results

During the study period, a total of 546 specimens from biopsies of head and neck lesions were received in the pathology laboratory of the University Hospital Oswaldo Cruz of the University of Pernambuco, among the 98 cases received histopathological diagnosis of lesion of odontogenic origin, being 57 (58.1%) odontogenic cysts and 41 (41.9%) odontogenic tumors (Table 1).

Among the 98 cases of odontogenic cysts and tumors analyzed, 62 were male (63.7%) and 36 female (36.3%). The age of the individuals ranged from 8 to 81 years, with a mean of 32.99 years, with greater frequency between the second and third decade of life. The most affected location was the mandible 58 (59.2%), especially the posterior region 31 (79.5%). For maxillary cases 29 (29.6%), the anterior region was the most affected area 12 (92.3%) (Table 1).

Table 1. General characteristics of the patients with odontogenic cysts and odontogenic tumours.

General characteristics of the patients		Total		Odontogenic Cysts		Odontogenic Tumours	
		N	%	N	%	N	%
Gender	Male	62	63,7	37	59,7	25	40,3
	Female	36	36,3	20	55,6	16	44,4
Age	Mean	32,99					
Age decades (years)	0-9	1	1	1	1,7	0	0
	10-19	23	23,5	18	33,3	5	12,2
	20-29	26	26,5	15	26,3	11	26,8
	30-39	20	20,4	7	12,2	13	31,7
	40-49	16	16,3	8	13,8	8	19,5
	50-59	3	3,1	1	1,7	2	4,8
	60-69	3	3,1	2	3,4	1	2,4
	70-79	4	4,1	4	7	0	0
Anatomical jaw location	80-89	2	2	0	0	2	4,8
	Maxilla	29	29,6	10	76,9	3	23,1
	Mandible	58	59,2	16	41	23	59
Anatomical region location	Uniformed	11	11,2	31	54,3	15	36,5
	Maxila Anterior	12	92,3	9	75	3	25
	Maxila Posterior	1	7,7	1	100	0	0
	Mandible Anterior	8	20,5	3	37,5	5	62,5
	Mandible Posterior	31	79,5	13	41,9	18	58,1
	Uniformed	46	46,9	31	54,3	15	36,5

Source: Own authorship.

The most frequent cystic lesion was odontogenic keratocyst 21 (21.4%), being more common in mandible 14 (73.7%), followed by root cysts 19 (19.4%) and dentigerous cysts 14 (14.3%). Among the odontogenic tumors, the most frequent were ameloblastoma 27 (27.6%), being more common in mandible 24 (92.3%), followed by odontoma 6 (6.1%) and myxoma 5 (5.1%). The other cases correspond to 9.1% of the odontogenic tumors analyzed (Tables 2 and 3).

Table 2. Frequency and distribution of Odontogenic Cysts and Tumours per gender and age range.

HISTOPATOLOGIC DIAGNOSIS	Gender		Age								Total	
	Male	Female	0-9	10-19	20-29	30-39	40-49	50-59	60-69	70 >	N	%
Inflammatory Cysts	12	7	0	3	5	4	5	1	0	1	19	19,4
Root cyst	12	7	0	3	5	4	5	1	0	1	19	19,4
Developmental Cysts	24	13	1	15	10	3	3	0	2	3	37	37,7
Odontogenic keratocyst	13	8	0	10	5	2	2	0	0	2	21	21,4
Dentigerous cyst	10	4	1	5	3	1	1	0	2	1	14	14,3
Calcifying odontogenic cyst	1	1	0	0	2	0	0	0	0	0	2	2
Epithelial odontogenic tumours	20	9	0	1	9	10	4	2	1	2	29	29,6
Ameloblastoma	18	9	0	0	8	10	4	2	1	2	27	27,6
Calcifying epithelial odontogenic tumour	2	0	0	1	1	0	0	0	0	0	2	2
Mesenchymal odontogenic tumours	2	5	0	1	1	2	3	0	0	0	7	7,1
Myxoma	2	3	0	1	1	0	3	0	0	0	5	5,1
Odontogenic fibroma	0	2	0	0	0	2	0	0	0	0	2	2
Mixed odontogenic tumours	4	2	0	3	1	1	1	0	0	0	6	6,1
Odontoma	4	2	0	3	1	1	1	0	0	0	6	6,1

Source: Own authorship.

Table 3. Distribution of Odontogenic Cysts and Tumours per location.

HISTOPATOLOGIC DIAGNOSIS	Anatomical jaw location						Anatomical region location					
	Maxilla		Mandible		NI		Posterior		Anterior		NI	
	N	%	N	%	N	%	N	%	N	%	N	%
Inflammatory Cysts	9	50	9	50	1	5,2	2	10,5	3	15,7	14	73,6
Root cyst	9	50	9	50	1	5,2	2	10,5	3	15,7	14	73,6
Developmental Cysts	14	42,4	19	57,6	4	10,8	12	32,4	9	24,3	16	43,2
Odontogenic keratocyst	5	26,3	14	73,7	2	9,5	9	42,8	6	28,5	6	28,5
Dentigerous cyst	8	66,7	4	33,3	2	14,2	2	14,2	3	21,4	9	64,2
Calcifying odontogenic cyst	1	50	1	50	0	0	1	50	0	0	1	50
Epithelial odontogenic tumours	3	11,1	24	88,9	2	6,9	14	48,2	7	24,1	8	27,5
Ameloblastoma	2	7,7	24	92,3	1	3,7	14	51,8	7	25,9	6	22,2
Calcifying epithelial odontogenic tumour	1	100	0	0	1	50	0	0	0	0	2	100
Mesenchymal odontogenic tumours	1	20	4	60	2	28,5	3	42,8	0	0	4	57,2
Myxoma	1	25	3	75	1	25	3	60	0	0	2	40
Odontogenic fibroma	0	0	1	100	1	50	0	0	0	0	2	100
Mixed odontogenic tumours	2	50	2	50	2	16,6	1	16,6	1	16,6	4	66,6
Odontoma	2	50	2	50	2	16,6	1	16,6	1	16,6	4	16,6

Source: Own authorship.

A statistically significant association ($p < 0.05$) was found between the variable anatomical location of the jaws, age and the presence of odontogenic cysts and tumors (Table 4). Odontogenic tumors were less frequent than odontogenic cysts in the maxillary region. However, the occurrence of odontogenic cysts and tumors was significantly greater in the mandible than in the maxilla ($p = 0.002$).

There was no statistically significant association between the distribution of odontogenic cysts and tumors, according to gender and anatomical region. (Table 4).

Table 4. Frequencies and p value test of the distribution of odontogenic cysts and tumors associated with independent variables.

VARIÁVEL	ODONTOGENIC CYSTS		ODONTOGENIC TUMOURS		p value
	N	%	N	%	
Gender	N	%	N	%	0,425
Male	37	59,7	25	40,3	
Famale	20	55,6	16	44,4	
Age	N	%	N	%	0,013
Up to 29 years old	35	70	15	30	
30 years or more	22	45,8	26	54,2	
Anatomical jaw location	N	%	N	%	0,002
Maxila	24	82,8	5	17,2	
Mandible	28	48,3	30	51,7	
Anatomical region Location	N	%	N	%	0,196
Anterior	12	60	8	40	
Posterior	14	43,8	18	56,2	

Source: Own authorship.

4. Discussion

In the period surveyed, 546 lesions were diagnosed in a region of head and neck being 98 (17.94%) of these classified as cysts and odontogenic tumors. Equivalent manner, a study with similar methodology held in the Brazilian Northeast obtained 20.3% (Barros et al., 2019) and another in the Brazilian midfielder obtained 21.22% (Jaeger, 2017). In the meantime, international research diverges from the aforementioned findings, Amrita and employees when analyzing the records of an oral pathology service in Kanpur, India, observed a frequency of only 8.49% for such injuries (Raj et al., 2017). Such a discrepancy of results shows that epidemiological surveys of head and neck lesions in a given geographical area establish the real needs of said population, as well as provide professionals facilities in the elaboration of treatment plans and preventive actions (Karen et al., 2015).

With regard to the prevalence of odontogenic cysts and tumors, it is noted that the cysts are common lesions in the gnathical bones (Açikgöz et al., 2012) and may vary their frequency in the studies between 3.65% to 22.41% (Avelar et al., 2009; Souza et al., 2010; Açikgöz et al., 2012; AlSheddi et al., 2015; Raj et al., 2017). On the other hand, odontogenic tumors are rare (Serkerçi et al., 2015) to be observed the frequency of 2.74% to 11.51% (AlSheddi et al., 2015; Serkerçi et al., 2015; Da Silva et al., 2016; Raj et al., 2017). In this research, the quantitative of odontogenic lesions are almost similar when comparing cysts and tumors. But most of the sample consisted of odontogenic cysts which is in line with the literature. A possible justification for the fact was the inclusion of the odontogenic keratocyst and calcifying odontogenic cyst as odontogenic cysts in the classification of WHO head and neck tumors. The large amount of odontogenic tumors in the sample may be associated with the service assistance profile.

We observed a greater number of cases in males, with no significant difference between odontogenic cysts and odontogenic tumors. This gender distribution was also observed in other studies on odontogenic cysts (Ledesma-montes et al., 2000; Meningaud et al., 2006; Prockt et al., 2008; Tortorici et al., 2008; Açikgöz et al., 2012; Selvamani et al., 2012; Khosravi et al., 2013; Baghaei et al., 2014; Demirkol et al., 2014; Kambalimath et al., 2014) and odontogenic tumors (Lawal et al., 2013; Oginni et al., 2015; Aregbesola et al., 2018). Odontogenic cysts occurred more frequently in the maxilla when compared to odontogenic tumors that occurred more commonly in the jaw, and in accordance with the findings of other population cohorts in odontogenic cysts and odontogenic tumors.

The vast majority of the studies reports a greater frequency of odontogenic lesions in individuals between the second and fourth decade of life (Avelar et al., 2009; Souza et al., 2010; Açikgöz et al., 2012; AlSheddi et al., 2015; Serkerçi et al., 2015; Da Silva et al., 2016; Lima-verde-osterne et al., 2017). These data resemble this study, with the highest frequency of cases between the second and the fourth decade of life and the age range ranging from 8 to 81 years.

As for the anatomical location, there was a predominance of cases in jaw when compared to jaw. The information collected are in consensus with other works, where the jaw is considered the most frequent location of cysts (Avelar et al., 2009; Açikgöz et al., 2012; AlSheddi et al., 2015) and odontogenics tumors (AlSheddi et al., 2015; Sekerci et al., 2015; Da Silva et al., 2016; Lima-verde-osterne et al., 2017). It is worth noting that there is a greater chance of being observed cases in the jaw in cysts than in odontogenic tumors (Avelar et al., 2009; Açikgöz et al., 2012; AlSheddi et al., 2015), similar to the data found in this study.

Odontogenic cysts are classified between inflammatory and development (Castro-Núñez, 2015; Speith & Takata, 2018) being the most common radicular and dentigerous representatives, respectively. In the descending order of prevalence, root cysts are (apical, vertical and lateral variant), followed by the odontogenic cyst and odontogenic keratocyst (Avelar et al., 2009; Souza et al., 2010; Açikgöz et al., 2012; AlSheddi et al., 2015). In this study, distinct findings were obtained, since the odontogenic keratocyst was more common than the others. Divergent data is possibly associated with the service care profile,

amendment in the classification of this injury by WHO 2017 and professional negligence as to the adequate forwarding of specimen with a diagnostic hypothesis of inflammatory basis for the purposes of histopathological analysis.

In relation to the epidemiological data of odontogenic tumors, ameloblastoma was found more frequently (n = 27) when compared to odontoma (n = 6). When the literature considers these epidemiological variations, the explanation lies in possible ethnic and / or geographical differences (Daley et al., 1994; Arotiba et al., 1997; Lu Y et al., 1998; Santos et al., 2001). It is also common to suggest that ameloblastomas are more common in blacks than in Caucasians (Regezi et al., 1978; Gunhan et al., 1990; Daley et al., 1994; Odukoya, 1995; Arotiba et al., 1997; Lu Y et al., 1998). In an epidemiological study of the odontogenic tumors of two Brazilian institutions, both in the state of São Paulo, was observed that at the dental hospital, of 113 cases, odontomas and ameloblastomas corresponded to 39.4 and 22.12% of odontogenic tumors, respectively. On the other hand, at the medical hospital, 75 cases, 60% were ameloblastomas and only 5.3% were diagnosed as odontomas (Fregnani et al., 2002). It should be considered that in many countries ameloblastomas are usually diagnosed in the Faculty of Dentistry, but the treatment is carried out in a medical hospital. This is not the case of odontomas. However, some comparative studies considering dental and medical institutions help clarify the subject (Gunhan et al., 1990; Mosqueda-Taylor et al., 1997). In a retrospective study of 349 Odontogenic tumors in Mexico, including dental and medical hospitals, no odontoma was treated in the last. Therefore, the data encountered in this study are possibly associated with the service assistance profile (Mosqueda-Taylor et al., 1997).

5. Conclusion

Knowledge about clinical-pathological distribution of cysts and odontogenic tumors brings benefits to professionals and patients. The best understanding of the behavior and clinical-pathological profile of the cases diagnosed in this reference service in Pathological Anatomy of the State of Pernambuco contributes to the scientific literature with regard to the study of the pathologies of this anatomical region, as well as subsidizing management in decision-making and resources applied in this health care segment. Further studies including large series should be performed in different regions of the world in order to determine the global epidemiologic profile of these lesions.

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