



Frequency of central odontogenic tumors: a retrospective study in an Iraqi population utilizing 2022 WHO head and neck tumors classification

Frequência de tumores odontogênicos centrais: um estudo retrospectivo em uma população iraquiana utilizando a classificação de tumores de cabeça e pescoço da OMS de 2022

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ABSTRACT

Objectives: Odontogenic tumors occupy an important position among head and neck tumors. Although, rarely encountered in medical practice but they considered increasingly challenging lesions for the clinicians due to their overlapping clinical and histopathological features. This study was designed to determine the relative frequency of central odontogenic tumors in an Iraqi population by utilizing 2022 WHO tumor classification. **Material and methods:** Sixty cases of central odontogenic tumors from a total of 1869 case records were retrieved retrospectively from the file archive of the histopathology laboratory in Baghdad medical city from the period of 2016 to 2021. For each individual case, data regarding age, gender, location, and tumor type were collected and analyzed. **Results:** odontogenic tumors constituted 3.2% of the total cases analyzed mostly benign. The male to female ratio was 1/1. The age of the patients ranged from 11 to 75 years. Most cases were recorded in the third and fourth decades of life (n=31, 51.6%). The most common benign and malignant tumors were ameloblastoma and ameloblastic fibrosarcoma respectively. Most of these tumors located in the mandible (n= 45, 75%). The most common mandibular tumor was ameloblastoma followed by ameloblastic fibroma, and odontogenic myxoma. Regarding maxillary tumors, the predominant tumor was ameloblastoma followed by ameloblastic fibroma, ameloblastic fibrosarcoma, and clear cell odontogenic carcinoma. **Conclusions:** Odontogenic tumors in an Iraqi population occurred more commonly in the mandible and showed no sex predilection. Most cases were diagnosed in third and fourth decades of life and ameloblastoma was the most frequent odontogenic tumor. The relative frequency of malignant odontogenic tumors was 11.67% of all cases studied mostly ameloblastic fibrosarcoma.

KEYWORDS

Odontogenic tumors, WHO tumor classification, Frequency, Ameloblastoma.

RESUMO

Objetivos: Os tumores odontogênicos ocupam uma posição importante entre os tumores de cabeça e pescoço. Embora raramente encontrados na prática médica, eles consideram lesões cada vez mais desafiadoras para os clínicos devido às suas características clínicas e histopatológicas sobrepostas. Este estudo foi desenhado para determinar a frequência relativa de tumores odontogênicos centrais em uma população iraquiana, utilizando a classificação de tumor da OMS de 2022. **Materiais e métodos:** Sessenta casos de tumores odontogênicos centrais de um total de 1.869 registros de casos foram recuperados retrospectivamente do arquivo do laboratório de histopatologia na cidade médica de Bagdá no período de 2016 a 2021. Para cada caso individual, dados sobre idade, sexo, localização e tipo de tumor foram coletados e analisados. **Resultados:** os tumores odontogênicos constituíram 3,2% do total de casos analisados em sua maioria benignos. A proporção entre homens e mulheres era de 1/1.

A idade dos pacientes variou de 11 a 75 anos. A maioria dos casos foi registrada na terceira e quarta décadas de vida (n=31, 51,6%). Os tumores benignos e malignos mais comuns foram ameloblastoma e fibrossarcoma ameloblástico, respectivamente. A maioria desses tumores localizava-se na mandíbula (n= 45, 75%). O tumor mandibular mais comum foi o ameloblastoma, seguido do fibroma ameloblástico e do mixoma odontogênico. Em relação aos tumores maxilares, o tumor predominante foi o ameloblastoma seguido de fibroma ameloblástico, fibrossarcoma ameloblástico e carcinoma odontogênico de células claras. Conclusões: Os tumores odontogênicos em uma população iraquiana ocorreram mais comumente na mandíbula e não mostraram predileção por sexo. A maioria dos casos foi diagnosticada na terceira e quarta décadas de vida, sendo o ameloblastoma o tumor odontogênico mais frequente. A frequência relativa de tumores odontogênicos malignos foi de 11,67% de todos os casos estudados principalmente fibrossarcoma ameloblástico.

PALAVRAS-CHAVE

Frequência, Tumores odontogênicos centrais; Ameloblastoma, Classificação tumoral da OMS.

INTRODUCTION

Residual cells of tooth forming apparatus may give rise to odontogenic tumors (OTs), a heterogenous group of lesions with diverse biological behavior ranging from hamartomatous tumor like growth to malignant tumors with metastatic capability. Despite being fortunately quite uncommon these lesions provide a diagnostic difficulty for clinicians due to their overlapping clinical, radiographical and histopathological characteristics [1]. The world health organization (WHO) classification scheme of tumors in 2017 reclassify (keratocystic odontogenic tumor) as an odontogenic cyst with “odontogenic keratocyst (OKC)” designation. This major modification created a relative change in the incidence of these tumors among literature [2,3]. The current 5th edition of the WHO classification of odontogenic tumors, which was introduced in 2022, was broadly identical to the previous one with the exception of a newly defined entity, adenoid ameloblastoma, which was categorized as a benign epithelial odontogenic tumor [4]. The purpose of current study was to investigate the relative frequency of different histopathological subtypes of central OTs among Iraqi population within five-year period.

MATERIALS AND METHODS

Ninety-six odontogenic tumors from a total of 1869 case records were retrieved retrospectively from the file archive of the histopathology laboratory in Baghdad medical city from the period of 2016 to 2021. Thirty-six files were ignored as they included duplicate entries or insufficient histological data. Recurrent tumors

were considered individual cases. In addition, the exclusion included all cases of odontomas and peripheral odontogenic tumors. Information including age, gender, and anatomical location, was gathered for each case. Whenever there was a doubt regarding the diagnosis, a histopathological examination was carried out using the available slides stained with hematoxylin and eosin (H&E). All data analysis was done via Microsoft Office Excel 2016 sheets.

RESULTS

In this study, 60 cases were odontogenic tumors, which constituted 3.2% of the total cases analyzed. Of these (n = 53 cases, 88.33%) were benign and (n = 7 cases, 11.67%) were malignant. The male to female ratio was 1:1. Most of these tumors were epithelial in origin (n = 46, 76.6%), whereas mixed and mesenchymal tumors were (n = 4, 6.6%), and (n = 10, 16.6%) respectively. The age of the patients ranged from 11 to 75 years, with an average of 34.4 years for males and 35.4 years for females. Most cases were recorded in the third and fourth decades of life (n = 31, 51.6%). As shown in Table I, the most common benign and malignant tumors were ameloblastoma and ameloblastic fibrosarcoma. Mandibular tumors were (n = 45, 75%) while maxillary tumors were (n = 15, 25%). The most common mandibular tumor was ameloblastoma (n = 35, 77.7%), followed by ameloblastic fibroma (n = 5, 11.1%) and odontogenic myxoma (n = 2, 4.4%). Regarding maxillary tumors, the predominant tumor was ameloblastoma (n = 5, 33.3%), followed by ameloblastic fibroma, ameloblastic fibrosarcoma, and clear cell odontogenic carcinoma, respectively.

DISCUSSION

Information derived from literature indicates that published series regarding specifically central odontogenic tumors in the Iraqi population that follow the 2017 and 2022 WHO classifications of tumors is limited [5,6]. Odontogenic tumors (OT) are relatively uncommon lesions with variable incidence [7]. In those used 1992 WHO classification of tumors, the frequency recorded was (3%±2.9%), whereas reports utilizing the 2005 WHO classification documented slightly higher rates of (4.0% ±1.3%) due to the inclusion and reclassification of odontogenic keratocyst as a tumor [8].

The relative frequency of OTs in the current study was 3.2% of the total oral biopsied specimens between 2016 and 2021. However, lower rates were recorded in literature utilizing different WHO tumor classifications worldwide as showed in (Table II). The disparity of OTs incidence among different countries may be related to variations in the environmental, educational, and socioeconomical status of the population. In addition, studies may cover only a limited area of the country or restricted to a few research or medical centers which provides an incorrect perception about the frequency. The mean age of odontogenic tumor cases as this study recorded was 35.4 years with peak occurrence in the third

Table I - Frequency of odontogenic tumors relative to gender, site, and age

Tumor type	Frequency		Gender			Site		Age distribution			
	n	percentage	M	F	M/F	Man	Max	0-19	20-39	40-59	60-79
Ameloblastoma	40	66.67%	21	19	1.10	35	5	2	22	11	5
Ameloblastic carcinoma	2	3.33%	0	2		1	1	0	1	1	0
Adenomatoid odontogenic tumor	1	1.67%	1	0		0	1	1	0	0	0
Clear cell odontogenic carcinoma	2	3.33%	2	0		0	2	0	0	1	1
Calcifying epithelial odontogenic tumor	1	1.67%	1	0		1	0	0	1	0	0
Odontogenic fibroma	1	1.67%	1	0		0	1	0	1	0	0
Odontogenic myxoma	3	5.00%	0	3		2	1	1	2	0	0
Ameloblastic fibroma	7	11.67%	4	3	1.33	5	2	4	3	0	0
Ameloblastic fibrosarcoma	3	5.00%	0	3		1	2	1	1	0	1
Total	60	100%	30	30	1	45	15	9	31	13	7

M, male; F, female; Man, mandible; Max, maxilla; ET, epithelial tumor; MT, mesenchymal tumor; MOT, mixed odontogenic tumor; n, mean number of cases.

Table II - Worldwide studies concerning the frequencies of odontogenic tumors

	Total number of cases	Number of Odontogenic tumors	Rate of OTs in study	number of cases after exclusion of odontomas, POT, COF and OK	Rates of OTs after exclusion	MOT	Rate of odontogenic tumors
Al-Aroomy et al. (2022) [3]	8974	230	2.56%	175	1.95%	5	2.85%
Kaur et al. (2021) [9]	8801	345	3.92%	299	3.39%	11	3.67%
Waheed et al. (2021) [5]	7384	64	0.80%	50	0.67%	1	2%
Suluk-Tekkesin et al. (2020) [10]	53869	1231	2.30%	896	1.66%	16	1.70%
Lima-Verde-Osterne et al. (2017) [8]	9170	376	4.10%	156	1.70%	1	0.64%
Nalabolu et al. (2017) [7]	7400	161	2.17%	98	1.32%	0	0.00%
Sekerci et al. (2015) [11]	7,942	218	2.74%	110	1.38%	13	11.81%
Ramos et al. (2014) [12]	2600	78	3.00%	23	0.88%	0	0.00%
Chrysomali et al. (2013) [13]	29088	652	2.20%	168	0.57%	1	0.59%
Siriwardena et al. (2012) [14]	44,720	1677	3.75%	1047	2.34%	23	2.19%
Varkhede et al. (2011) [15]	2075	120	5.78%	60	2.89%	0	0
Gupta et al. (2010) [16]	11,843	489	4.12%	441	3.72%	15	3.40%
Buchner et al. (2006) [17]	91178	1088	1.10%	245	0.26%	5	2.04%
Ladeinde et al. (2005) [18]	3,337	339	9.60%	314	9.40%	11	3.24%

POT, peripheral odontogenic tumor; COF, cemento-ossifying fibroma; OK, odontogenic keratocyst; MOT, malignant odontogenic tumor.

and fourth decades of life comparable to that reported by others [18,19].

Across several reviews for comparison, ameloblastoma was the most commonly reported OT [7,9,20,21]. This comes in accordance with this study. Controversy regarding which gender predominates in ameloblastoma exists. Cavalcante et al. 2016 reported slight female preference [22]. Ramos et al. [12] stated that ameloblastoma exhibits no gender predilection. The present study showed a slight prevalence in male patients (52.5%). Similar conclusion was documented by other series [23,24]. The jaw most commonly affected was the mandible, with a maxilla to mandible ratio of 1:7. This was consistent with studies from different countries [2,12,24]. A current study showed that ameloblastoma was most commonly reported in the third and fourth decades of life (55%). Equal predilection was reported worldwide [3,8,25].

Odontoma (a hamartomatous tumor like lesion) was thought to be one of most common OT with fluctuating frequencies in many records [11,13]. Underrating odontomas and overrating of tumors with aggressive behavior which required extensive medical interference, might be responsible for such discrepancy. Furthermore, the diagnosis of odontoma can be made with high confidence using clinical and radiographic methods; as a result, odontoma biopsies are occasionally referred for histopathological evaluation, or the diagnosis is not documented. For all these reasons, odontoma was excluded in this study.

Ameloblastic fibroma (AF) represent a rare benign OT with an epithelial and mesenchymal neoplastic proliferation [26]. In the current study, AF was recognized in 7 cases, encompassing 11.6% of the entire sample. The patients ranged in age from (11 to 33) years. The highest incidence (57.14%) was recorded in the 2nd decade. There were (n = 4, 57.14%) males and (n = 3, 42.86%) females. The site of the lesion demonstrates a predilection for the mandible (71.4%). Disparity has been reported among series globally regarding the frequency of this tumor [8]. One explanation is that histologically, a disorganized combination of both dental epithelium and mesenchyme creates confusion between an emerging odontoma and lesions like ameloblastic fibroma (AF) and ameloblastic fibro-odontoma, which are considered true

neoplasms thus some ameloblastic fibroma may be diagnosed as odontoma which is the most frequent OT. Clinical and radiographic features may be required to reach a definite diagnosis.

Odontogenic myxoma (OM) is a benign mesenchymal tumor histologically composed of stellate, spindle cells in a plentiful myxoid stroma [1]. The current study reported (n = 3, 5%) of the studied sample ranked three after ameloblastoma and ameloblastic fibroma. The mean age of occurrence was 23.6 years, almost exclusively in females. The mandible was the predominant site (n = 2, 66.6%), with a mandible to maxilla ratio of 2/1. Buchner et al. [17] reported significant female preference with a mandible to maxilla ratio of 2/1 which was in accordance with this study. However, they documented a higher mean age of 36.1 years. Another study reported a higher incidence of OM (9.7%) [3]. In disagreement with this study, Lima-Verde-Osterne et al. [8] reported maxillary predilection (after exclusion of odontoma).

Adenomatoid odontogenic tumor (AOT) is a rare benign neoplasm of epithelial origin presenting histologically a characteristic duct like structure and shows predilection for maxilla. Variable frequencies were recorded for this tumor the highest was in the continent of Asia. AOT was recognized as the fourth or fifth most common OT [27,28]. This study reported one case representing 1.67% of all sample located in the maxilla.

The present study excluded cemento-ossifying fibroma (COF) which was added as mesenchymal odontogenic tumor in WHO 4th edition of head and neck classification of tumor in 2017 to differentiate it from more aggressive form of ossifying fibroma (the juvenile subtype). However, it is discussed in depth along with other ossifying fibromas in the section of fibro-osseous lesions in this edition [2,29].

Papers reported sporadic cases of calcifying epithelial odontogenic tumor (CEOT) representing only slight percentage of the total OTs [30]. Present study registered one case of CEOT representing 1.6% of total cases.

The current study documented 7 malignant OTs, comprising 11.67% of the total investigated sample. The most common malignant OT was ameloblastic fibrosarcoma (n = 3, 42.8%), followed by ameloblastic carcinoma (AC) and

clear cell odontogenic carcinoma (CCOC) (n = 2, 28.5%) for both tumors. Fluctuating frequencies of malignant OT were recorded in other studies as demonstrated in (Table II). However, current study demonstrated high rate of prevalence in comparison to others which may be related to the patients' delaying medical treatment, which might increase the likelihood of transformation of benign into malignant tumor. We reported 3.33% frequency of ameloblastic carcinoma. Lower rates were described in reports from Egypt (1.7%), Brazil (0.64%) and Turkey (0.33%) respectively [2,3,8]. As previously mentioned, the 2017 WHO reclassification of odontogenic keratocyst and inclusion of COF as an odontogenic tumor beside the exclusion of odontoma in this study render the comparison between current study and others more difficult and complicated.

CONCLUSIONS

This study reported comparable and divergent findings with other studies worldwide. Attention should be paid to the high frequency of malignant odontogenic tumors in Iraqi population as this study recorded relative to benign tumors in a rate that appear higher than other parts of the world. However, it should be kept in mind that this study excluded one of most common odontogenic tumors, odontoma, which was excluded in this study for the reasons previously described. In addition 2017 WHO reclassification of tumors changes somewhat the frequency and prevalence of odontogenic tumors globally.

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Author's Contributions

OMG: Conceptualization, Writing – Original Draft Preparation, Writing – Review & Editing.

Conflict of Interest

No conflicts of interest to mention.

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Regulatory Statement

The study was approved by the Ethical Committee of the College of Dentistry at the University of Al-Qadisiyah (REF. 233; Date: 23/5/2022).

Compliance with ethical principles

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