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Original Article

Frequency of Maxillofacial Bone and Cartilage Tumors in Cairo Governorate: A Retrospective Multi-centered Study

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Abstract

Aim: The aim of this study was to identify the prevalence of the primary non-odontogenic maxillofacial bone and cartilage tumors.

Methodology: Clinical data and histopathological diagnoses of primary non-odontogenic maxillofacial bone and cartilage tumors, diagnosed between January 2010 and December 2019, were collected from the histopathological reports of Cairo governorate's educational hospitals and institutions.

Results: Out of 11,444 archival reports -found in the maxillofacial bones and paranasal sinuses- collected from the mentioned study centers, 186 were reported as non-odontogenic maxillofacial bone and cartilage tumors, yielding a 1.63% prevalence. Osteosarcoma was the most common lesion, followed by chondrosarcoma and osteoma, respectively. Females were found to be more prone to the investigated tumors.

Conclusion: The non-odontogenic maxillofacial bone and cartilage tumors represent 1.63% in Cairo governorate. Demographic variations were seen in some of the tumors studied, which differed from the literature; however, osteosarcoma was found to be the most prevalent lesion, accounting for over half of the lesions studied.

Keywords: Prevalence; Epidemiology; Maxillofacial; Bone Tumors; Osteosarcoma; Cairo

Introduction

Epidemiology is the study concerned with the occurrence, determinants and distribution of a disease. It is a scientific discipline that has evolved throughout the years. It was defined in 2018 by the World Health Organization (WHO) as "the study of the distribution and determinants of health-related states or events (including disease), and therefore the application of this study to the control of diseases and other health problems. Various

methods can be used to carry out epidemiological investigations: surveillance and descriptive studies can be used to study distribution; analytical studies to study determinants." Hence, Epidemiology is concerned with studies ranging from surveillance to analytical research **(Bonita et al., 2006; Frérot et al., 2018).**

An example of observational epidemiological study is "Prevalence Study". It is defined as: the number of people in a given population that have a specific disease or attribute at a specified point of time or over a specified period of time. Determining the prevalence and incidence rates of any disease in a given population is usually considered the first step in describing the epidemiology of a disease; in terms of highlighting the impact of the disease. They are also considered to be the base for monitoring the population's general health. Many organizations, including the Center for Disease Control and Prevention (CDC), have stated that there are different types of prevalence present, 'Point Prevalence', 'Period Prevalence' and 'Lifetime/Contact Prevalence' **(Bonita et al., 2006; Ward et al., 2012; Spronk et al., 2019).**

In 2008, National Cancer Registry Program in Egypt (NCRPE) was established through the collaboration of 3 ministries: Ministry of Health, Ministry of Communication and Information Technology and Ministry of Higher Education and Scientific Research. Even though the main aim of NCRPE was to determine the prevalence of different types of cancer, it was faced with many challenges. One of the biggest challenges was designing a sample that could represent a big country such as Egypt, and that could be used as the sole source of cancer prevalence in the country. The sustainability of the program was another challenge faced by NCRPE **(Ibrahim et al., 2014)**.

In spite of the excellence of NCRPE in attempting to cover the cancer cases all over the country and trying to be the main, if not the sole, source of cancer rates in Egypt, the program still had its weak points. One of the major limitations of this program was the lack of recording of the frequencies of many types of cancer occurring in the head and neck area. Although the prevalence rates of various cancer types occurring in different body sites were recorded in the NCRPE, the program did not provide any information regarding the specific cancer types occurring in the head and neck region. Instead, head and neck cancers were stratified according to anatomical site rather than histological cancer type. The cause behind the lack of recording head and neck cancer based on the histological type could not be traced. Examples of such unregistered cancer types include those of odontogenic origin and those affecting maxillofacial bones, as well as many other types **(Ibrahim et al., 2014).**

Maxillofacial bone and cartilage tumors (MF BCT) are a group of non-odontogenic lesions affecting the maxillofacial area (Table 1). MF BCT are usually difficult to diagnose due to their overlapping features **(Kindblom, 2009; Takata & Slootweg, 2017)**.

In addition, their diagnoses and management are somewhat difficult, since their occurrences are reported to be rare in the literature. The light is always shed on odontogenic lesions, because they are more commonly reported than MF BCT in the head and neck area, but this should not mean that MF BCT are of less importance **(Sivapathasundharam et al., 2019).**

In WHO 2017 classification, a group of unrelated lesions were placed under the heading of "Maxillofacial Bone and Cartilage Tumors", claiming that they occur in the same sites or in the interest of differential diagnosis. One of the odd lesions that was found in this classification was the melanotic neuroectodermal tumor of infancy (MNTI), which was not related to the rest of the lesions shown in table (1).

Table (1): WHO 2017 Classification

Benign Maxillofacial Bone and Cartilage Tumors	Malignant Maxillofacial Bone and Cartilage Tumors
Osteoma	Osteosarcoma
Osteoid Osteoma	Chondrosarcoma
Osteoblastoma	Mesenchymal Chondrosarcoma
Chondroma	
Chondroblastoma	
Desmoplastic Fibroma	
Chondromyxoid Fibroma	
Melanotic Neuroectodermal Tumor of Infancy	

If the classification was based on site only, fibrosarcoma, which also occurs in jaw bones, would have been included. However, this was not the case; it was argued that since fibrosarcoma originates from fibroblasts, it cannot be included among the category of "Bone and Cartilage Tumors" **(Sivapathasundharam et al., 2019).** Following this point of view, desmoplastic fibroma, which also originates from fibroblasts, should have no place in this classification. However, as shown in table (1), desmoplastic fibroma was considered as one of the maxillofacial bone and cartilage tumors. To add to the confusion, not all bone- and cartilage-forming tumors were included in the classification. Osteochondroma, for instance, was not found under the heading of "Maxillofacial Bone and Cartilage Tumors".

WHO has introduced a new classification recently in 2022, after the conduction of this study in 2019. Compared to 2017 WHO classification, tumors such as MNTI and osteoid osteoma were excluded from "Maxillofacial Bone and Cartilage Tumors" in 2022. On the other hand, other lesions as osteochondroma and rhabdomyosarcoma with TFCP2 rearrangement were added **(*Vered and Wright,* 2022)**.

To the best of our knowledge, no epidemiological studies have been conducted on MF BCT in Cairo governorate. Therefore, the aim of this study was to assess the frequency of such group of lesions in educational hospitals and institutions in Cairo governorate.

So this study aimed to determine the prevalence of non-odontogenic maxillofacial bone and cartilage tumors diagnosed histopathologically, during the years between 2010 and 2019, in educational hospitals and institutions in Cairo governorate.

Material and Methods

1. Data sources

The clinical data and histopathological diagnoses were retrieved from the patients' histopathological reports. The study included all cases diagnosed histopathologically with any of the maxillofacial benign or malignant bone and cartilage tumors (MF BCT), according to WHO

2017 classification of head and neck tumors, during the period between January 2010 and December 2019. Reports were reviewed from the archives of the following:

- Cairo University, Faculty of Dentistry, Oral and Maxillofacial Pathology Department.
- Cairo University, Faculty of Medicine, General Pathology Department.
- Ain Shams University, Faculty of Dentistry, Oral Pathology Department.
- Al-Azhar University, Faculty of Dentistry, Oral Pathology Department (For Boys).
- Al-Azhar University, Al-Hussein Hospital, General Pathology Department.
- Al-Azhar University, El-Sayed Galal Hospital, General Pathology Department.
- Ahmed Maher Teaching Hospital.
- National Cancer Institute.

2. Participants

Eligibility criteria and selection methods

Patients adhering to the following criteria, were included:

- Maxillofacial non-odontogenic bone and cartilage tumors.
- All age groups.
- Both sexes.
- From the archives of the patient records from 2010-2019.
- Following the histopathological classification of the World Health Organization (2017).

Any of the following cases were excluded:

- Odontogenic neoplasms of the jaw.
- Fibro-osseous lesions of the jaw.
- Metastatic tumors to the jaw.
- Records not within the period of 2010-2019.

3. Study design

Retrospective analysis of the patients records in Cairo governorate's educational hospitals and institutions during the 10 years between 2010 and 2019.

4. Statistical methods

Qualitative data were presented as frequencies and percentages. Quantitative data were presented as mean, standard deviation (SD), median, range and 95% Confidence Interval (95% CI). Chi-square test or Fisher's Exact test was used for comparisons regarding qualitative variables. Age data showed non-normal (non-parametric) distribution; so Mann-Whitney U test was used to compare between ages of patients with intra- and extra-oral non-odontogenic bone and cartilage tumors.

5. Ethical considerations

The study was approved by the Research Ethics Committee of Faculty of Dentistry, Cairo University (no. 15 12 19). In addition, patients' names found in the histopathological reports were kept confidential and were not used in this study.

Results

I. Descriptive statistics

A. Overall prevalence of non-odontogenic maxillofacial bone and cartilage tumors in the ten years between 2010 and 2019

From the archival records of 8 educational hospitals and institutions in Cairo governorate, 186 cases were diagnosed as non-odontogenic maxillofacial bone and cartilage tumors, out of 11,444 lesions found in the maxillofacial bones and paranasal sinuses, in the 10-year time frame, from 2010-2019, as shown in the pie chart in figure (1).

B. Prevalence of each type of non-odontogenic maxillofacial bone and cartilage tumors

The most common type is osteosarcoma (46.2%) followed by chondrosarcoma (19.4%), while chondromyxoid fibroma and melanotic neuroectodermal tumor of infancy (MNTI) showed the lowest prevalence (0.5% for each lesion). Five lesions (2.7%) were diagnosed as non-odontogenic bone and cartilage tumors without specifying the type. The prevalence of each type of non-odontogenic bone and cartilage tumors is shown in (Table 2).

1. Osteosarcoma

The most common non-odontogenic MF BCT is osteosarcoma. 86 (46.2%) cases were histopathologically diagnosed as osteosarcoma, affecting 39 (45.3%) males and 47 (54.7%) females. The age ranged from 7 to 68 years with a mean (±standard deviation) of 36.6 (±15.4) years (Table 3). The most commonly affected site intraorally was found to be the mandible, followed by the maxilla and the least commonly affected site intraorally was found to be the hard palate.

2. Osteoma

The most common benign non-odontogenic MF BCT and the third most common non-odontogenic MF BCT is osteoma. 30 (16.1%) cases were histopathologically diagnosed as osteoma, affecting 18 (60%) males and 12 (40%) females, with a ratio of 3:2 respectively. The age ranged from 6 to 64 years with a mean (±standard deviation) of 32.6 (±15.9) years (Table 4). The most commonly affected site intraorally was found to be the mandible, followed by the maxilla then the hard palate. In addition to the 30 reported cases, one case was found in the tongue and was diagnosed as osteoma mucosae.

C. Comparison for the site of occurrences of non-odontogenic maxillofacial bone and cartilage tumors

1. Intra- and extra- oral sites

Intra-oral sites were more commonly affected, by non-odontogenic MF BCT, than the extra-oral sites. 158 cases were recorded intra-orally, giving a prevalence of 84.9%, while only 21 cases were found extra-orally, representing 11.3% of the studied sample. The site of seven lesions was reported to be non-specific, giving a prevalence of 3.8%.

2. Intra-oral sites

The most common intra-oral site was the mandible (52.5%) while the palate was the least common site (3.2%). Three lesions (1.9%) were diagnosed from the oral cavity without specifying the site.

3. Extra-oral sites

The most common extra-oral site was the nasal cavity (38.1%) while the least common sites were sphenoid sinus, supraorbital and zygomatic arch (4.8% for each site).

D. Association between gender and site of non-odontogenic bone and cartilage tumors

There was no statistically significant association between gender and site of non-odontogenic MF BCT (*P*-value = 0.651, Effect size = 1.025). Females are 1.025 folds prone to non-odontogenic MF BCT than males, as shown in (Table 5 and figure 2).

D. Association between age and site of non-odontogenic bone and cartilage tumors

1. Age categories

There was no statistically significant association between age category and sites of non-odontogenic MF BCT (*P*-value = 0.418, Effect size = 0.218) (Table 6 and figure 3).

2. Age values

There was no statistically significant difference between median age values in patients with intra- and extra-oral non-odontogenic bone and cartilage tumors (*P*-value = 0.073, Effect size = 0.271), as shown in (Table 7 and figure 4).

Discussion

In the current work, histopathological diagnoses were extracted from the archival reports of different histopathological laboratories in educational hospitals and institutions in Cairo governorate from January 2010 to December 2019. Among these educational institutions, some receive biopsies from the maxillofacial region only while others have a broader diagnostic field including bone biopsies from the whole body. In such laboratories, the total cases of maxillofacial lesions from 2010 to 2019 were only collected.

In the present study, 11,444 cases were recorded in the maxillofacial bones and paranasal sinuses from 2010 to 2019. Out of the total lesions, 186 cases were diagnosed as MF BCT, representing a prevalence of 1.63%. This came in agreement with the WHO 2017 classification of head and neck tumors, stating the rarity of these group of lesions **(Takata & Slootweg, 2017)**. **Wang et al., 2012**, **Singh & Solomon, 2017**, **Moreau et al., 2018**, and **Cleven et al., 2020** are examples of many researchers who have also disclosed the rarity of such lesions.

In addition, it was found that 124 cases were diagnosed as malignant MF BCT, out of the total 186 cases, giving a prevalence of 66.7%. This was in line with **George & Mani, 2011**, and **de Souza et al., 2019**. These authors haven't studied the prevalence of the malignant MF BCT collectively, but have reported the high prevalence of osteosarcoma, and chondrosarcoma, respectively, among MF BCT.

Out of the rest of the 186 cases, 57 were diagnosed as benign MF BCT, giving a prevalence of 30.6% but unfortunately, this finding couldn't be opposed to any study in the literature. This is due to the lack of actual prevalence records of the benign MF BCT. The rest of the lesions (5 cases) were diagnosed as bone or cartilaginous tumors, NOS (not otherwise specified), giving a prevalence of 2.7%. The reason behind the higher prevalence of malignant MF BCT is not fully understood. It could be because malignant tumors develop symptoms like pain, swelling, or non-specific symptoms or it could be because the benign tumors are asymptomatic.

This study showed that osteosarcoma was found to be the most common non-odontogenic MF BCT, representing 46.2%. This is similar to what was stated by the WHO 2017 classification of head and neck tumors. In addition to many other studies conducted in different countries, such as India by **George & Mani, 2011**, Nigeria by **Ibikunle et al., 2018** and Vienna by **Eder-Czembirek et al., 2019,** all these authors concluded that osteosarcoma was the most common maxillofacial bone tumor.

Regarding the site of occurrence, non-odontogenic MF BCT's most commonly affected intra-oral site was the mandible. Almost half of the lesions (52.5%) occurred in the mandible, followed by the maxilla. Concerning the malignant non-odontogenic MF BCT, specifically, there was no significant difference between the maxilla and the mandible (i.e. almost the same number of lesions were found in both). As for the extra-oral sites, the nasal cavity was found to be the most prevalent site for the non-odontogenic MF BCT, while the zygomatic arch was the least affected site.

Table (2): Frequencies (n) and percentages (%) of main types of non-odontogenic MF BCT (n = 186)

Type of non-odontogenic bone and cartilage tumors	n	%
Osteosarcoma	86	46.2
Chondrosarcoma	36	19.4
Osteoma	30	16.1
Osteoblastoma	12	6.5
Desmoplastic fibroma	9	4.8
Osteoid Osteoma	4	2.2
Mesenchymal Chondrosarcoma	2	1.1
Chondromyxoid fibroma	1	0.5
MNTI	1	0.5
Non-specified types	5	2.7

Table (3): Frequencies (n) and percentages (%) of osteosarcoma and male and female distribution in each age category.

Age category	Male		Female		Total	
	n	%	n	%	n	%
<10 y	0	0	2	2.3	2	2.3
11 – 20 y	6	6.9	3	3.5	9	10.5
21 – 30 y	6	6.9	19	22.1	25	29.1
31 – 40 y	11	12.8	8	9.3	19	22.1
41 – 50 y	10	11.6	4	4.7	14	16.3
51 – 60 y	5	5.8	2	2.3	7	8.1
61 – 70 y	1	1.2	9	10.5	10	11.6

As for the age range of non-odontogenic MF BCT, it was found to range from 0.6 to 83 years, with a median of 34.5 years for the intra-oral lesions, while the extra-oral lesions ranged from 13 to 77 years, with a median of 41 years. Most of the intra- and extra-oral lesions were most commonly seen within the age range of 21 to 40 years.

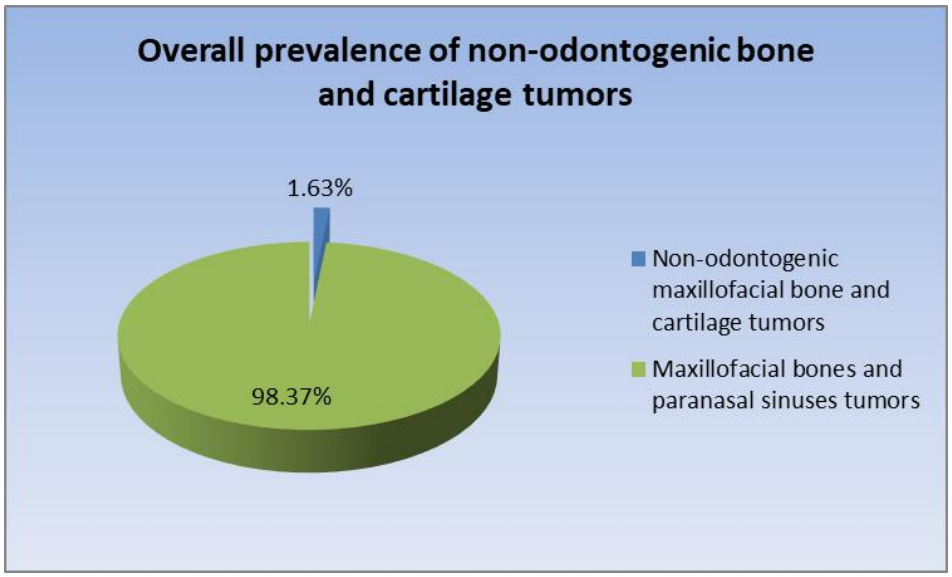

Figure (1): Pie chart representing overall prevalence of non-odontogenic MF

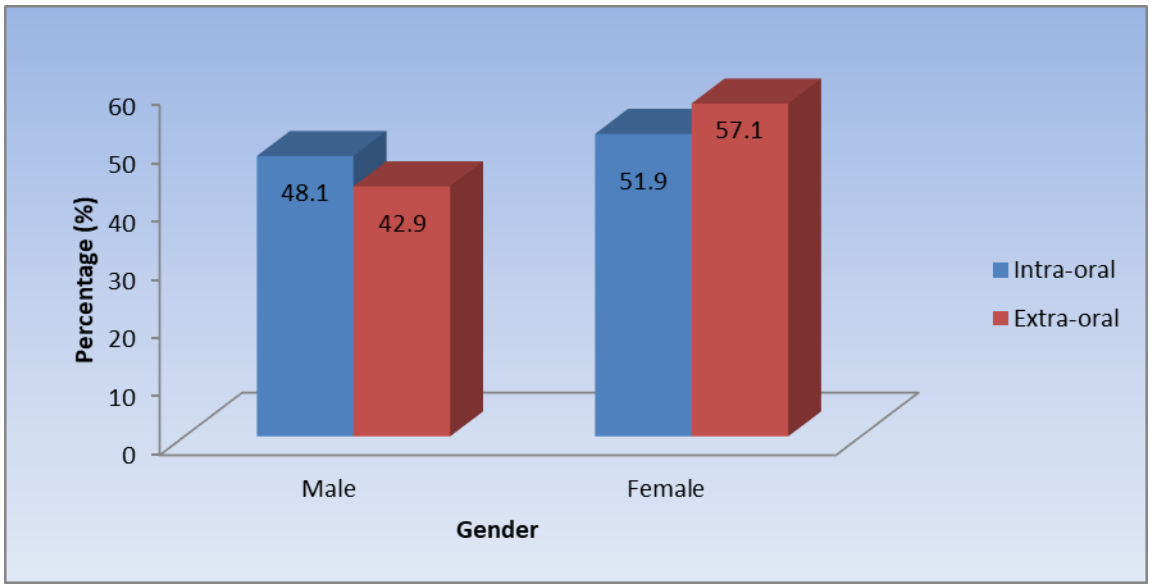

Figure (2): Bar chart representing the association between gender and sites of non-odontogenic MF BCT

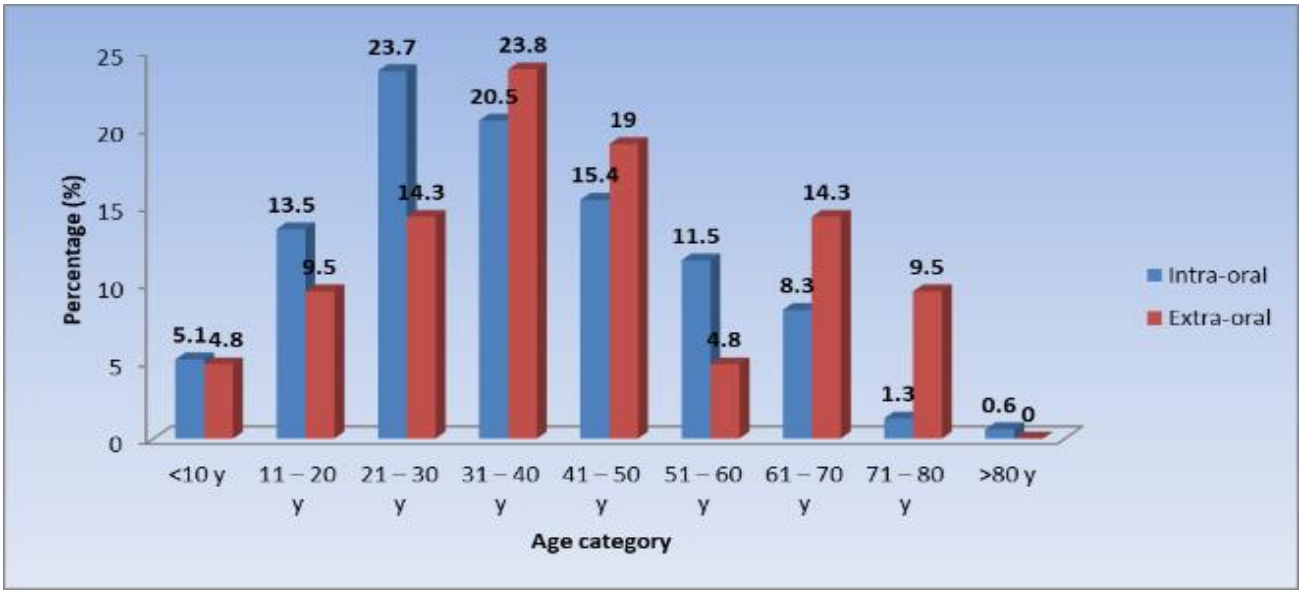

Figure (3): Bar chart representing the association between age categories and sites of non-odontogenic MF BCT

Table (4): Frequencies (n) and percentages (%) of osteoma and male and female distribution in each age category (*: One case was excluded, because age was not specified).

Age category	Male		Female		Total (n=29)*	
	n	%	n	%	n	%
<10 y	1	3.4	1	3.4	2	6.9
11 – 20 y	5	17.2	1	3.4	6	20.7
21 – 30 y	3	10.3	3	10.3	6	20.7
31 – 40 y	3	10.3	2	6.9	5	17.2
41 – 50 y	5	17.2	2	6.9	7	24.1
51 – 60 y	0	0	1	3.4	1	3.4
61 – 70 y	0	0	2	6.9	2	6.9

Table (5): Descriptive statistics and results of Chi-square test for the association between gender and site of non-odontogenic MF BCT

Gender	Intra-oral (n = 158)		Extra-oral (n = 21)		*P*-value	*Effect size (OR)*
	n	%	n	%		
Male	76	48.1	9	42.9	0.651	1.025
Female	82	51.9	12	57.1		

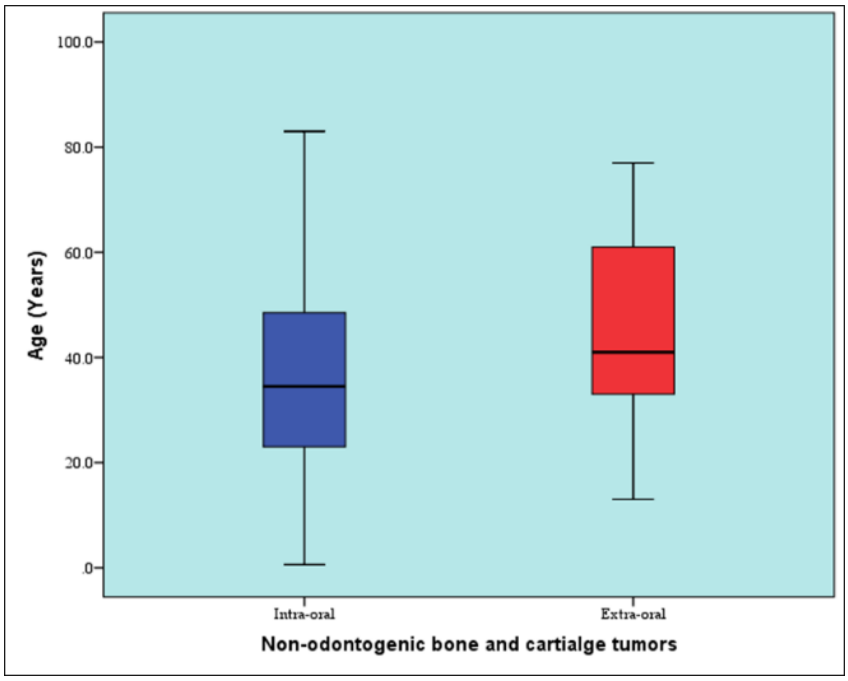

Figure (4): Box plot representing median and range values for ages of patients with different sites of non-odontogenic MF BCT

Table (6): Descriptive statistics and results of Fisher's Exact test for the association between age categories and sites of non-odontogenic MF BCT

Age category	Intra-oral (n = 156) †		Extra-oral (n = 21)		*P*-value	*Effect size (v)*
	n	%	n	%		
<10 y	8	5.1	1	4.8		
11 – 20 y	21	13.5	2	9.5		
21 – 30 y	37	23.7	3	14.3		
31 – 40 y	32	20.5	5	23.8		
41 – 50 y	24	15.4	4	19	0.418	0.218
51 – 60 y	18	11.5	1	4.8		
61 – 70 y	13	8.3	3	14.3		
71 – 80 y	2	1.3	2	9.5		
>80 y	1	0.6	0	0		

†: Two cases were excluded because age was not specified

Table (7): Descriptive statistics and results of Mann-Whitney U test for comparison between age values in patients with intra- and extra-oral non-odontogenic MF BCT (*: Significant at $P \leq 0.05$, †: Two cases were excluded because age was not specified).

Intra-oral (n = 156) †		Extra-oral (n = 21)		*P*-value	*Effect size (d)*
Mean (SD)	Median (Range)	Mean (SD)	Median (Range)		
36.2 (17.3)	34.5 (0.6-83)	43.8 (18.4)	41 (13-77)	0.073	0.271

This study showed that there were no statistically significant differences between the median age values in patients with intra- and extra-oral non-odontogenic MF BCT**.** These findings, however, could not be compared elsewhere due to the lack of a similar collective study.

In the present study, females were more prone to MF BCT than males. They were also more affected by osteosarcomas than males, which was inconsistent with the studies conducted by **Ibikunle et al., 2018, and Eder-Czembirek et al., 2019,** where they all recorded that the lesions were more commonly seen in males. On the other hand, osteomas were more frequent in males than in females, which was consistent with **Larrea-Oyarbide et al., 2008**, but in contrast with **Boffano et al., 2012**, where they reported that the females were higher than the males.

Knowing the prevalence of different lesions is an eye-opener. However, many developing countries and their governmental hospitals and institutions, including Egypt, need to work on archiving their records and attempting to standardize the formats of the histopathological reports. That could either be done by standardizing the database used, or by unifying the electronic system through which the histopathological reports are saved. In addition, NCRPE needs to add the odontogenic and non-odontogenic tumors, and all hospitals need to report the prevalence of each tumour periodically to NCRPE, to have a constant update.

It is also recommended for governorates, other than Cairo, to conduct the same study to be able to conclude the overall prevalence of occurrence of MF BCT in Egypt. And since the classification of the WHO has been recently updated, after the conduction of such study, the overall prevalence of maxillofacial bone and cartilage tumors would be slightly affected. For instance, since MNTI and osteoid osteoma have been removed from the new WHO classification, excluding them from the above studied lesions would cause a decrease in the overall prevalence in Cairo governorate, from 1.63% to 1.58%.

Conflict of Interest:

The authors declare no conflict of interest.

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