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Odontogenic Tumors in Children and Adolescents: A Retrospective Analysis at a Tertiary-Level Health Care Facility in Bangladesh

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ABSTRACT:

Introduction: This study presents the oral and maxillofacial pathological features of several types of odontogenic tumors in children and adolescents less than 18 years of age from a Bangladeshi tertiary-level healthcare facility. **Materials and Methods:** In this retrospective cross-sectional study, the World Health Organization (WHO) categorization for head and neck tumors in 2022 was used to assess the frequency of odontogenic tumors in children and adolescents. Data relating to sex, age, and tumor site were retrieved and presented. The Chi-square test was implemented to study the association between gender, age, and site with different types of odontogenic tumors. **Results:** A total of 106 cases were found, and all the cases were benign. Ameloblatoma (75.47%) was the most common tumor, followed by odontogenic myxoma (9.44%), cemento-ossifying fibroma (5.66%), and odontoma (4.72%). A male (60.38%) predominance was observed in the gender distribution, but no statistically noteworthy (P = 0.43) relationship was found. Most of the cases were found in the 13–18 age group (63.21%), with a mean age (standard deviation) of 13.01 (2.29) years. There was no statistical significance found between age group and tumor type (P = 0.78). The mandibular (95.28%) predominance was found over the maxilla (4.72%) (Mandible: Maxilla = 20.2:1) and was statistically significant (P= 0.002). In the mandible, the posterior region (88.68%) was mostly affected. **Conclusions:** Early detection of odontogenic tumors is rare, and as people aged, they become more common. Ameloblatoma was the most prevalent odontogenic tumor in the jaws, followed by odontogenic myxoma, cemento-ossifying fibroma, and odontoma. Adolescents experience odontogenic tumors more frequently than children do. The mandible was observed to predominate over the maxilla in terms of site distribution. The mandible's posterior portion was primarily affected.

Keywords: Adolescent, ameloblastoma, children, odontogenic tumors, retrospective study

INTRODUCTION:

Odontogenic tumors are a heterogeneous collection of diseases that originate from the cells that develop teeth or their remnants. They can originate from epithelial, mesenchymal (ectomesenchymal), or mixed components and have a variety of clinical and histopathologic characteristics. [1] In 1971, the World Health Organization (WHO) released the first odontogenic tumor classification that was universally recognized. [2] Then it was reviewed twice, in 1992 [3] and 2005. [4] The fourth version of the WHO categorization of odontogenic tumors was released in 2017. [5] This revised categorization follows a more straightforward type from the development of the germ cell layer, which includes mixed, mesenchymal (ectomesenchymal), and epithelial odontogenic tumors. Similar to past editions, this one primarily classified odontogenic tumors as either benign or malignant, depending on their biologic nature. Odontogenic keratocysts and calcifying odontogenic cysts, respectively, have replaced the terms keratocystic odontogenic tumor (KCOT) and calcifying cystic odontogenic tumor (CCOT). This categorization also included the primordial odontogenic tumor, cement-ossifying fibroma, and sclerosing odontogenic carcinoma. [5] In March 2022, the updated 5th version of the WHO Classification of Head and Neck Tumors became accessible online in a handy

electronic form for the first time. The current edition is basically identical to the earlier categorization of odontogenic lesions. The only recently described odontogenic tumor is adenoid ameloblastoma, which is classified as a benign epithelial odontogenic tumor. [6]

Except for a few studies that utilized the 2017 classification [7–9], the majority of papers reported after 2005 employed the 2005 WHO categorization. [10–16] A publication was reported that used the WHO categorization of head and neck tumors from 2022 to report benign pediatric jaw lesions in 2023. [17] There are not enough studies that focus on the occurrence of odontogenic tumors in children and adolescents in Bangladesh. This study is intended to explore the incidence of various odontogenic tumor types in children and adolescents in light of the 2022 change to the WHO classification for head and neck tumors.

MATERIALS AND METHODS:

In this retrospective cross-sectional study, the latest WHO classification for head and neck tumors in 2022 was used to evaluate the prevalence of odontogenic tumors in children and adolescents. This study was developed relying on the medical data from the Department of Oral and Maxillofacial Surgery at Dhaka Dental College and Hospital from January 2011 to June 2018. Since this was a retrospective analysis and all information was acquired from the departmental record books, no ethical clearance was required.

Patients who had odontogenic tumors reported by histological analysis and were under the age of 18 were included. Patients with incomplete data were excluded from the study. Medical record data were searched for information on each patient's age, gender, and anatomical location of the lesion. In the hospital documents, 106 cases of odontogenic tumors in patients under the age of 18 were identified. The patients were separated into three different categories for analysis based on age: 0–6 years, 7–12 years, and 13–18 years. The anterior and posterior anatomic regions of the maxilla and mandible were separated based on site prevalence. The anterior region was demarcated from the canine to the canine tooth of either jaw. The posterior region was defined in the maxilla from the first premolar or deciduous molar to the tuberosity and from the first premolar or deciduous molar to the third molar tooth, including the angle-ramus area in the mandible.

The SPSS program (version 23; SPSS, Inc., Chicago, IL) was the software employed for the statistical analysis. The prevalence and distribution of each of the odontogenic tumors as per distinct age categories, sexes, and sites were presented by percentage in separate tables. The Chi-square test was implemented to study the association between sex, age, and site with different types of odontogenic tumors. The threshold for statistical significance was set at P <0.05.

RESULTS:

We identified 106 cases in total with odontogenic tumors below 18 years of age recorded in the hospital documents. All the cases were benign. No malignant case was found in the records. Table 1 demonstrates the frequency of various odontogenic tumors. Ameloblatoma was the most common tumor, constituting about 75% of the patients, followed by odontogenic myxoma (9.44%), cemento-ossifying fibroma (5.66%) and odontoma (4.72%). The male (60.38%) predominance was observed in the gender distribution with a ratio of 1.52:1. However, there was no evidence of a significantly noteworthy relationship between tumor types and gender (P = 0.43).

Table 1. Prevalence of odontogenic tumors and demographic data by histological categorization

Odontogenic Tumour	Frequency	Percentage (%)	Gender			*P-value* (*P*<0.05) *
			Male (%)	Female (%)	Male :Female ratio	
Benign, 106 cases, 100%						
Odontogenic epithelium origin						
AME	80	75.47%	49 (46.22)	31 (29.25)	1.58:1	
CEOT	1	0.94%	1(0.94)	0	NA	
SOT	1	0.94%	0	1(0.94)	NA	
AOT	2	1.89%	0	2(1.89)	NA	
Odontogenic epithelium mixed with odontogenic ectomesenchyme origin						0.43#
AF	1	0.94%	1 (0.94)	0	NA	
OD	5	4.72%	4 (3.77)	1 (0.94)	4:1	
Mesenchyme and/or odontogenic ectomesenchyme origin						
OM	10	9.44%	6 (5.67)	4 (3.77)	1.5:1	
COF	6	5.66%	3 (2.83)	3 (2.83)	1:1	
No malignant case was found						
Total	106	100%	64 (60.38)	42 (39.62)	1.52:1	

AF, Ameloblastic fibroma; AME, Ameloblastoma; AOT, Adenomatoid odontogenic tumor; CEOT, Calcifying epithelial odontogenic tumor; COF, Cemento-ossifying-fibroma; NA, Not applicable; OD, Odontoma; OM, Odontogenic myxoma/myxofibroma; SOT, Squamous odontogenic tumor.
#, Statistically not significant; *, Statistically significant.

Most of the cases were found in the 13–18 age group (63.21%), followed by the 7–12 age group (31.13%). The age group 0–6 had the fewest patients (5.66%). The mean age (standard deviation, SD) of prevalence was 13.01 (2.29) years, with a range of 4–18 years. There was no statistical significance found between age group and tumor type (P = 0.78). Table 2 presents the age distribution of odontogenic tumors.

Table 2. The age distribution of the patient with odontogenic tumors

Tumour	Total cases	Age (Years)					*P*- value (*P*<0.05)*
		0-6	7-12	13-18	Mean ± SD	Range (years)	
AME	80	6	24	50	13.8 ± 3.67	4-18	0.78#
CEOT	1	0	0	1	NA	15	
SOT	1	0	1	0	NA	11	
AOT	2	0	0	2	16 ± 2	14-18	
AF	1	0	1	0	NA	7	
OD	5	0	2	3	14.2 ± 3.19	10-18	
OM	10	0	2	8	14.2 ± 2.36	10-18	
COF	6	0	3	3	13.67± 2.98	9-18	
Total	106	6 (5.66%)	33 (31.13%)	67 (63.21%)	13.01± 2.29	4-18	

AF, Ameloblastic fibroma; AME, Ameloblastoma; AOT, Adenomatoid odontogenic tumor; CEOT, Calcifying epithelial odontogenic tumor; COF, Cemento-ossifying-fibroma; NA, Not applicable; OD, Odontoma; OM, Odontogenic myxoma/myxofibroma; SD, Standard deviation; SOT, Squamous odontogenic tumor.
#, Statistically not significant; *, Statistically significant.

The mandibular (95.28%) predominance was found over maxilla (4.72%) (Mandible: Maxilla = 20.2:1) and was statistically significant (*P*= 0.002). In the mandible, the posterior region (88.68%) was mostly affected. Data regarding the anatomic site allocation of the tumors are given in Table 3.

Table 3. Site distribution of odontogenic tumors

Tumour	Maxilla@			Mandible@			Maxilla: Mandible ratio	*P*- value (*P*<0.05)*
	A	P	Total	A	P	Total (%)		
AME	1 (0.94%)	2(1.89%)	3 (2.83%)	5 (4.72%)	72 (67.92%)	77 (72.64%)	1: 25.67	0.002*
CEOT	0	0	0	0	1(0.94%)	1 (0.94%)	NA	
SOT	0	1 (0.94%)	1 (0.94%)	0	0	0	NA	
AOT	0	0	0	1(0.94%)	1 (0.94%)	2 (1.89%)	NA	
AF	0	0	0	0	1 (0.94%)	1 (0.94%)	NA	
OD	0	1 (0.94%)	1(0.94%)	0	4 (3.77%)	4 (3.77%)	1:4	
OM	0	0	0	0	10 (9.43%)	10 (9.43%)	NA	
COF	0	0	0	1(0.94%)	5 (4.72%)	6 (5.66%)	NA	
Total (%)	1 (0.94%)	4 (3.78%)	5 (4.72%)	7 (6.6%)	94 (88.68%)	101 (95.28%)	1: 20.2	

A, Anterior; AF, Ameloblastic fibroma; AME, Ameloblastoma; AOT, Adenomatoid odontogenic tumor; CEOT, Calcifying epithelial odontogenic tumor; COF, Cemento-ossifying-fibroma; NA, Not applicable; OD, Odontoma; OM, Odontogenic myxoma/myxofibroma; P, Posterior; SOT, Squamous odontogenic tumor.
#, Statistically not significant; *, Statistically significant.
@A, Anterior (canine to canine teeth in either jaw); P, posterior (first premolar/deciduous molar tooth to tuberosity in maxilla, first premolar/deciduous molar tooth to third molar tooth including angle-ramus area in the mandible).

DISCUSSION:

Odontogenic tumors in children and adolescents are uncommon lesions, and the outcomes of this study are consistent with earlier reports. [10, 11] The findings of this current study supported the previously published literature by demonstrating a prevalence of benign tumors. [10, 11] Our analysis did not reveal any malignant tumors, which is consistent with a report that was previously published. [11] Malignant odontogenic tumors are less common in children and adolescents than has been documented. [8, 10]

In this study, the mean age of the patients was 13 years. The 13–18 age group was the most common, followed by the 7–12 age group. It has been hypothesized that the incidence of odontogenic tumors rises with advancing age. The majority of studies have found that odontogenic tumors are more common in adolescents than in childhood. [8, 10, 11, 18, 19] This may be explained by the fact that asymptomatic slow tumor growth delays diagnosis and may be related to increased densities of epithelial remnants during odontogenesis with advancing age. [10]

Regarding the gender distribution, we found a higher frequency of males than females. Chen *et al.* [7] reported a slight male predominance, accounting for 52.8% in their study titled "Benign Pediatric Jaw Bone Lesions." However, most of the authors reported an equal or similar distribution by gender.[9-12, 18, 19]

The incidence and frequency of each form of odontogenic tumor have been reported to differ. Our findings on the frequency of ameloblastoma are consistent with those reported by various authors. [12, 18, 19] The frequency of ameloblastoma was found to range between 49 and 54% in the previous studies, where it was reported as the most common tumor. [18, 19] But our findings demonstrate a higher frequency of ameloblastoma, accounting for approximately 75%. In contrast, in the studies they conducted, certain scholars stated that the odontoma was the most typicl odontogenic tumor, accounting for a range between 41% and 61%. [7, 9–11, 14] In the present study, odontomas were found to be the fourth most common tumor, accounting for around 5%. While odontoma was the most commonly reported odontogenic tumor in America and Europe, ameloblastoma appears to be more prevalent in Asian and African nations. [7-19]

With a prevalence of 9.44%, odontogenic myxoma was the second most prevalent odontogenic tumor in the study we conducted. The documented prevalence of odontogenic myxoma in children differs from 1.2% to 39%. [10, 11, 17–19] Lawal *et al.* and Adebayo *et al*. reported myxoma as the second most frequent odontogenic tumor in Nigerian children and adolescents. [16] Crasnean *et al.* conducted a pilot study in Romanian children and reported myxoma as the second most common, accounting for around 29% of odontogenic tumors. [17] Ulmansky *et al.* found that odontogenic myxoma was the most frequent tumor in Israeli children, with 38.8% prevalence. [20]

Cemento-ossifying fibroma has historically been classified as both an odontogenic neoplasm and a benign fibro-osseous lesion, but it has now been revised and classified under the odontogenic tumor category. [5, 6] In the present study, Cemento-ossifying fibroma was the third most prevalent tumor. There are no studies reporting cemento-ossifying fibroma in children and adolescents. Cemento-ossifying fibroma was found to be the second most common tumor in a retrospective study carried out by Al-aroomy *et al.* in the Egyptian population to determine the incidence of odontogenic tumors. [21] A low frequency of cemento-ossifying fibromain was reported by Mascitti *et al.* in an epidemiology study conducted in Italy. [22] For the first time, cemento-ossifying fibroma was categorized under mesenchymal odontogenic tumors in the 2017 classification, although it was also extensively covered alongside the other ossifying fibromas in the part that focused on fibro-osseous lesions. [6]

With a mandibular-to-maxillary ratio of 20.2:1, the mandible was the site with the highest involvement. These results were in agreement with a number of previously published studies; however, the ratio was in disagreement with their conclusions. [7, 9, 17–19] In the current study, the posterior mandibular region was likewise the most severely affected area. The results of our study agreed with those of numerous other studies. [10, 18, 19] The anterior mandibular area came next. Seeveto *et al.* reported the anterior maxilla as the second most prevalent site for odontogenic tumors in children and adolescents. [10] Additionally, tumor type and location of tumor showed a statistically meaningful relationship.

CONCLUSIONS:

Most odontogenic tumors that appear in the jaws are benign, with ameloblatoma being the most common, followed by odontogenic myxoma, cemento-ossifying fibroma, and odontoma. Odontogenic tumors are more common in adolescents than in childhood**.** Regarding the site distribution, the mandibular predominance was found over the maxilla. In the mandible, the posterior region was mostly affected. Considering that our database includes instances that were referred to our hospital for histopathologic evaluation from various parts of the country, the findings acquired in the current study can be regarded as reflecting the children and adolescents in our population. It may also be used to make a probable diagnosis before taking a biopsy. The keratocystic odontogenic tumor was excluded from the 4th edition of the odontogenic tumor classification by the WHO in 2017. The 5th edition in 2022 is basically identical to the earlier categorization of odontogenic lesions. This is evident in the present study. In terms of the relative incidence of odontogenic tumors, we found some differences. The 5th edition of WHO classification and predetermined age specifications should serve as the foundation for further research to facilitate comparisons between various global populations.

CONFLICT OF INTEREST: The authors declare no conflict of interest.

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DATA AVAILABILITY STATEMENT: The data presented in this study are available on reasonable request from the corresponding author.

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