

## Odontogenic Diseases And Disorders Of Infectious Nature In Relation To Paediatric Population– A systematic review

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### ABSTRACT

**Background:** Most Odontogenic tumors that affect children and adolescents are benign and constitute about 1% to as high as 28% of all oral lesions in children. It is important to know that the reported frequency of odontogenic tumors in the pediatric age group may likely be related to ethnic or geographic consideration of the population studied. This systematic review presents the detailed data about the occurrence of pediatric odontogenic tumors, the predominant type of tumor, most common sites, gender predominance and so on.

**Materials and Methods:** A systematic literature review and retrospective studies of Pediatric odontogenic tumors published in two electronic databases was conducted based on the guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) Statement.

**Results:** The predominant odontogenic tumor that was reported is Odontoma which accounted to 641 cases which was 52% of the total cases reported under the age of 20. Following Odontoma, it was Ameloblastoma which was 36% with 448 cases reported. Next in line were Keratocystic odontogenic tumors (147) and Adenomatoid Odontogenic tumor (116). The other odontogenic tumors that were encountered in the studies are Cementoblastoma (15), Ameloblastic fibroodontoma (48), Ameloblastic fibroma (56), Odontogenic myxoma (96), Odontogenic fibroma (75) and Calcifying epithelial odontogenic tumor (19).

**Conclusion:** The result of the study was in line with the previous literature reviews suggesting that even though Pediatric odontogenic tumors are uncommon and usually present as benign tumors, the malignant variants do occur and the detailed information regarding it is necessary for the better understanding of the tumor in regard to treatment.

**Keywords:** paediatric, oral, odontogenic, pathology, dental

### 1. INTRODUCTION

Odontogenic tumors (OTs) are a heterogeneous group of lesions originating from odontogenic apparatus and their remnants.<sup>[1]</sup> These tumors arise from the tissues that form our teeth and are derived from epithelial, ectomesenchymal and/or mesenchymal components and constitute less than 5% of all tumors.<sup>[1]</sup> Odontogenic lesions present diverse clinical behavior and histopathology which ranges from simple hamartomas to life-threatening malignancy. The OTs are commonly located within bone (intra-osseous) but there are few that occur in the soft tissue overlying tooth-bearing areas (peripheral/extra-osseous).<sup>[2]</sup> The fourth edition of World Health Organization (2017) classification, odontogenic tumors were classified as

epithelial odontogenic, mesenchymal (ectomesenchymal), mixed odontogenic and malignant odontogenic tumors (either carcinomas or sarcomas).<sup>[3,4]</sup> Odontogenic tumours are slow growing and vary in size from a small to a large swelling, causing cortical bone perforation with the displacement of the adjoining normal anatomic structures.<sup>[5]</sup> Tumors of the pediatric maxillofacial skeleton are uncommon with a broad differential diagnosis.<sup>[6]</sup> Most of the intra osseous tumors are misinterpreted as infection at the early stage and they are treated with antibiotics. Subsequently, due to an unresponsive antibiotic therapy the intraoral radiographs are taken to analyze the radiolucency or radio dense lesion in the jaws. So, finally a tissue biopsy becomes necessary in order to diagnose and initiate proper therapy.<sup>[7, 8]</sup> Most Odontogenic tumors that affect children and adolescents are benign and constitute about 1% to as high as 28% of all oral lesions in children. It is important to know that the reported frequency of odontogenic tumors in the pediatric age group may likely be related to ethnic or geographic consideration of the population studied.<sup>[9]</sup> This systematic review presents the detailed data about the occurrence of pediatric odontogenic tumors, the predominant type of tumor, most common sites, gender predominance and so on.

## 2. MATERIALS AND METHODS

A systematic literature review and retrospective studies of Pediatric odontogenic tumors published in two electronic databases was conducted based on the guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) Statement. An electronic search was conducted in PubMed, Google scholar and Scopus. The search strategy used in electronic base was Pediatric Odontogenic Tumors OR Odontogenic Tumors of Jaw OR Retrospective study of pediatric Odontogenic Tumors OR Benign Oral Lesions in Pediatric Population. The articles that were published between 2004-2021 were selected. The reference lists of the articles included and of the relevant reviews on the issue were also screened for additional studies that might have been missed in the electronic searches.

### Eligibility criteria

Inclusion criteria were articles reporting cases and studies on Pediatric Odontogenic Tumors. The studies need to have the complete details regarding the occurrence of the odontogenic tumors, the age group, the most common site and type of odontogenic tumors in pediatric population. Retrospective studies, reviews, and case reports were also searched. Articles need to be in English. Exclusion criteria were studies reporting randomized controlled trials and cohort study.

### Study selection

The titles/abstracts of all references retrieved through the electronic searches were read independently. The study was included if the title/abstract met the eligibility criteria. The full texts of the articles with titles/abstracts were obtained to get the appropriate information. Following the assessment of full text, the reference list was also screened in order to check for the eligibility criteria.

### Data Extraction

For each study included, the following data were extracted on a standard form: author names and year of publication and country where the study was conducted, the age groups, number of odontogenic tumors encountered in the study, the anatomical location (maxilla/mandible) and the predominant type of odontogenic tumor seen in that specific population.

### Data analysis

Data analysis was performed using the Statistical Package for the Social Sciences (SPSS) software, version 23.0 (SPSS Inc., Chicago, IL, USA).

## 3. RESULTS

Author	Year	Population	Age	Odontogenic tumors	Predominant site	Predominant tumor
Tekkesin et al	2016	Turkey	0-17	149	Mandible	Odontoma, Ameloblastoma, Myxoma.
Ajayi et al	2004	Nigeria	0-19	92	Mandible	Ameloblastoma, AOT, Myxoma

Guerrisietal	2007	Argentina	0-20	153	Mandible	Odontoma,Ameloblastoma,Myxoma
Perryetal	2015	Baltimore	0-16	20	Mandible	Ameloblastoma
Leietal	2014	Southern Taiwan	0-15	158	Mandible	Odontoma, Ameloblastoma
Mortellaroetal	2008	Italy	1-15	86	Mandible	Odontoma,Odontogenic fibroma,Ameloblastoma
Silveiraetal	2008	Brazil	0-14	79	-	Odontoma Keratocystic odontogenic tumors
Tanrikuluetal	2004	Turkey	0-15	21	Mandible	Odontoma, Ameloblastic fibroma
MartinsFilhoetal	2015	Northeastern Brazil	0-18	66	Maxilla	Odontoma,Keratocystic
						odontogenic tumor
Wang et al	2009	Taiwan	0-14	178	Maxilla	Odontoma, Ameloblastoma
Yamani et al	2011	Saudi Arabia	1-18	15	-	Odontoma,Ameloblastoma
Prosdócimo et al	2018	Brazil	0-19	154	-	Odontoma,Ameloblastoma,Odontogenic fibroma
Chen et al	2020	Massachusetts USA.	0-18	36	-	Odontoma
Silva et al	2016	Brazil	0-19	94	Mandible	Ameloblastoma, Keratocystic odontogenic

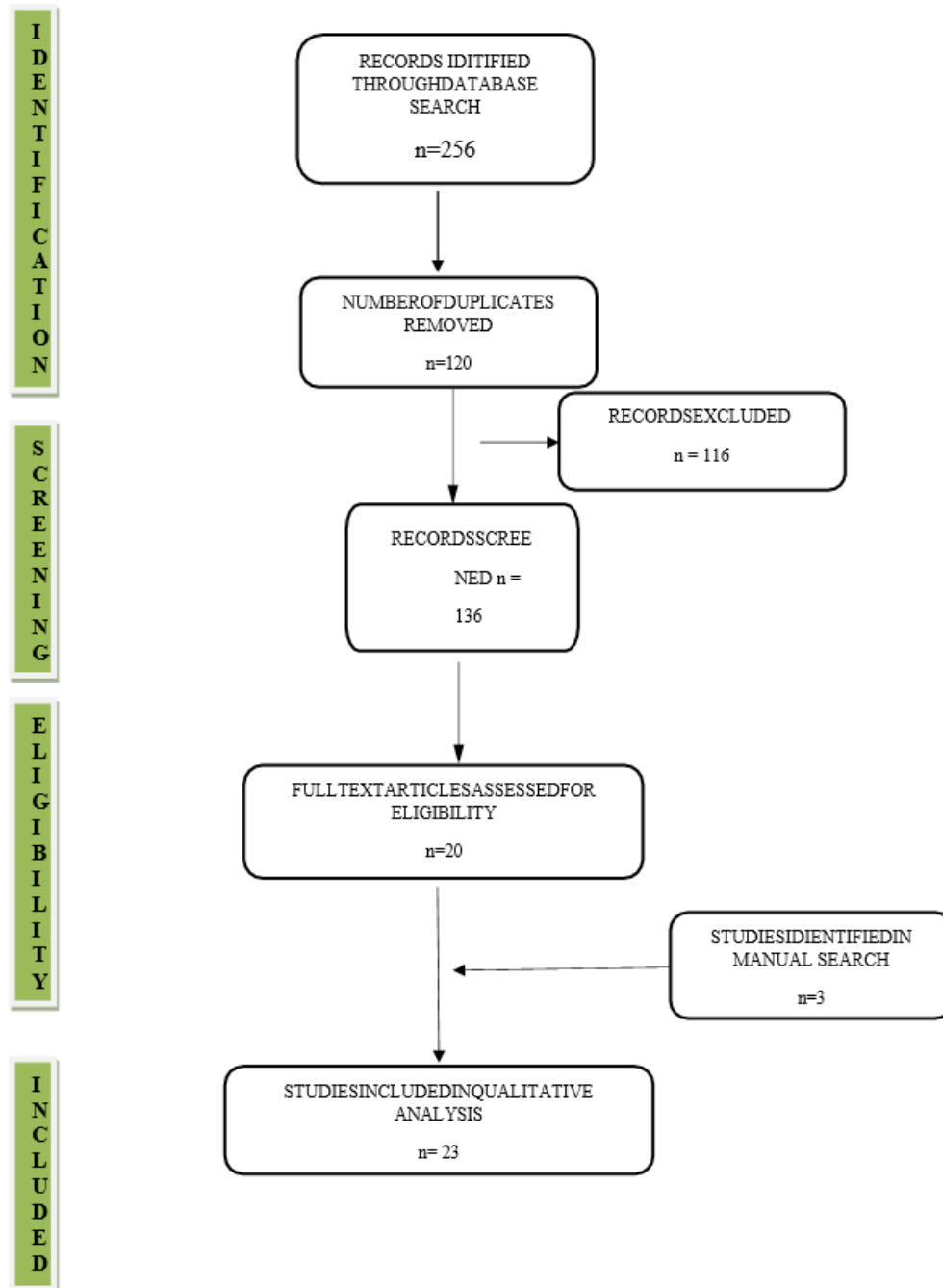
						tumors
Kebede et al	2017	Ethiopia	0-19	34	Mandible	KCOT,Amelobl astoma.
Costa et al	2012	Brazil	0-19	48	Mandible	Odontoma,Kerat ocystic odontogenic tumor
Bhagwat et al	2017	India	0-19	-	Maxilla	Ameloblastoma, Odontome
Al Sheddi et al	2015	Saudi Arabia	0-20	63	Mandible	Keratocystic odontogenic tumor,Amelobla stoma
Mamabolo et al	2011	South Africa	0-20	244	Mandible	Ameloblastoma,
						Keratocystic odontogenic tumors
Lawal et al	2013	Nigeria	0-19	48	Mandible	Ameloblastoma, Fibromyoma
Tandon et al	2020	India	0-15	26	Mandibular posterior	Ameloblastoma, Adenomatoid odontogenic tumor.
Deepthi et al	2016	India	0-19	70	Mandibular posterior	Odontoma,Amel oblastoma
Bhat et al	2019	India	1-20	14	Mandible	Unicystic ameloblastoma, Odontoma

#### **NUMBER AND TYPES OF ODONTOGENIC TUMORS ENCOUNTERED IN THE STUDIES**

AUTHOR	YEAR	Ameloblastoma	AOT	Odontoma	KC OT	CB	AFO	AF	OM	OF	CEOT
Tekkesinetal	2016	31	4	60	-	4	5	7	24	-	-

Ajayietal	2004	45	18	4	-	-	-	5	8	7	0
Guerrisietal	2007	28	8	78	-	3	7	3	13	5	2
Perryetal	2015	5	-	0	-	-	1	0	-	0	-
Leietal	2014	14	2	108	-	1	1	4	5	10	-
Mortellaroetal	2008	10	7	34	-	-	-	4	10	11	3
Silveira et al	2008	-	-	27	10	-	-	-	-	14	-
Tanrikulu et al	2004	2	-	8	-	-	4	-	-	3	-
Martins Filho et al	2015	8	6	22	21	-	1	-	3	1	1
Wang et al	2009	25	2	83	-	-	7	2	-	-	-
Yamani et al	2011	5	-	7	-	-	-	2	1	-	-
Prosdócimo et al	2018	43	12	80	-	-	4	4	3	7	-
Chen et al	2020	8	-	22	-	-	-	3	3	-	-
Silva et al	2016	27	6	21	25	-	1	2	1	3	1
Kebede et al	2017	17	-	1	5	-	-	4	5	1	-
Costa et al	2012	6	2	18	11	1	1	-	3	-	0
Al Sheddi et al	2015	10	3	17	17	3	3	0	4	1	0
Mamabolo et al	2011	109	25	21	39	2	11	11	12	10	3
Lawal et al	2013	23	4	-	4	-	-	4	-	1	2
Tandon et al	2020	11	9	-	-	-	-	-	-	-	6
Deepthi et al	2016	15	5	27	15	1	2	1	1	1	1
Bhat et al	2019	6	3	3	-	-	-	-	-	-	-

AOT- Adenomatoid odontogenic tumor, KCOT- Keratocystic odontogenic tumor, CB- Cementoblastoma, AFO- Ameloblastic fibroodontoma, AF- Ameloblasticfibroma, OM- Odontogenic myxoma, OF- Odontogenic fibroma, CEOT- Calcifying epithelial odontogenic tumor.



The literature review yielded 256 articles of which 120 duplicate records were excluded. Remaining 136 records were screened and out of which 116 records were excluded from the review. Inclusion and exclusion criteria were applied and hence 20 articles were eligible for the study. 3 more articles were included for the study which was identified by the manual search engine satisfying the eligibility criteria. So, total of 23 cases were included in the review. The countries were more common per pediatric case seen : Brazil, Saudi Arabia, Turkey, India, Nigeria, Taiwan, Ethiopia, Argentina, Italy, Baltimore and Massachusetts, USA. The age distribution of individuals included in the study was 0- 20 years or it was restricted only to first two decades of life. The odontogenic tumors were more predominant in the mandibular posterior region (84%) than that of the maxilla (16%) (Chart 1). In few of the studies the exact anatomical location of the tumors were not mentioned. The predominant tumors that were reported are Odontoma, Ameloblastoma, Keratocystic Odontogenic tumor and Adenomatoid Odontogenic tumor (Tab.1). The predominant odontogenic tumor that was reported is Odontoma which accounted to 641 cases which was 52% (Chart 2) of the total cases reported under the age of 20. Following Odontoma, it was Ameloblastoma

which was 36% with 448 cases reported. Next in line were Keratocystic odontogenic tumors (147) and Adenomatoid Odontogenic tumor (116). The other odontogenic tumors that were encountered in the studies are Cementoblastoma (15), Ameloblastic fibroodontoma (48), Ameloblastic fibroma (56), Odontogenic myxoma (96), Odontogenic fibroma (75) and Calcifying epithelial odontogenic tumor (19).

#### 4. DISCUSSION

Oral and Maxillofacial tumors are not the common entities in the pediatric population and if reported it usually presents as benign ones. Very few soft tissue tumors such as Hemangioma, Papilloma are seen in the Pediatric cases and hence the Odontogenic tumors cover only one third of the maxillofacial pathologies. Odontogenic tumors are the rare pathological lesions that affect the maxillofacial region and they are basically divided into three types based on their origin namely epithelial, mesenchymal and mixed epithelial & mesenchymal odontogenic tumors. [33] Odontogenic tumors are least common in children with an range of 4.2% to 31.0% and hence a study was conducted in the year 2014 at Department of Oral Maxillofacial Surgery, School of Stomatology, China Medical University in regard to it and reviewed records between 1987 and 2008. Medical records of patients younger than 19 years with a diagnosis of tumor or tumor-like lesions were included in the study and found that out of 1028 pediatric tumors, only 310 were odontogenic in the children aging between (0-18 years). [34] In the study, majority of the odontogenic tumors were reported during the later stages of the second decade (i.e) during the permanent dentition stage compared to the early primary dentition and mixed dentition stages. This is due the fact that development of these odontogenic tumors occurs only after the age of 6 in the pediatric population and this is the time when the permanent tooth development takes place with the dental crown being formed. As a result of alteration in the process leads to formation of odontogenic tumors. [33] Odontogenic tumors in children in this series, had predilection for the mandible (84%) which was in agreement with most other studies though, studies in India, Taiwan and Brazil [26, 18, 19] found more cases in the maxilla. In the present study, Odontoma (52%) was the most commonly presented Odontogenic tumors in the Pediatric population and followed by Ameloblastoma (36%), which is then the most common tumors. These two Pediatric tumors were predominantly seen in countries such as Turkey [10], Argentina [12], Italy [15], Taiwan [14, 19], Saudi Arabia [20], Brazil [21], India [26, 31, 32]. In few of the studies, the Odontoma was most frequently encountered [16, 18, 22]. This was in accordance to the study reported by Jones A and Franklin CD [35] which stated that odontomas were largely seen compared to other Pediatric odontogenic tumors. In few of the reports, Ameloblastoma was the common odontogenic tumors seen in children [24, 28, 29]. A study done by Adebayo et al [36] supports the fact that Ameloblastoma was the common occurring Odontogenic tumors compared to all the other tumors. The variation in relative incidences of odontogenic tumors among different populations were noted which may be attributed to the geographic variations. The next in line was the Keratocystic Odontogenic tumor and Adenomatoid Odontogenic tumors. These tumors are seen in the Pediatric population in lesser extent but are the commonest among the Pediatric Odontogenic Tumors. In accordance to their report, a study by Santos R L et al., [37] came out with the information that KOT (2.66%) and AOT (0.58%) were commonly seen in the Children and Adolescents.

The other least common odontogenic tumors presented were Cementoblastoma, Ameloblastic fibroodontoma, Ameloblastic fibroma, Odontogenic myxoma, Odontogenic fibroma and Calcifying epithelial odontogenic tumor. This lower incidence can also be observed in other studies, conducted in various literature reviews. Very few studies have investigated Pediatric OTs and thus a comparison between our results, and those published in the other literature reviews remains very difficult to be compared due to large ethnic diversity found worldwide and geographic conditions.

#### 5. CONCLUSION

Odontogenic tumors are the unique set of neoplasms and are in many instances classified as hamartomas. These Odontogenic tumors usually present as benign entities in children with few to no symptoms. Though the pediatric odontogenic tumors are considered to be rare entities, its occurrence has impact on the growth of jaw bones and tooth development of young children. Our present study gives an overview of the occurrence of the Odontogenic tumors in the pediatric population across the globe. The result of the study was in line with the previous literature reviews suggesting that even though Pediatric odontogenic tumors are uncommon and usually presents as benign tumors, the malignant variants do occur and the detailed information regarding it is necessary for the better understanding of the tumor in regard to treatment.

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