



# A Sixteen-year Retrospective Study of Biopsied Pediatric Oral Lesions from North Kerala

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## ABSTRACT

**Aim:** This study aimed at providing updated information on biopsied oral and maxillofacial lesions from children below 12 years, received between 2001 and 2016, from a tertiary health care center in North Kerala, India.

**Materials and methods:** The archives of the Department of Oral Pathology and Microbiology, Government Dental College, Kozhikode, India, were retrospectively analyzed over a period of 16 years (2001–2016). Patients aged 12 years and below were considered as the pediatric population. Age, gender, site, and histopathologic diagnosis were recorded. Subjects were divided into three age groups: 0 to 6, 7 to 9, and 10 to 12 years. The oral and maxillofacial lesions were classified into eight different categories. Data were analyzed using descriptive statistics.

**Results:** Of the total 8,306 biopsied cases, 334 cases were from the pediatric population. Cases showed almost equal predilection between males (48%) and females (52%); M:F = 1:1.08. Both mandible (26.94%) and maxilla (26.05%) showed almost equal site predilection followed by lower lip (19.76%), gingiva (14.67%), and tongue (5.39%). The most common condition diagnosed individually was periapical cyst (21.86%), followed by mucocele (17.37%) and dentigerous cyst (13.17%). Regarding the diagnostic categories, most lesions were in the cystic group (37.43%) followed by the inflammatory/reactive group (31.44%) and benign odontogenic tumors (10.48%).

**Conclusion:** This study showed similar trends as well as contradictory results when compared with other studies. This can be due to geographical and ethnic variations, difference in criteria, and difference in age groups selected.

**Keywords:** Children, Mucocele, Odontome, Pediatric lesions, Periapical cyst.

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## INTRODUCTION

Children are a distinct part of the general population. Continuous emotional, physiological, socioeconomic, and psychological changes in them lead to the development of various types of alterations and lesions in the orofacial region.<sup>1,2</sup> Certain diseases have a predilection for the pediatric population compared with their adult counterparts. Reviews of oral pathological lesions in children are rare. Some of the previous studies on pediatric oral lesions from different parts of the world are shown in Table 1.<sup>3</sup>

There is difference in the distribution of these lesions in different parts of the world as racial and environmental factors and the lifestyles of populations influence the prevalence of these lesions.<sup>5-7,18</sup> Literature reveals very few studies involving pediatric pathologies from India. To the best of our knowledge, no studies have been done in the pediatric population of northern Kerala. Since geographic distribution is a source of variation,<sup>7,18</sup> the occurrence of this type of lesions in different geographic areas may be a relevant topic to investigate. Thus, the aim of this study was to evaluate retrospectively the prevalence and characteristics of biopsied pediatric oral lesions in this geographical area.

**Table 1:** Studies of biopsied oral and maxillofacial lesions in pediatric populations from different countries

Authors	Country	Cases	Age in years	Period
Skinner et al <sup>4</sup>	USA	1,525	0–19	14
Keszler et al <sup>5</sup>	Argentina	1,289	0–15	25
Das and Das <sup>6</sup>	USA	2,370	0–20	11
Chen et al <sup>7</sup>	Taiwan	534	0–15	12
Lawoyin <sup>8</sup>	Nigeria	561	0–16	10
Sousa et al <sup>9</sup>	Brazil	2,356	0–14	15
Gultelkin et al <sup>10</sup>	Turkey	472	0–15	8
Jones and Franklin <sup>11</sup>	UK	4,406	0–16	30
Dhanuthai et al <sup>12</sup>	Thailand	1,251	0–16	15
Lima et al <sup>13</sup>	Brazil	625	0–14	20
Wang et al <sup>14</sup>	Taiwan	797	0–14	29
Shah et al <sup>15</sup>	USA	5,457	0–16	16
Zuniga et al <sup>16</sup>	Chile	542	0–16	15
Lei et al <sup>3</sup>	Taiwan	1,023	0–15	15
Krishnan et al <sup>17</sup>	South India	97	0–15	10
Heera et al <sup>18</sup>	South Kerala, India	540	0–12	20
Resmi et al (present study)	North Kerala, India	334	0–12	16

## MATERIALS AND METHODS

After obtaining the Institutional Ethics Committee clearance, archives of biopsy reports of pediatric patients were retrieved from the Department of Oral Pathology and Microbiology, Government Dental College, Kozhikode, India, during the time period from January 2001 to December 2016. Children aged 12 years and below were considered as the pediatric population. Subjects were categorized into three age groups; 0 to 6, 7 to 9, 10 to 12 years respectively. Age, gender, anatomic location, and histopathologic diagnosis of pediatric cases were recorded. Lesions were classified into eight categories:

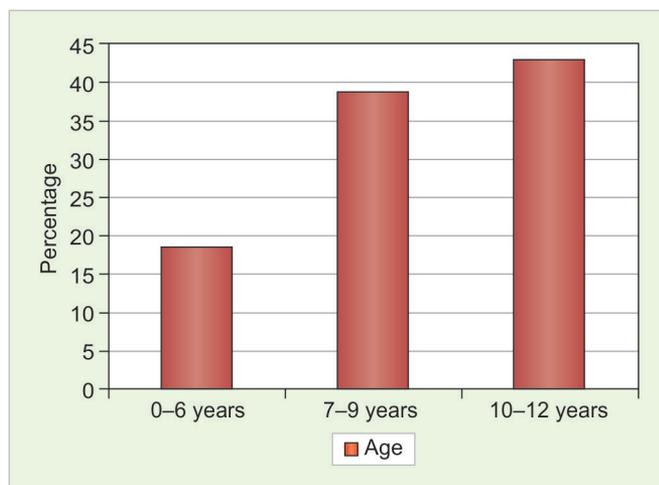
1. Cystic lesions
2. Inflammatory/reactive lesions
3. Odontogenic tumors
4. Soft tissue neoplasms
5. Bone pathologies
6. Developmental conditions
7. Autoimmune diseases
8. Miscellaneous lesions/others

### Statistical Analysis

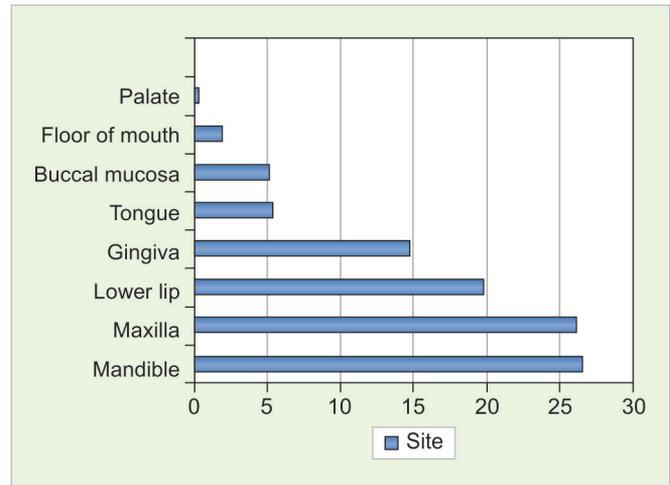
Data were recorded and analyzed by descriptive statistics using computer software Statistical Package for the Social Sciences.

## RESULTS

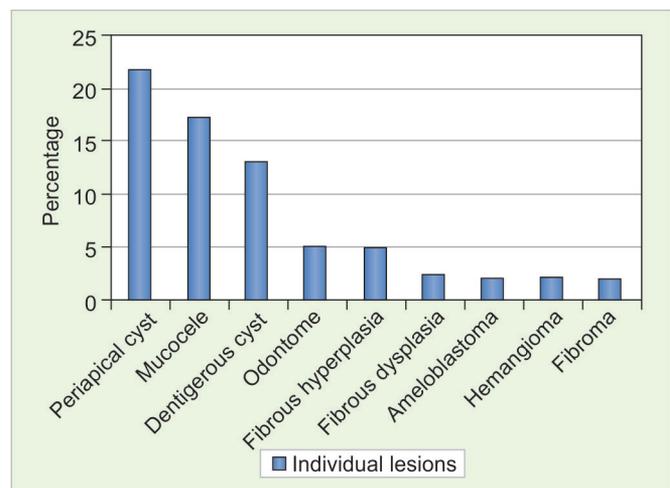
During the 16-year period from January 2001 to December 2016, 8,306 biopsy specimens were received, of which 334 cases were from the pediatric population between 0 and 12 years of age. This patient pool represented 4.02% of all the biopsies received for that period. The prevalence of lesions was the highest in the age group 10 to 12 years (42.81%) followed by 7 to 9 (38.62%) and 0 to 6 years (18.56%; Graph 1).



Graph 1: Percentage distribution of lesions according to age



Graph 2: Percentage distribution of cases according to site



Graph 3: Percentage distribution of the most frequent oral lesions

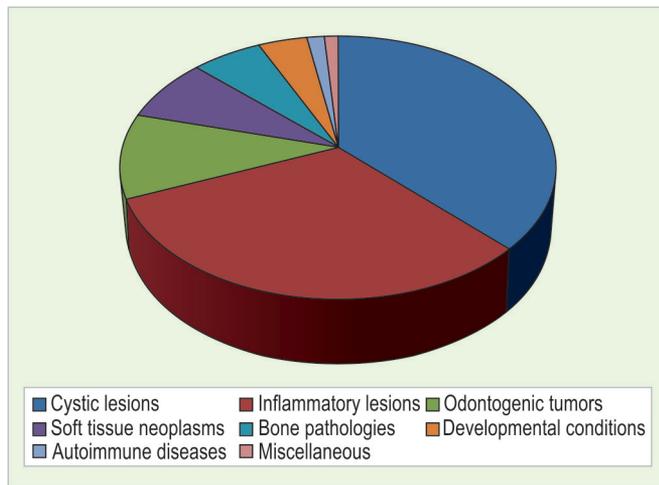
The lowest age observed was 11 months, diagnosed histopathologically as traumatic ulcerative granuloma with stromal eosinophilia.

Almost similar prevalence was observed between boys (48%) and girls (52%) (male:female ratio 1:1.08).

Maxilla and mandible showed almost equal site predilection (26.05 and 26.94% respectively). Other sites were lower lip (19.76%), gingiva (14.67%), tongue (5.39%), buccal mucosa (5.09%), floor of mouth (1.80%), and palate (0.3%; Graph 2).

Periapical cyst was the most common condition (21.86%) followed by mucocele (17.37%) and dentigerous cyst (13.17%; (Graph 3).

Regarding the diagnostic categories, major groups were cystic (37.43%), inflammatory/reactive group (31.44%), odontogenic tumors (10.48%), soft tissue neoplasms (7.78%), and bone pathologies (5.99%). The minor categories were developmental conditions (4.19%), autoimmune diseases (1.5%), and miscellaneous group (1.2%; Graph 4).



**Graph 4:** Percentage distribution of diagnostic categories

Periapical cyst was the most common condition in the cystic group (58.4%) followed by dentigerous cyst (35.2%) and odontogenic keratocyst (4%). Mucocele (55.24%) was the most common condition in the inflammatory/reactive category. Odontome was the most common odontogenic tumor, which accounted for 48.57% of all odontogenic tumors. Ameloblastoma was the second commonest lesion in this category (20%). Hemangioma and fibroma showed equal prevalence in the soft tissue tumors (26.9%). Among the bone pathologies, fibrous dysplasia was the commonest one (40%; Table 2). Malignant tumors and salivary gland tumors were not reported in the sample.

## DISCUSSION

In the current study, we analyzed the prevalence of the biopsied oral and maxillofacial lesions occurring in a pediatric cohort in our institution. Our institution is a major tertiary referral center in northern Kerala, catering to patients from five northern districts of Kerala. Hence, the data presented in this study can, in the most part, represent the prevalence of the pediatric oral and maxillofacial lesions in northern Kerala.

Previous studies on oral and maxillofacial lesions in pediatric patients worldwide showed that the number of pediatric biopsies accounted for less than 10% of all cases referred to histopathology services.<sup>7,18,19</sup> Similar prevalence was observed in our study also, which accounted for 4.02% of the total biopsies. This graph was in accordance with the study from south Kerala, which accounted for 3.47% of the total biopsies.<sup>18</sup> However, some other authors have found the percentage of oral and maxillofacial biopsies ranging between 11 and 27.2% of the total number of pediatric cases.<sup>19</sup> This disparity between different studies may be due to differences in the inclusion criteria. Some studies recruited children up to 15 years of age, whereas others included older children in their study group. In

**Table 2:** Type and prevalence of oral lesions

Categories of lesion	Number of lesion	Percentage of pediatric population
<b>Cystic lesions (n = 125)</b>		<b>37.43</b>
Periapical cyst	73	
Dentigerous cyst	44	
Odontogenic keratocyst	5	
Calcifying odontogenic cyst	2	
Glandular odontogenic cyst	1	
<b>Inflammatory/reactive lesions (n = 105)</b>		<b>31.44</b>
Mucocele	58	
Fibrous hyperplasia	17	
Pyogenic granuloma	12	
Nonspecific ulcer	5	
Peripheral giant cell granuloma	4	
Remaining lesions	9	
<b>Odontogenic tumors (n = 35)</b>		<b>10.48</b>
Odontome	17	
Ameloblastoma	7	
Peripheral odontogenic fibroma	5	
Adenomatoid odontogenic tumor	4	
Ameloblastic fibroma	2	
<b>Soft tissue neoplasms (n = 26)</b>		<b>7.78</b>
Hemangioma	7	
Fibroma	6	
Papilloma	1	
Neurofibroma	1	
Neurilemmoma	1	
Granular cell tumor	1	
Others	9	
<b>Bone pathology (n = 20)</b>		<b>5.99</b>
Fibrous dysplasia	8	
Juvenile ossifying fibroma	4	
Central giant cell granuloma	3	
Others	5	
<b>Developmental conditions (n = 14)</b>		<b>4.19</b>
Hyperplastic follicle	4	
Dentin dysplasia	3	
Dentinogenesis imperfecta	2	
Others	5	
<b>Autoimmune diseases (n = 5)</b>		<b>1.5</b>
Pemphigus	4	
Lichen planus	1	
<b>Miscellaneous/others (n = 4)</b>		<b>1.2</b>
Moderate epithelial dysplasia	2	
Traumatic keratosis	1	
Melanin hyperpigmentation	1	

addition, factors, such as time interval during which study was conducted, geographical region, genetic background of the population, and type of institution where the study was conducted contributed to the difference.<sup>19</sup> It is difficult to determine in which age interval pediatric oral and maxillofacial lesions occur most frequently because of the different age stratifications used in different studies.<sup>19</sup> In

our study, the pediatric populations were categorized into three age groups: 0 to 6, 7 to 9, and 10 to 12 years for the ease of comparison. The majority of the pediatric lesions occurred in the older age group of 10 to 12 years followed by the age interval of 7 to 9 years. This observation is similar to the studies of Heera et al,<sup>18</sup> Krishnan et al,<sup>17</sup> Das and Das,<sup>6</sup> and Chen et al.<sup>7</sup> In some other studies, most of the oral and maxillofacial lesions were seen in mixed dentition period.<sup>9,10,19</sup>

Most of the earlier studies have shown almost an equal distribution between both genders as reported by Gultelkin et al,<sup>10</sup> Das and Das,<sup>6</sup> and Jones and Franklin.<sup>11</sup> However, in the study by Krishnan et al<sup>17</sup> and Heera et al,<sup>18</sup> it was found that pathologies were more common in females. Our study also showed almost equal distribution between both genders and, therefore, it can be inferred that there was no greater propensity for oral and maxillofacial lesions in either sex.

Studies by Maia et al<sup>20</sup> and Lima et al<sup>13</sup> reported maxilla as the most common site, but studies from South India showed predilection for mandible.<sup>17,18</sup> Our study showed almost equal predilection between maxilla and mandible. This is because of the fact that most of the lesions were odontogenic cysts and tumors. The most common soft tissue sites were lower lip followed by gingiva and tongue.

Samples evaluated in the study comprised a wide spectrum of lesions ranging from inflammatory processes to neoplasms.

### Cystic Lesions

Odontogenic cysts, both developmental and inflammatory, represented the major category. Periapical cysts constituted 58.4% of all odontogenic cysts followed by dentigerous cyst (35.2%) and odontogenic keratocyst (4%). This finding was in accordance with the distribution of cysts in the general population in southern India, which showed a predominance of inflammatory cysts.<sup>21</sup> However, our findings were not in accordance with Dhanuthai et al,<sup>12</sup> Heera et al,<sup>18</sup> and Krishnan et al,<sup>17</sup> which showed highest prevalence of dentigerous cyst when compared with radicular cyst. We are of the opinion that increased prevalence of inflammatory cysts in pediatric population of north Kerala may be due to the unawareness of the general population about preventive measures and novel treatment modalities of dental caries. The lack of proper interventions at the appropriate times may be a contributing factor. This necessitates the need for conducting awareness classes at the community and school levels. Dentigerous cyst was the second cystic lesion followed by odontogenic keratocyst. The increased number of developmental cysts suggests the probable

role of genetic factors in its formation.<sup>18</sup> A similar trend was observed in studies by Jones and Franklin<sup>11</sup> and Skiavounou et al<sup>22</sup> in which radicular cysts were the most common lesion.

### Inflammatory/Reactive Lesions

The next major category was the inflammatory and reactive lesions. It is worth noting that the largest group in this category was mucocele (55.24%). This was similar to previous studies.<sup>4,6,18</sup> In our study, the lower lip was the most common site with female predilection. Lower lip, being a trauma-prone site, supports the role of trauma as an etiologic factor either in the form of sharp tooth cusp/biting habit in children. The greater number and density of salivary glands in the lower lip combined with downward force of gravity also play a role in the predilection of mucocele development in the lower lip.<sup>18</sup> The second most reactive lesion in our study was fibrous hyperplasia followed by pyogenic granuloma. However, pyogenic granuloma was the second most reactive lesion in studies from South India.<sup>18</sup> The toothbrushing techniques, which have not been mastered in children, may be considered as a significant cause of microtrauma and inflammation in the gingiva. Trauma to deciduous teeth, aberrant tooth development, and occlusal interferences may also be other precipitating factors.<sup>18</sup> Some other studies showed that peripheral giant cell granuloma and inflammatory fibrous hyperplasia were the commonest lesions.<sup>10,19</sup>

### Odontogenic Tumors

In this study, there were 35 odontogenic tumors that constituted only 10.48% of all pediatric cases. This observation confirms the view of other researchers that odontogenic tumors are rare in children.<sup>11</sup> Odontome was the most common tumor, which accounted for 48.57% of all odontogenic tumors. This is in accordance with studies by Saxena et al,<sup>23</sup> Dhanuthai et al,<sup>12</sup> and Arotiba.<sup>24</sup> Ameloblastoma was the most common odontogenic tumor in a study from south India.<sup>17</sup> Etiology of odontome is unclear as infections or local trauma may be a cause.<sup>18</sup> In our study, ameloblastoma and peripheral odontogenic fibroma ranked in second and third positions followed by adenomatoid odontogenic tumor. Two cases of ameloblastic fibromas were also reported in the study group.

### Soft Tissue Neoplasms

Among the soft tissue neoplasms, hemangioma and fibroma showed almost equal predominance followed by other benign neoplasms. This was in accordance with previous literature.<sup>17,19</sup> It is important to emphasize the fact that all hemangiomas may not always be biopsied. Therefore, occurrence of hemangioma might be

even higher than the number of actual cases.<sup>18</sup> On the contrary, malignant soft tissue neoplasms and salivary gland tumors were not reported in this pediatric cohort. Previous studies also reported very few malignancies in their study samples.<sup>11,18</sup>

### Bone Pathologies

Among the bone pathologies, fibrous dysplasia was the commonest one. Juvenile ossifying fibroma, central giant cell granuloma, osteoma, and exostoses were the other lesions reported. Craniofacial fibrous dysplasia is commonly seen in the second and third decades of life. The present study included children up to 12 years and this may be the reason for the lesser number of fibrous dysplasia cases reported.<sup>18</sup> This is in accordance with the findings by Saxena et al.<sup>23</sup>

### Minor Categories

Three remaining minor categories were classified as developmental conditions, autoimmune diseases, and miscellaneous/other lesions.

#### *Developmental Conditions*

Developmental conditions comprised 4.19% of all pediatric cases of which hyperplastic follicle was the commonest one followed by dentin dysplasia (n = 3) and dentinogenesis imperfecta (n = 2).

### Autoimmune Diseases

Lichen planus (n = 1) and pemphigus (n = 4) were also reported in the sample, which comprised 1.5% of all cases.

### Miscellaneous Other Lesions

Miscellaneous lesions constituted 1.2% of all pediatric cases. One case each of traumatic keratosis and melanin hyperpigmentation and two cases of moderate epithelial dysplasia were reported.

An important aspect observed when analyzing the results of the present study is that there are variations in the prevalence of individual lesions in any given population group. Several salient points regarding regional differences are highlighted by the data in this study. Moreover, it is worth mentioning that the data retrieved in the present study do not reproduce the actual prevalence of oral and maxillofacial lesions in this geographic area since some common pathologies occurring in children, such as herpes and aphthous ulcerations are diagnosed mostly based on their typical clinical features, while the present study was based exclusively on biopsied lesions. The results show a similar trend as well as contradictory findings in comparison with the reported previous

literature. The important difference observed in the study was that the cystic group formed the major diagnostic category predominated by inflammatory cysts followed by the inflammatory/reactive group. The majority of the tumors were benign odontogenic tumors with high rate of odontomas. Hemangiomas and fibromas were the most frequent benign nonodontogenic tumors reported. Salivary gland tumors and malignant tumors were not reported.

### CONCLUSION

In conclusion, little data of this nature have been reported from India and none from north Kerala. This is a large-scale study of pediatric oral and maxillofacial lesions conducted in northern Kerala, India. This type of study also contributes to the characterization of lesions in the pediatric population providing a solid background for diagnosis and treatment of these entities.

### CLINICAL SIGNIFICANCE

This study will throw some light regarding the prevalence and characteristics of the lesions observed in the pediatric population in this specific geographic area, which, in turn, will be useful in treating these lesions appropriately. This information may be valuable in teaching and epidemiology.

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