

Incidence and Distribution of Jaw Pathologies among 0–15 Years Age Group at a Tertiary Rural Health-Care Center of Maharashtra: A Retrospective Study of 10 Years

Abstract

Objective: This study was undertaken to investigate the prevalence of intraosseous jaw lesions among pediatrics (0–15 years of age) in a rural health-care center of Maharashtra and to determine the most common types of lesions and their distribution according to gender and anatomical site involved. **Study Design:** Histopathological data were collected from a database of lesions classified as intraosseous jaw lesions dated between January 1, 2005, and December 31, 2015, from the archives of the Department of Oral and Maxillofacial Surgery, Rural Dental College, Maharashtra, India. All the cases of jaw cysts and tumors among children under 16 years of age group were segregated and scrutinized further under the headings of type of pathologies, anatomical locations where they are most commonly seen, and the age/gender most pertinent to these pathologies. **Results:** The present study revealed that 114/3896 jaw lesions were pediatric jaw pathologies, of which odontogenic cysts were 67/114 and jaw tumors were 47/114. Among the cysts, 70% were developmental cysts and 30% were inflammatory cysts. The majority (71.6%) of the jaw cysts were found in the mixed dentition phase (7–15 years). Among the jaw tumors, 55% of them were categorized under odontogenic tumors and 45% were under fibro-osseous lesions. Similar to intraosseous jaw cysts, tumors of jaws were more prevalent under the age of mixed dentition, which was found to be 87%. Male predominance was seen and the overall male:female ratio was calculated as 1:0.52 and 1:0.5 for cysts and tumors, respectively. Mandibular posterior segment was found to be the most commonly encountered anatomical site in both the groups of cases. **Conclusion:** A large number of cases were recorded, which led us to a path of interrogation through which the reason for increased incidence was extracted, and it was agreed that though the pathogenesis remains the same in every individual, routine health checkups and early diagnosis may reduce the incidence and aggressiveness of pathologies, respectively, which was lacking at the rural area.

Keywords: *Odontogenic cysts, pediatrics, treatment, tumors*

Introduction

Odontogenic cysts and tumors are an important group of pathologies that encounters maxillofacial region.^[1] Odontogenic cysts are derived from the epithelium associated with the development of the dental apparatus. They are broadly classified as developmental group, including keratocysts and dentigerous cysts and an inflammatory group including radicular cysts.^[2]

The orofacial region, including the jaw bones, maxilla, and mandible, is a site for a multitude of neoplastic conditions common to find are odontogenic tumors and fibro-osseous lesions (FOLs). Odontogenic tumors constitute a wide range and diverse

kind of lesions derived from tooth-forming apparatus and its remnants. Odontogenic tumors originate from epithelium or ectomesenchyme or from both, showing varying degrees of inductive interaction between these embryonic components of the developing tooth germ.^[3] Neoplasms affecting children and adolescents are most frequently benign.^[4,5] Odontogenic tumors include entities of a hamartomatous nature, such as odontoma, benign neoplasms, some of which are aggressive as is the case of ameloblastoma and myxoma, and malign neoplasms capable of metastasis.^[6-8] FOLs are a generic designation of poorly defined group of lesions which are recognized to affect the jaws and the craniofacial bones which are known for their confusing area in diagnostic pathology. The disease comprises

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How to cite this article: Tandon P, Shah S, Dadhich A, Saluja H, Chauhan H. Incidence and distribution of jaw pathologies among 0–15 years age group at a tertiary rural health-care center of Maharashtra: A retrospective study of 10 years. *Contemp Clin Dent* 2020;11:39-45.

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Submitted : 25-Apr-2018
Revised : 03-Jul-2018
Accepted : 11-Feb-2020
Published : 13-Jul-2020

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Access this article online

Website:

www.contempclindent.org

DOI: 10.4103/ccd.ccd_328_18

Quick Response Code:



varied processes, in which the normal architecture of bone is replaced by fibrous tissue containing varying amount of foci of mineralization. Genomic alterations are involved in their pathogenesis.

Cysts, tumors, and jaw lesions affect wide range of age groups, most commonly the adult population. Although existing literature on the jaw is extensive, little has been discussed about these cysts occurring in the pediatric population. This study is, therefore, carried out to survey the extent of involvement of jaw pathologies among children aged 0–15 years over a 10-year period (2005–2010), with the aim of determining the prevalence of all histologically diagnosed pathologies.

Materials and Methods

The past 10-year records of the Department of Oral surgery, Rural Dental College, Loni Maharashtra, India, were retrieved from the archives dated between January 1, 2005, and December 31, 2015. It was aimed to target the most common form of jaw pathologies based on previous experiences. Because our study was aimed at estimating the frequency of jaw cysts, tumors, and FOLs in the pediatric population, only cases belonging to the age group 0 to 15 years were included in the sample population. All these cases were analyzed for age, gender, site, and histopathologic type. The age range was categorized into two groups: 0 to 6 years accounting mostly for primary dentition and 7 to 15 years accounting mainly for mixed dentition. The clinical data were cross-checked for recurrences, in order to avoid duplication of cases. The recurrence was assessed based on clinical, radiological, as well histopathological examination. The study followed the Declaration of Helsinki on medical protocol and ethics, and the regional Ethical Review Board of Rural Dental College, PIMS, Loni, approved the study.

Inclusion criteria

Pediatric population <16 years of age with swelling or enlargement noted on either of the arches, correlated radiographically as radiolucent lesions or mixed radiolucent-opaque lesions and histopathologically confirmed as odontogenic cysts, tumors, and FOLs with no malignant transformation, were included in this study.

Results

The total number of biopsy-proven cysts and tumors of jaws accounted in our department in a period of 10 years was 3896, out of which cysts and tumors accounted 2442 (63%) and 1454 (37%) cases, respectively. Among them, pediatric cases of odontogenic cysts and tumors encountered were 114 (2.9%), out of which 67 (59%) cases were diagnosed and confirmed histopathology as odontogenic cysts and 47 (41%) cases as jaw tumors [Figure 1]. Out of total 67 odontogenic cysts, the peak occurrence of 29 (43%) cases were diagnosed as dentigerous cysts, followed by 18 (27%)

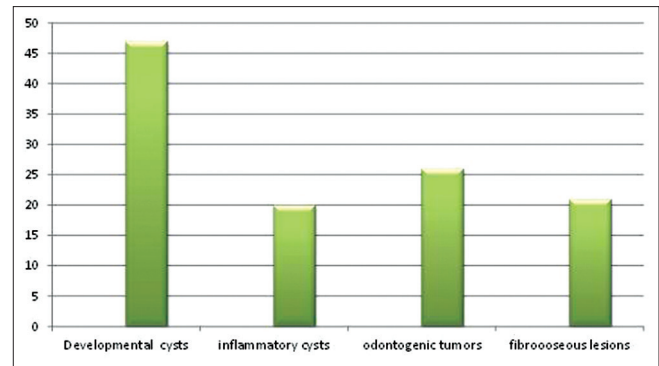


Figure 1: Prevalence of intraosseous jaw lesions

cases of odontogenic keratocysts and the remaining 20 (30%) cases were diagnosed as radicular cysts. Among 26 cases of odontogenic tumors, 11 (23%) cases were diagnosed as ameloblastoma, 6 (13%) cases as calcifying epithelial odontogenic tumor, and 9 (19%) as adenomatoid odontogenic tumor. A total of 21 cases of FOLs reported, out of which the peak incidence of 15 (32%) cases were found as ossifying fibroma and the rest 6 (13%) cases as central giant-cell granuloma.

Based on the location of these lesions, it was observed that mandibular posterior segment was most commonly involved in both the developmental and inflammatory group of cyst [Table 1]. The overall ratio of male to female in cases of odontogenic cysts was found to be 1:0.52, with males being 44 and females as 23 in total. Sixty-two percent of developmental cysts were seen in the age group of 7–15 years and the remaining 38% in the age group of 0–6 years. In case of inflammatory cysts, 95% of cysts were seen in 7–15 years of age and 5% were seen in 0–6 years of age. The overall frequency, age, and gender distribution of various cysts is shown in Table 2. The year-wise distribution of prevalence of jaw pathologies is categorized in Table 3.

In case of odontogenic tumor, posterior mandible was the most prevalent site (36%) in our study [Table 4]. Total 21 males and 5 females were diagnosed as cases of odontogenic tumor. The overall male:female ratio was found to be 1:0.2. 88% of odontogenic tumors were found to be prevalent in the mixed dentition age and 12% in the primary dentition age. Among FOLs, maxillary posterior region was the most prevalent site (19%) according to our data. The overall male-to-female ratio was 1:0.9. In the mixed dentition phase, 86% of cases were found and 14% in the primary dentition phase. The overall frequency, age, and gender distribution of various tumors is shown in Table 5.

The study was a retrospective data collected and documented with an intention of putting forward the increased incidence and prevalence of jaw pathologies in rural areas unlike in any growing urban area. The reason behind this as these pathologies are majority of

Table 1: Site distribution of jaw cysts

| Type of cysts | Maxillary anterior (%) | Maxillary posterior (%) | Mandibular anterior (%) | Mandibular posterior (%) |
|------------------------|------------------------|-------------------------|-------------------------|--------------------------|
| Developmental | | | | |
| Dentigerous cyst | 13 (19) | - | 6 (9) | 10 (12) |
| Odontogenic keratocyst | - | - | - | 18 (27) |
| Total | 13 (19) | - | - | 28 (42) |
| Inflammatory | | | | |
| Radicular cyst | 8 (12) | 3 (4) | - | 9 (13) |
| Total | 8 (12) | 3 (4) | - | 9 (13) |
| Total odontogenic cyst | 21 (31) | 3 (4) | 6 (9) | 37 (55) |

Table 2: The frequency, gender, and age distribution of cysts of the jaws

| Type of cysts | Prevalence of cysts (%) | Males (%) | Females (%) | Male:female ratio | 0-6 years (%) | 7-15 years (%) |
|---------------------------|-------------------------|-----------|-------------|-------------------|---------------|----------------|
| Developmental cyst | | | | | | |
| Dentigerous cyst | 29 (43) | 16 (55) | 13 (45) | 1:81 | 11 (38) | 18 (62) |
| Odontogenic keratocyst | 18 (27) | 14 (78) | 4 (22) | 1:0.28 | 7 (39) | 11 (61) |
| Total | 47 (70) | 30 (64) | 17 (36) | 1:0.56 | 18 (38) | 29 (62) |
| Inflammatory cyst | | | | | | |
| Radicular cyst | 20 (30) | 14 (70) | 6 (30) | 1:0.42 | 1 (5) | 19 (95) |
| Total | 20 (30) | 14 (70) | 6 (30) | 1:0.42 | 1 (5) | 19 (95) |
| Total odontogenic cysts | 67 (59) | 44 (66) | 23 (34) | 1:0.52 | 19 (28) | 48 (72) |

Table 3: Annual representation of occurrence of jaw pathologies

| Year wise distribution | Prevalence of developmental cysts | Prevalence of inflammatory cysts | Prevalence of odontogenic tumors | Prevalence of fibro-osseous lesions |
|------------------------|-----------------------------------|----------------------------------|----------------------------------|-------------------------------------|
| 2005 | 2 | 2 | - | 2 |
| 2006 | 5 | 1 | 4 | - |
| 2007 | 4 | 3 | 2 | - |
| 2008 | 2 | 2 | 2 | 2 |
| 2009 | 4 | - | 3 | - |
| 2010 | 3 | 1 | 1 | 3 |
| 2011 | 3 | 3 | 4 | 4 |
| 2012 | 5 | 2 | 2 | 1 |
| 2013 | 5 | 1 | 1 | 3 |
| 2014 | 8 | 1 | 3 | 4 |
| 2015 | 6 | 4 | 4 | 2 |

Table 4: Site distribution of jaw tumors and lesions

| Type of tumors | Maxillary anterior (%) | Maxillary posterior (%) | Mandibular anterior (%) | Mandibular posterior (%) |
|---|------------------------|-------------------------|-------------------------|--------------------------|
| Odontogenic tumors | | | | |
| Ameloblastoma | - | - | - | 11 (23) |
| Calcifying epithelial odontogenic tumor | - | - | - | 6 (12.7) |
| Adenomatoid tumor | 7 (15) | - | 2 (4) | - |
| Total | 7 (15) | - | 2 (4) | 17 (36) |
| Fibro-osseous lesion | | | | |
| Ossifying fibroma | - | 9 (19) | 6 (13) | - |
| Central giant-cell granuloma | - | - | 6 (14) | - |
| Total | 7 (15) | 9 (19) | 6 (14) | 6 (13) |
| Total bony tumors | 7 (15) | 9 (19) | 8 (17) | 23 (49) |

the times asymptomatic and since people of rural area do not visit or regular checkups thus many a times they go unnoticed till they transform as obvious deformity. These

pathologies may sometimes if secondarily infected become symptomatic; however, because of the poor socioeconomic culture where people strive for normal daily living, their

Table 5: The frequency, gender and age distribution of tumors of the jaws

| Type of tumor | Prevalence of tumor (%) | Males (%) | Females (%) | Male:female ratio | 0-6 years (%) | 7-15 years (%) |
|---|-------------------------|-----------|-------------|-------------------|---------------|----------------|
| Odontogenic tumor | | | | | | |
| Ameloblastoma | 11 (23) | 9 (82) | 2 (18) | 1:0.2 | 2 (18) | 9 (82) |
| Calcifying epithelial odontogenic tumor | 6 (13) | 5 (83) | 1 (17) | 1:0.2 | - | 6 (100) |
| Adenomatoid tumor | 9 (19.) | 7 (78) | 2 (22) | 1:0.3 | 1 (11) | 889) |
| Total odontogenic tumor | 26 (55) | 21 (81) | 5 (19) | 1:0.2 | 3 (12) | 23 (88) |
| Fibro-osseous lesions | | | | | | |
| Ossifying fibroma | 15 (32) | 6 (40) | 9 (60) | 1:1.5 | 3 (20) | 12 (80) |
| Central giant-cell granuloma | 6 (13) | 5 (83) | 1 (17) | 1:0.2 | - | 6 (100) |
| Total fibro-osseous lesions | 21 (45) | 11 (52) | 10 (48) | 1:0.9 | 3 (14) | 18 (86) |
| Total bony tumors | 47 (41) | 32 (68) | 15 (32) | 1:0.5 | 6 (13) | 41 (87) |

threshold of dental pain as they correlate with any pain and discomfort in oral cavity is increased and thus because of ignorance these pathologies could not be treated at an early age.

Discussion

In our retrospective analysis of 10 years of span, out of total 3896 cases of jaw pathologies, only 114 cases accounted for the cysts, tumors, and FOLs of the jaws in the pediatric population. These figures have been reported from a rural trust hospital in the state of Maharashtra of India, where we receive patient referrals from various surrounding districts. The occurrence rate of jaw pathologies in the pediatric age group is relatively low. In the present study, we found three types of cysts: cysts of developmental origin (dentigerous and odontogenic keratocyst) and cysts of inflammatory origin (radicular cyst).

The incidence of pediatric jaw cysts showed predominance of developmental origin (70%) in contrast to those of inflammatory origin (30%). Similar findings were reported by Padmakumar *et al.*^[9] where cysts of developmental origin (63.2%) while those of inflammatory origin accounted only for 33.6% in the pediatric population. Vinicius *et al.*^[10] reported the same results in pediatric patients as 66.7% developmental while 20% as inflammatory cysts in a 21 years of study. Similarly, Bodner^[11] also showed that 45% of cystic lesions seen in children were dentigerous cyst, whereas radicular cyst represented only 13.3% of such lesions.

The increased number of developmental cysts suggests that at the time of development of a child, factors such as eruption and exfoliation of teeth along with the simultaneous growth of jaws and a probable role of genes interplay with each other that attributes to more prevalence in pediatrics.^[12-14] On the other hand, the origin of inflammatory cysts is environmentally related and thus the exfoliation of primary teeth or the absence of tooth follicles

of permanent teeth limits its occurrence unlike in the adult population, which are more affected by cystic lesions of inflammatory origin.^[15-17]

The developmental and inflammatory cysts showed a male predominance with an overall male-to-female ratio of 1:0.52. This was in accordance with the various literature reviews.^[18-20] Relatively poor oral hygiene and increased susceptibility to trauma among boys might be the reason for male predominance.^[21] The anterior maxilla was the most common site affected by dentigerous cyst (19.4%) and posterior mandible was most commonly affected by odontogenic keratocyst (26.8%). This was in accordance with the study of Nannan *et al.*, who reported dentigerous cyst (57.6%) predominantly located at the anterior maxilla and odontogenic keratocyst (60.3%) predominantly located at the posterior mandible.^[22] The reason for increased occurrence of dentigerous in maxillary anteriors may be because of more chances of unerupted maxillary canines in the age of 0–15 years; however, mandibular predominance of dentigerous cyst was reported in few studies.^[10,23,24] Manor *et al.*^[25] have attributed that the incidence of inflammatory jaw cysts in the pediatric population is probably less when compared to adult population owing to the fact that exfoliation/loss of primary teeth may result in the resolution of certain cystic lesions that are limited in size and are asymptomatic, particularly when they do not involve the underlying tooth follicles of permanent teeth. In primary teeth, the inflammatory stimulus would not have enough time to act as a chronic irritant. The occurrence of inflammatory cysts in permanent teeth is also infrequent in children and adolescents, because their pathogenesis starts from the pulp necrosis and recently erupted permanent teeth are generally healthy, not showing the condition necessary for the development of cystic lesion.

In our study, 13% of inflammatory cysts were seen to involve mandibular posterior quadrants, followed by maxillary anterior segment (12%). The inflammatory cysts are more prevalent in the maxillary anterior and mandibular

posterior segment probably due to the fact that maxillary anterior teeth are more prone to trauma and mandibular posterior teeth present more fissures and grooves, being more susceptible to caries and, consequently, to periapical inflammatory lesions.^[18]

Odontogenic tumors are considered highly infrequent because they account for a very low percentage of the diseases that affect both hard and soft tissues.^[4,5,7] However, their frequency is relevant when considering tumors of the jaws.^[26,27] In general, odontogenic tumors in the pediatric population are rare and considerably more than in the adult population. Odontogenic tumors constituted 26 (55%) cases, which represented quite a substantial figure. Our study was in accordance with Adebayo *et al.*,^[28] Sato *et al.*,^[26] Tanaka *et al.*,^[27] and Guerriasi *et al.*^[29] who attributed high percentage of odontogenic tumors in their respective series of patients; however, findings of the study of Jones and Franklin and other researchers were in contrast to ours, which showed that odontogenic tumors were rare in children.^[5] Odontogenic tumors showed a male predilection in our study, which was in accordance with Urs *et al.*,^[13] whereas the study of Jones and Franklin showed a female predilection.^[5]

Among the odontogenic tumors, ameloblastoma showed 23% of cases and found this tumor as the most frequent in our study, which was similar to the findings of Jones *et al.*^[5] and Tanrikulu *et al.*^[30] Ulmansky *et al.*^[31] observed significantly lower percentages of 11%; however, the study of several authors^[26-28,32,33] have found frequency of ameloblastoma within total cases of odontogenic tumors to range between 24% to 53.8%. Ameloblastomas are rare in children^[34] when compared to adults. According to Ord *et al.*, majority of ameloblastomas in children are unicystic,^[34] which was similar to our findings. In our study, out of four cases of ameloblastoma, three of them were unicystic. Ameloblastomas showed male predominance with mandibular posterior region as the most commonly involved site. This was in accordance with the results of Urs *et al.*^[13]

Adenomatoid odontogenic tumor in our study was the second most common odontogenic tumor encountered, 9 (20%), which was in accordance with Asamoia *et al.*^[35] However, Guerriasi *et al.*^[29] stated that this tumor is rare type and accounted for 5.2% of odontogenic tumors in their series of cases. It shows female predilection with propensity toward anterior maxilla.^[36] Similar findings were recorded in our study also.

Among the FOLs, juvenile ossifying fibromas were the most common types seen with total of 32% out of total 45% cases of FOLs. These lesions showed a female predilection in our study, which was similar to the finding of Scholl *et al.*^[37] and Urs *et al.*^[13] Pieter^[38] suggested as although both maxilla and mandible are affected with a slight predominance of maxilla similar as our study

however according to Liu Y *et al.* findings mandibular predominance was seen.^[39]

In our study, we found total 12.7% of cases as central giant-cell granuloma with a mandibular predominance. This was similar to the findings of Kaffe *et al.*^[40] and Whitaker and Waldron,^[41] in which mandibular/maxillary ratio of 2:1 and 3:1 was found, respectively. The lesions are more common in the anterior region and frequently extend across the midline Hegde.^[42] In our 6 cases of central giant-cell granuloma, one patient showed a lesion extending beyond the midline.

The planning and execution of treatment for intraosseous bony lesion in pediatric patients represents the most difficult challenge in clinical practice^[43] as most of the cysts and tumors tend to reoccur if conservatively treated; however, radical procedures will affect the child's functional, social, as well as psychological life. The treatment recommended for these lesions varies from most conservative measures including marsupialization to extensive procedures including enucleation with adjuvant procedures such as application of Carnoy's solution in cases of odontogenic keratocysts. In cases of inflammatory cysts, endodontic treatment of the diseased tooth followed by enucleation was the treatment of choice to remove all the infected periapical tissue. This method was chosen over marsupialization, as the radicular cysts unlike developmental cysts do not attain big size and thus can easily be curretted. Marsupialization was the most commonly performed surgical treatment modality for developmental cysts with large size posing a threat to fracture jaw and/or risk of damaging adjacent tooth or neurovascular structure. This is in sharp contrast to the treatment usually rendered in case of the jaw cysts in adults where enucleation is the most commonly employed technique in the management of jaw cysts.^[21,44] Various studies in literature show that there are less chances of recurrence in cases of odontogenic cysts, especially with odontogenic keratocysts if treated with marsupialization. Browne proposed that recurrence of the keratocysts was caused by the nature of lesion itself, i.e., the presence of additional remnants of dental lamina from which a cyst could develop and which was not related to its method of treatment.^[45] In our study, 12% of the total operated cysts showed recurrence. Odontogenic keratocysts showed maximum recurrence among all the types with a total of 33% in a follow-up period of 10 years. The published recurrence rate ranges from a maximum of 62% to a minimum of 0%.^[18,46-48]

Among the odontogenic tumors, ameloblastomas in children are generally less aggressive unicystic variants which do not extend beyond the cystic wall of the tumor cell.^[49] This type of tumor has a much better prognosis. We treated all the cases of ameloblastoma by complete removal of the tumor, followed by application of modified Carnoy's solution as a chemical cauterizing agent to destroy leftover

tumor cells. In our four cases of unicystic ameloblastoma, none of the cases showed recurrence, whereas out of remaining seven patients of multicystic ameloblastoma, three patients showed recurrence after approximately 2 years. Fung stated that in younger patients, there would be maximum cancellous bone, with the lesion growing more rapidly, with extensive destruction.^[50] Thus, we choose marginal resection with 0.5 to 1 cm past what appears to be normal bone^[51] and did not encounter recurrence in a follow-up period of 5 years. Odontogenic tumors other than ameloblastomas and all the FOLs at our center were treated by local curettage and excision of the tumor and none except one patient of central giant-cell granuloma showed recurrence after 5 months which was then treated by segmental resection followed by reconstruction [Table 6].

The recurrence depends on stage at which a case is diagnosed and surgical technique used for its treatment. The physical signs and symptoms of odontogenic cysts and tumors will depend on a certain extent on the dimensions of the lesion. A small lesion is unlikely to be diagnosed on a routine examination of the mouth because signs will not be demonstrable. Such lesions are only likely to be detected at an early stage as the result of routine radiographic examination. In these areas, pathologies may become extremely large before expansion is observed clinically. Because of the poor oral hygiene, relative late diagnosis and irregular follow-ups in rural districts due to obvious restraints unlike in urban areas which may explain the recurrence in our study.

This study, thus, aids in better knowledge of the prevalence of developmental odontogenic cysts and bony tumors of jaws in a rural pediatric population and shows that early detection and short intervals of regular follow-ups hold an importance next to adequate treatment.

Importance of conducting this study

- To analyze the prevalence of odontogenic pathologies such as cysts and tumors among younger population
- The scenario of extent and occurrence of these

pathological conditions inclines more toward rural population

- To bring forward the importance of extension of health services where early diagnosis can be made which may help in reversing the graph.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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Table 6: Recurrence rate

| | Type of pathology | Recurrence (%) |
|---------------------------------------|---|----------------|
| Odontogenic cysts | Dentigerous cyst | - |
| | Odontogenic keratocyst | 6 (33) |
| | Inflammatory cyst | 2 (10) |
| | Total | 8 (12) |
| Odontogenic and fibro-osseous lesions | Ameloblastoma | 3 (28) |
| | Calcifying epithelial odontogenic tumor | - |
| | Adenomatoid odontogenic tumor | - |
| | Ossifying fibroma | - |
| | Central giant-cell granuloma | 2 (33.3) |
| | Total | 5 (11) |

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