

Surgical Management of intraosseous cystic lesions in paediatric patients: A prospective study

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ABSTRACT

Objectives: To present the characteristics and management of intra-osseous cystic lesions of jaws in paediatric patients.

Methods: Study Design: This prospective study was done from March 2018- February 2022 in a tertiary care centre. Thirteen paediatric patients in the age group of 7 to 14 years presenting with cystic lesions were treated surgically in more conservative manner. Treatment performed is early and exact methods of intervention depends on the chronological age, location of cysts and state of dental development.

Interventions: Depending upon the involvement of the vital structures by cystic lesion, obliteration of the buccal sulcus, displacement of the teeth and associated structures, type of dentition and location of the cystic lesion, the patients were treated with complete enucleation of the smaller lesions and extraction of primary/permanent teeth followed by primary closure of the surgical cavity and marsupialization followed by enucleation. Follow up period was 6 months.

Results: In 92.3% of the cases, the cysts were surgically enucleated followed by extraction of teeth whereas in 7.69% of the cases, the cystic lesions were marsupialized. The most common cysts were radicular/periapical cysts (12 patients) followed by dentigerous cysts (1 patient) evaluated on histopathological evaluation.

Conclusion: In paediatric population the cystic lesion should be treated early and the intervention is guided by the site of the cyst development, age of the patient and status of the dental development. Treatment was surgical removal without interfering, whenever possible, with the development of the dentition. Surgical approach was usually enucleation and, to a lesser extent, marsupialization.

Keywords: surgical, enucleation, marsupialization, cystic lesion, treatment.

INTRODUCTION

A cyst is 'a pathological cavity having fluid, semifluid or gaseous contents and which is not created by the accumulation of pus' ¹. Cystic jaw lesions may be odontogenic or non-odontogenic, epithelial or non-epithelial, developmental, or inflammatory in origin ². Odontogenic cysts are those cysts that arise from odontogenic epithelium and occur in the tooth-bearing regions of the jaws. It is usually considered that proliferation and/or degeneration of this epithelium leads to odontogenic cyst development. The distribution and characteristics of jaw cysts in children are different from those in adults. In children there is a relatively high rate of developmental cysts, whereas in adults the inflammatory cysts are more common².

The cystic lesions in paediatric patients has been the issue of importance. There has been wide variety of clinical presentation associated with the intra-osseous cystic lesions in children. These cysts are often asymptomatic unless there is an acute inflammatory exacerbation and, therefore, these lesions are usually diagnosed on routine radiographic examination³. The presentation varies with the location and severity of the cystic lesion. The most frequent findings associated includes the expansion of the cortical bone plate, obliteration of the sulcus, extra oral swelling, distortion of the face. Swelling, tooth mobility, teeth displacement, and sensitivity may be present if the cyst reaches the size larger than 2 cm in diameter⁴. Radiographs show a unilocular radiolucent lesion with well-defined sclerotic margins that is associated with the crown of an unerupted tooth. Radicular resorption of teeth in the region of the lesion is common. The complications associated with dentigerous cyst include pathologic bone fracture, loss of permanent tooth, bone deformation, and development of squamous cell carcinoma. Since the cyst may increase in size, the indicated treatment is surgical removal of lesion and involved teeth, or decompression to salvage the involved teeth⁵.

Management of dentigerous cyst in primary dentition needs special consideration regarding the preservation of the developing permanent tooth buds. The principle involved the treatment are the same irrespective of the age of the patient. However, the technique in children is necessarily modified by the certain anatomical, physiological and psychological factors. The surgical approach to cystic lesions of the jaws were: total enucleation for small lesions, and enucleation with bone grafting, or resection, marsupialization for decompression of large volume cysts, or a combination of both^{6,7}. The treatment of choice is dependent on the size and localization of the lesion, the bone integrity of the cystic wall, and its proximity to vital structures. It is been suggested that marsupialization of the cyst lining is the treatment of choice for dentigerous cyst in children in order to give a chance to the unerupted tooth to erupt⁸, the major disadvantage of marsupialization is that pathologic tissue is left in situ, without a thorough histologic examination;^{9,10}. The correct diagnosis of an odontogenic cyst is important as some of them have an aggressive behaviour and a propensity to recur along with only limited data on the prevalence of odontogenic cysts in paediatric patients¹¹.

Although existing literature on the incidence and clinico-pathological features of cysts of the jaw in adult population is extensive, little has been discussed regarding management of intra-osseous cysts specially in the paediatric population. Furthermore, the management of the cystic lesions possess a great challenge specially in children. To the best of our knowledge, there is limited reported literature on jaw cysts in the paediatric age group within the Indian population.

The present study was aimed to present the distribution, characteristics, histopathologic diagnosis and comprehensive management of intra-osseous cysts of the jaws in the paediatric population reporting to a tertiary government dental healthcare educational institution located in the state of Haryana, India.

MATERIALS AND METHODS

Study design

This prospective study was done from March 2018 to February 2022 in a tertiary care centre. Thirteen paediatric patients, 7-14 years old, with intraosseous cystic lesions of the jaws, were treated. All cysts were surgically removed. No bone grafts were used and the teeth involved were preserved whenever possible. Removed tissues were examined histologically. Follow-up period was 6 months.

Interventions:

Depending upon the age of the patients, size and volume of the cystic lesions, location of the cyst and approximate to the vital structures and status of dentition the patients were treated with complete enucleation of the smaller lesions and extraction of primary/permanent teeth followed by primary closure of the surgical cavity and marsupialization followed by enucleation. In 2 cases wherein cystic lesions were associated with permanent teeth, the apicoectomy of the affected teeth were performed. Furthermore, in 1 case wherein large cystic lesion involved considerable amount of maxilla, a two-stage operation were performed, firstly marsupialization followed by complete enucleation and orthodontic treatment thereafter. The results were highly satisfactory.

Independent Variables:

Involvement of the vital structures by cystic lesion, obliteration of the buccal sulcus, displacement of the teeth and associated structures, type of dentition (primary, mixed and early permanent), location of the cystic lesion (maxilla or mandible).

Outcome variables:

Post-operative healing, intra-oral tenderness, occlusion status, post-operative pain, and/or discomfort, resolution of cortical plate expansion and extra-oral swelling.

The site distribution of the cysts was divided into three classes, class 1 including the dentoalveolar region involving the central incisors, lateral incisors and canines, class 2 spanning between mesial aspect of first premolar/first primary molar to the tuberosity in the maxilla and ramus in the mandible and class 3 including both classes 1 and 2. An incisional biopsy was taken for all cases where surgical enucleation was performed whereas excisional biopsy was the method of preference for 1 case wherein marsupialization was done followed by enucleation of the cystic lesion. The interventional/treatment approach associated with independent variables were evaluated throughout the surgical management of intraosseous cystic lesions performed in paediatric patients. The clinical presentations of the intraosseous cysts were related to the histopathological findings in patients. Moreover, post-operative outcome variables as per the treatment modalities of surgical management of intraosseous cysts in patients were noted and evaluated.

RESULTS

The table 1 reflects the treatment aspects for the surgical management of intraosseous cysts performed in the patients. The surgical enucleation of the cystic lesions followed by extraction of the primary tooth along with primary closure of the surgical cavity is being performed on 9 (69.23%) out of 13 patients (Figure 1a,b,c,d,e,f).



Figure 1 (a,b,c,d,e,f): Showing surgical enucleation of the cystic lesions followed by extraction of the primary tooth along with primary closure of the surgical cavity.

All the patients were in mixed dentition. The mean age of the patients is 9.78 years and all the cystic lesion were class II i.e lesion span between mesial aspect of first premolar/ first primary molar to the tuberosity in the maxilla and ramus in the mandible. The surgical enucleation of the cyst followed by the extraction of the primary tooth along with enucleation of the permanent tooth bud was carried out in 3 (23.07%) patients (Figure 2a,b,c,d,e) wherein patients were in mixed dentition.



Figure 2 (a,b,c,d,e): Showing surgical enucleation of the cyst followed by the extraction of the primary tooth along with enucleation of the permanent tooth bud.

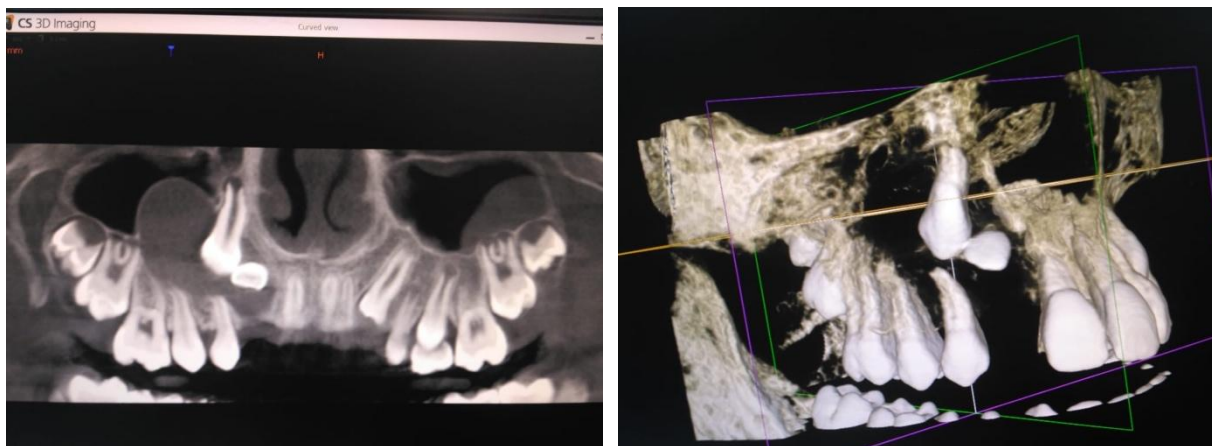




Figure 3 (a,b,c,d,e,f,g): Showing marsupialization is followed by the fixed orthodontic treatment.

The mean age was 8.5 years and the cystic location is class II. Marsupialization for decompression of cystic cavity followed by enucleation was carried out in 1 (7.69%) patient having permanent dentition. The age of the patient is 13 years and the cyst is class I including the dentoalveolar region involving the lateral incisors. The marsupialization is followed by the fixed orthodontic treatment the case (Figure 3a,b,c,d,e,f,g).

Table1:- Showing the treatment aspects for surgical management of intraosseous cysts performed in patients.

Sr. No	Study Subjects	Mean Age(years)	Location of the cyst	Status of dentition/ Teeth involved.	Treatment performed
1.	9	9.78	Class II (9 cases)	Mixed dentition (85, 84, 85, 84, 75, 85, 85, 75, 46)	Surgical enucleation of the cyst followed by extraction of the primary tooth. Primary closure of the surgical cavity.
2.	3	8.5	Class II (both cases)	Mixed dentition (Case 1- 75 & 35) (Case 2- 74 & 34)	Surgical enucleation of the cyst followed by extraction of primary tooth along with enucleation of permanent tooth bud. Primary closure of the surgical cavity.
3.	1	13	Class I	Permanent (12 & 13)	Marsupialization for decompression of cystic cavity followed by enucleation. Fixed orthodontic treatment.

The table 2 presented the histopathological findings with the clinical presentation of intraosseous cysts in patients. Based upon histopathological evaluation in the study, twelve cases of radicular cyst/periapical cyst were found with mean age of

the patients 9.83 years. In contrast, only one case of dentigerous cyst was confirmed on histopathological evaluation involving permanent premolar. The clinical presentation in all patients were extra-oral swelling, displacement of teeth and associated structures, obliteration of buccal sulcus, intra-oral swelling and approximation to vital structures.

Table 2:- Relating the histopathological findings with the clinical presentation of intraosseous cysts in patients.

Sr. No	Histopathologic evaluation	Tooth involved	Study Subjects	Mean Age (years)	Clinical presentation				
					Extra-oral swelling	Displacement of teeth & associated structures	Obliteration of buccal sulcus	Intra-oral swelling	Approximation to vital structures
1.	Dentigerous cyst	74 & 34	1	7	Y	Y	Y	Y	Y
2.	Radicular cyst/Periapical cyst	Mixed & Permanent Dentition	12	9.83	Y	Y	Y	Y	Y

(Y= yes)

Table 3 demonstrated the post-operative outcome variables in compliance to the various treatment modalities of surgical management of intra-osseous cysts in study. Out of a total 13 patients, nine (69.23%) patients were placed for surgical enucleation of the cystic lesion followed by 3 (23.07%) for surgical enucleation of cyst followed by enucleation of primary/permanent teeth and one (7.69%) marsupialization for decompression of cystic cavity respectively. The post-operative outcomes were found to be significant statistically ($p < 0.05$) after 6 months of follow up in case of surgical enucleation of cystic lesion along with primary closure of cystic cavity when compared with marsupialization for decompression of the cystic lesions in study population. Moreover, the reduction/resolution of post-operative healing, intra-oral tenderness and extraoral swelling were more in comparison to resolution of cortical plate expansion and occlusal status after 6 months of follow ups. However, on statistically evaluation the results were found to be non-significant. ($p < 0.05$).

Table 3:- Relating the post-operative outcome variables as per the treatment modalities of surgical management of intraosseous cysts in patients.

Sr. No	Surgical treatment modalities	Study subjects	Post-operative outcomes variables (After 6 months)				
			Post-operative healing	Intra-oral tenderness	Occlusal status	Extraoral swelling	Resolution of cortical plate expansion
1.	Surgical enucleation of the cyst followed by extraction of the primary tooth. Primary closure of the surgical cavity.	1	+++	+++	+	+++	+
		2	+++	+++	++	+++	++
		3	+++	+++	+	+++	+
		4	+++	+++	+	++	+
		5	+++	++	+	+++	+
		6	+++	+++	++	+++	++
		7	++	++	+	++	+
		8	+++	+	+	+++	++
		9	+++	++	+	++	+
2.	Surgical enucleation of the cyst followed by extraction of primary tooth along	1	++	+	+	+++	+

	with enucleation of permanent tooth bud. Primary closure of the surgical cavity.	2	+++	++	++	+++	++
3.	Marsupialization for decompression of cystic cavity followed by enucleation. Fixed orthodontic treatment.	1	+	+	+	+	+

DISCUSSION

Cystic lesions of the jaws can be either odontogenic or non-odontogenic, developmental, or inflammatory in origin. The occurrence of cystic lesion in jaws especially in pediatric population is relatively scarce^{1,11,12}. The distribution of cysts in the general population in India as reported by Padmakumar et al¹³ showed a predominance of inflammatory cysts. In the present study the incidence of the inflammatory cysts was found to be 91.66% whereas the occurrence of the development cyst particularly dentigerous cyst were 8.33%. However, Telang et al¹⁴ had reported 43.2% of radicular cysts and 20.2% dentigerous cysts in their 19-year retrospective study of cystic lesions of pediatric and adolescent population.

The difference in the distribution of developmental and inflammatory cysts may probably be attributed to the state of dynamism of dento-alveolar complex¹⁵. This may be a result of interplay of several factors including the development and eruption of the succedaneous dentition and the simultaneous skeletal growth of the maxilla and mandible in this age group. In addition, authors suggested that the incidence of jaw cysts in the pediatric population is probably under-reported 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. The exfoliation of primary teeth may also limit the development and expansion of these cystic lesions which again would help in widening the disparity in reported incidence of developmental and inflammatory cysts in the pediatric population when compared to that of the adult population. The increased number of developmental cysts also suggests a probable role of genetic factors in its formation whereas inflammatory cysts have obviously more of an environmental etiology^{13,15}.

The frequency of cystic lesion reported to be greater in males because they are more likely to neglect their teeth or they are more likely to sustain trauma to their teeth, compared to females, all of which may be attributed to the etiology for cyst formation^{1,16}. The majority of the cystic lesion in pediatric population were located in mandible. In the present study, one out of 13 cystic lesions were located in anterior maxilla while the rest of the cysts were located in mandibular jaw predominantly in posterior region. The most common site of the biopsy lesion was found to be the mandibular posterior region based upon the present operated cases. Moreover, the literature suggested that the common site of biopsy was the mandible (66.6%), followed by maxilla (33.4%), which differed with findings of Salako & Taiwo¹⁷, who found maxilla to be the commonest site. The odontogenic cysts usually presented as a single lesion on unilateral basis. However, there are certain lesions that appeared as bilateral involvement of both the half.

Radicular cyst was the most common diagnosis, accounting for 43.2% of the odontogenic cysts which was similar only to Jones & Franklin¹¹ and Lawoyin¹⁸. While most other studies reported dentigerous cysts as the common cyst in the pediatric populations^{17,19,20,21}. The reason for this difference may relate to the prevalence of caries and varied oral health regimes between different countries and regions.

Clinical presentation of infection at the cystic lesion is usually associated with acute or chronic inflammation at the cyst wall. In such cases the epithelial lining of the cyst wall may be destroyed, regardless of cyst origin, leaving the cyst wall with granulation tissue. There are wide variety of intraoral presentation associated with the cystic lesions. The most common intraoral findings were extra-oral swelling, expansion of the buccal cortical plate, obliteration of the buccal sulcus, malocclusions such as displacement of the permanent teeth, gross malocclusion. These clinical features were present in almost all radicular/periapical and dentigerous cystic lesion reported the present study in pediatric population²².

There are very few studies in the literature done over cystic lesion occurring in children. Correct diagnosis of odontogenic cysts and their variants is important owing to the propensity of certain lesions to recur and have an aggressive nature. There are wide variety of diagnostic aids for evaluation of the cystic lesions¹³. Imaging modalities like intraoral periapical

radiograph, panoramic radiograph and occlusal radiograph were employed in the evaluation of these pathologic jaw cysts.²³ Extraoral radiographs, computed tomography (CT) scans had also proved useful in few cases. The utility of CT scans is limited usually to cases of larger lesions where it is difficult to discern the borders and extent of these lesions²⁴. The radiographically the most important findings were radiolucent lesion surrounded by radiopaque borders. Histopathological examination using biopsy of the lesions were the definitive criteria for diagnosis of the cystic lesion. Fine Needle Aspiration Cytology (FNAC) is advisable in case of liquid/ fluctuant content of the cyst.

The management of the cystic lesions in children is mostly conservative. The factors such as the size and location of the cystic lesion can change the treatment option. The available options for the treatment of these lesions in children include total enucleation of the cyst with primary closure and/or marsupialization. Marsupialization of the cyst is the treatment of choice which gives a chance to the unerupted tooth to erupt in large cysts; however, this technique creates notable disadvantages. To illustrate, the two-stage surgical procedure may result an intolerable procedure for a child and lifting behind a pathological tissue²². Enucleation with primary closure was the treatment choice in our case as the cyst was small, and the displacement of the unerupted permanent tooth was severe. Although dentigerous cyst lesions are rare in the first decade of life, they can form in young children. For this reason, the true treatment choice with the early clinical diagnosis of these lesions may increase the rate of success. A long-term follow-up is important for the diagnosis of the recurrence as well.

CONCLUSION

The inflammatory radicular cyst is the most common in the pediatric age group. The lesions show a male predominance with posterior mandibular region being the common occurrence site. The treatment modalities or surgical management of cystic lesions were marsupialization, enucleation, and enucleation with allogeneic or xenogeneic bone grafting. Treatment was surgical removal without interfering, when possible, with the development of the dentition. Surgical approach was usually enucleation and, to a lesser extent, marsupialization. Following enucleation of a cystic jaw lesion, the entire surgical specimen and not only a biopsy specimen, should be examined histopathologically to prevent any possibility of recurrence.

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