

Treatment Modalities and Rehabilitation Procedures of Dentigerous Cyst Among the Paediatric Population - A Systematic Review of Case Reports

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ABSTRACT

BACKGROUND: The dentigerous cyst which wraps around the impacted teeth is the most prevalent developmental odontogenic cyst among the Paediatric population in India. Various treatment modalities were proposed previously, such as enucleation, marsupialization, fenestration, or a combination technique with or without preservation of the affected dentition.

OBJECTIVES: The aim of the Paediatric dentist should rely on the preservation of permanent teeth. Hence, this systematic review aimed to evaluate the prevalence and describe the different treatment methods and rehabilitation techniques conducted following the surgical excision of dentigerous cysts among the Paediatric population in India, which is significant to Paediatric dentists.

METHODS: Systematic review registered under PROSPERO following PRISMA guidelines. Thorough search was done in PubMed, EMBASE, ProQuest and Cochrane Library.

RESULTS: The results showed that the dentigerous cyst occurred commonly in the age group of children aged 6-10 years (72.22%), with a male predilection

(80.56%). The complete cyst enucleation with the removal of milk teeth and permanent dentition (52.78%) was preferred more than the preservation of the affected dentition. After complete enucleation of the cyst in only 57.89% of cases, rehabilitation procedures were planned. The results showed that proper rehabilitation was not planned in many cases, which may lead to poor oral hygiene maintenance and early space loss.

CONCLUSIONS: It is important for the Paediatric Dentist to know the importance of preserving the permanent teeth or plan a wise rehabilitation procedure to replace missing teeth to maintain the proper oral health of the children.

Keywords: Dentigerous Cyst, Paediatric Dentistry, Enucleation, Marsupialization, Oral Rehabilitation.

INTRODUCTION

A cyst is described as a hollow sac with an epithelial covering and soft material inside it. Odontogenic cysts develop from the components that form the tooth structure.^[1] Odontogenic Dentigerous cysts wrap the dental enamel and are connected to the cervical region of a developing or impacted tooth.^[2] The second most prevalent

odontogenic cysts are dentigerous cysts, with 0.91%–7.3% prevalence in all cases. [3] However, the underlying pathology of dentigerous cysts remains controversial. The tension of the developing tooth on the dental sac inhibits the vascular flow, resulting in exudate development between the reduced enamel epithelium and the unerupted dental crown. [4, 5] The formation of a dentigerous cyst can pose a problem with the formation of primary and permanent teeth, osseous deformation, and ectopic tooth eruptions and even lead to the development of malignant disorders such as squamous cell carcinoma, mucoepidermoid carcinoma, and ameloblastoma. [6, 7] Various therapies for dentigerous cysts include enucleation, marsupialization, or a combination of these, with or without preservation of the affected dentition. This systematic review aimed to determine the prevalence and list the various treatment modalities and rehabilitation procedures performed after the surgical removal of dentigerous cysts within the Paediatric population in India, which is of great importance to Paediatric dentists.

MATERIALS & METHODS

The methodology for the systematic review has been submitted to the Prospective Register of Systematic Reviews (PROSPERO CRD42022368732). This systematic review was conducted in compliance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) standards. [8]

I. RESEARCH QUESTION – According to PICO(TS) strategy,
Population – children of age group 0–15 years

Intervention – surgical intervention without extraction of affected teeth
Control – surgical intervention with the extraction of the affected teeth
Outcome – complete rehabilitation of affected cases
Time frame – 1999– December 2023
Study design – case series and reports documented on dentigerous cysts in India.

II. ELIGIBILITY CRITERIA

- a. Inclusion criteria
- All case studies and case series of dentigerous cysts recorded and documented in children below 15 years in India.
 - Confirmed diagnosis of dentigerous cysts by clinical, radiographic, and histological findings as per the WHO classification.
 - Documented only in the English language.
- b. Exclusion criteria
- Case reports with insufficient clinical, radiographical, and histological data.
 - Case reports of countries other than India.
 - Case reports in regional languages.
 - Dentigerous cysts related to supernumerary teeth or odontoma.
 - Dentigerous cyst related to any syndromes.
 - Reports of animal studies.

III. DATABASES SEARCHED AND SEARCH STRATEGY (TABLE 1) DATABASES USED AND KEYWORDS SEARCHED

The reports were reviewed by two authors manually to exclude duplicates and articles that met the exclusion criteria.

Table 1 DATABASES AND KEYWORDS USED

| DATABASES | KEYWORDS | RESULTS |
|------------------|--|---------|
| PUBMED | ("dentigerous cyst"[All Fields] AND ("india"[MeSH Terms] OR "india"[All Fields] OR "india s"[All Fields] OR "indias"[All Fields])) AND ((casereports[Filter]) AND (child[Filter] OR adolescent[Filter])) | 46 |
| EMBASE | Dentigerous cyst | 297 |
| COCHRANE LIBRARY | Dentigerous cysts | 5 |
| ProQuest | Dentigerous cyst and india and children | 180 |

IV. QUALITY ASSESSMENT OF STUDIES

Joanna Briggs Institute (JBI) Critical Appraisal Checklist for Case Reports had been used for quality evaluation (last amended in 2017).^[9] Two authors did the quality assessment for each report individually to determine the risk of bias in

each case report, after which they were included in the review.

V. DATA COLLECTION AND SYNTHESIS

The data collection is given in the PRISMA flowchart below. (Figure 1)

Description of included studies are given in Table 2.

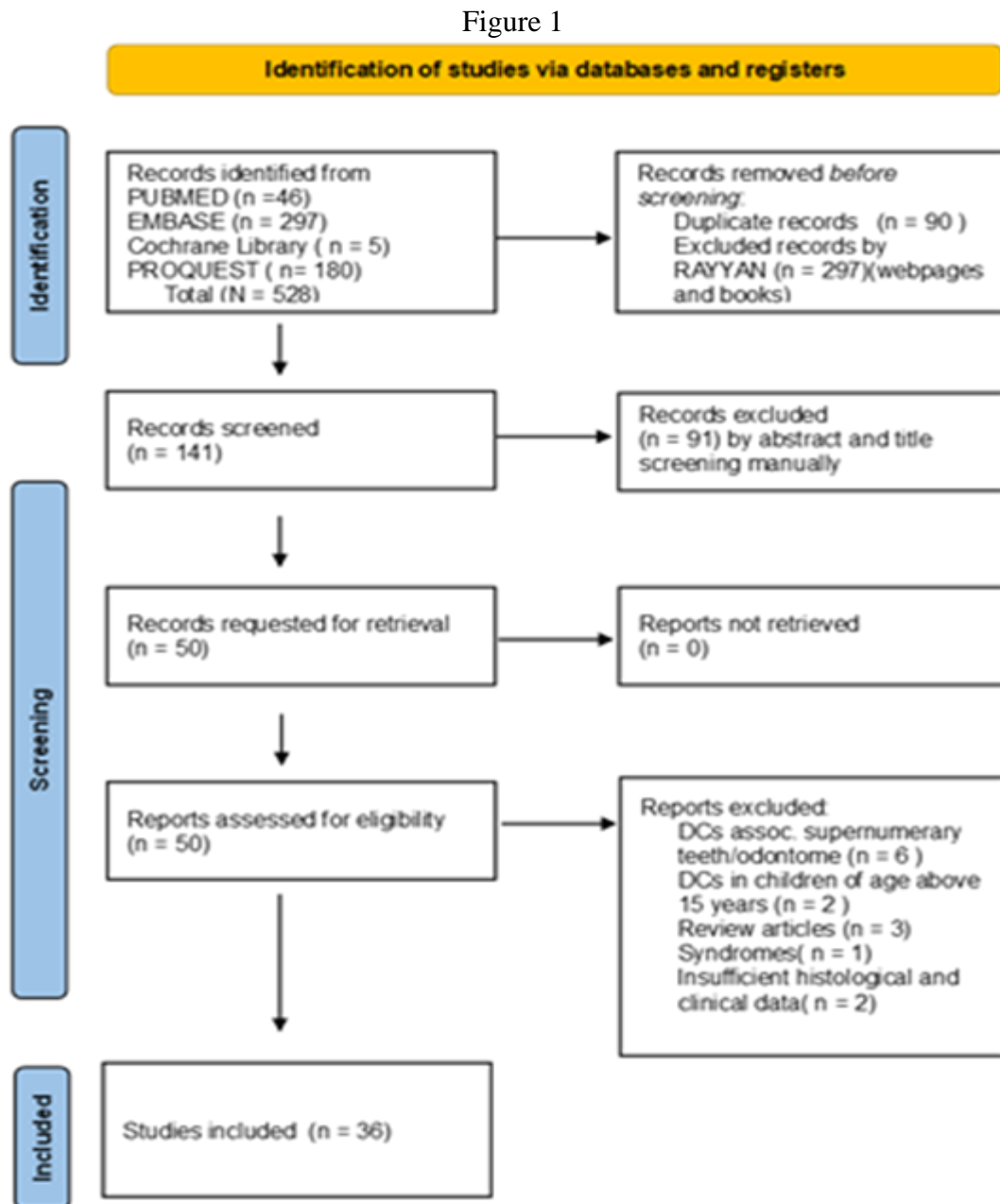


Figure 1 PRISMA FLOWCHART OF COLLECTED DATA

| AUTHOR AND YEAR | AGE/GENDER | REGION AND TEETH INVOLVED | TREATMENT DONE | REHABILITATION DONE | FOLLOW UP |
|--|------------|---------------------------|---|--|--|
| Tripathi GM et al 2014 ^[10] | 12/M | Right mandible - 43 | ENUCLEATION + extraction of 43 44 | NM (not mentioned) | 3 months – no complications |
| Desai RS et al 2005 ^[11] | 8/M | Left maxilla - 21 | ENUCLEATION + extraction of 61 62 | NM | NM |
| Premkishore K et al 2011 ^[12] | 10/M | Left mandible - 35 | ENUCLEATION + extraction of 32 33 34 35+extraction of 73 74 75 | Functional flexible removable space maintainer | 3 months – no complications |
| Passi S et al 2008 ^[13] | 10/M | Right mandible -44 45 | MARSUPIALIZATION + extraction of 83 84 85 | NM | 10 days and 1 month – No complications |
| Sardana D et al 2016 ^[14] | 8/M | Right mandible - 43 | MARSUPIALIZATION + extraction of 84 85 | Acrylic obturator | 3 months; 1 year – no complications |
| Bharath KP et al 2011 ^[15] | 9/M | Left maxilla – 21 22 | ENUCLEATION + extraction of 61 62 63 + extraction of 21 22 | Removable partial denture planned | NM |
| Pandey AK et al 2021 ^[16] | 6.5/M | Left maxilla - 21 | ENUCLEATION + extraction of 61 62+ extraction of 21 | NM | NM |
| Pant B et al 2019 ^[17] | 10/M | Maxilla bilateral – 13 23 | ENUCLEATION + extraction of primary teeth + extraction of 13 23 | NM | NM |
| Khandeparker RV et al 2018 ^[18] | 10/M | Maxilla bilateral | ENUCLEATION + extraction primary and permanent teeth | NM | NM |
| Sarangal H et al 2019 ^[19] | 10/M | Right mandible -45 | MARSUPIALIZATION + extraction of 84 85 | NM | 7 days – no complications |
| Singh S et al 2014 ^[20] | 13/M | Left mandible | MARSUPIALIZATION + extraction of 75 | NM | Up to 9 months – no complications |
| Kirtaniya BC et al 2010 ^[21] | 7/M | Left mandible - 34 | MARSUPIALIZATION + extraction of 74 | NM | 6 months |
| Kirtaniya BC et al 2010 ^[21] | 10/F | Right mandible - 44 | MARSUPIALIZATION + extraction of 83 | NM | 6 months |
| Singh M et al 2020 ^[22] | 10/M | Left maxilla 21 22 23 | MARSUPIALIZATION | Obturato | 6 months |
| Deepa KK et al 2021 ^[23] | 10/F | Right mandible -45 | ENUCLEATION + extraction of permanent teeth | NM | NM |
| Mishra R et al 2014 ^[42] | 12/F | Left mandible 35 | ENUCLEATION + extraction of 75 | NM | NM |
| Tikekar S et al 2010 ^[43] | 11/M | Mandible bilateral 45 35 | MARSUPIALIZATION + extraction of 75 85 | NM | NM |

| AUTHOR AND YEAR | AGE/GENDER | REGION AND TEETH INVOLVED | TREATMENT DONE |
|---|------------|---------------------------|---|
| Warad NM et al 2012 ^[24] | 11/M | Left maxilla 22 23 | ENUCLEATION + extraction of 23 |
| Arakeri G et al 2015 ^[25] | 13/F | Left mandible 37 | ENUCLEATION + extraction of 37 |
| Sargod SS et al 2020 ^[26] | 9/M | Right Mandible 46 | MARSUPIALIZATION + preservation of 46 |
| Shivaprakash P et al 2009 ^[27] | 10/F | Mandible Bilateral 45 35 | ENUCLEATION + extraction of deciduous teeth 75 85 |
| Suresh R et al 2011 ^[28] | 1/M | Left mandible 34 | ENUCLEATION + extraction of 34 |
| Parveen A et al 2014 ^[29] | 13/F | Left Mandible 33 | ENUCLEATION + extraction of 33 |
| Mahajan S et al 2006 ^[30] | 13/M | Mandible Bilateral 35 43 | ENUCLEATION + extraction of 35 43 |
| Lakkam B et al 2010 ^[31] | 10/M | Left mandible 35 | ENUCLEATION + extraction of deciduous and permanent |

| | | | |
|--|------|-------------------------|---|
| Tamgadge A et al 2011 ^[32] | 10/M | Maxilla bilateral 15 23 | ENUCLEATION+ extraction of permanent teeth |
| Kalaskar RR et al 2007 ^[33] | 7/M | Right maxilla 13 | ENUCLEATION + extraction of deciduous and permanent teeth |
| Gupta P et al 2016 ^[34] | 8/M | Left mandible 35 | MARSUPIALIZATION + extraction of 75 |
| Jain N et al 2018 ^[35] | 12/M | Left maxilla 24 | ENUCLEATION + extraction of 64 65 +extraction of 24 |
| Bhardwaj B et al 2016 ^[36] | 10/M | Left mandible 35 | MARSUPIALIZATION + extraction of 75 |
| Hegde RJ et al 2013 ^[37] | 9/M | Left mandible 35 | ENUCLEATION + extraction of 75 + extraction of 35 |
| Rohilla M et al 2011 ^[38] | 10/M | Left maxilla 21 | ENUCLEATION + extraction of 61+ extraction of 21 |
| Sadhvani S et al 2019 ^[39] | 10/F | Right mandible -45 | Extraction of 85 + MARSUPIALIZATION |
| Patil S et al 2017 ^[40] | 8/M | Right mandible 45 | Extraction of 85 + MARSUPIALIZATION |
| Nagarajan N et al 2021 ^[41] | 7/M | Right maxilla 11 | ENUCLEATION + extraction of 11+ extraction of 52 53 |

VI. DATA ANALYSIS

The analyzed data are given in Table 3. Write here procedure/technique of your research study.

Table 3 DATA ANALYSIS AND RESULTS

| PREVALENCE OF DENTIGEROUS CYST AMONG CHILDREN UNDER 15 YEARS OF AGE ACCORDING TO DIFFERENT AGE GROUPS | |
|--|--------------------------|
| Age groups | No of cases(n=36) |
| 0 – 5 years | 1 (2.78%) |
| 6 – 10 years | 26 (72.22%) |
| 11–15 years | 9 (25.00%) |
| Total cases | 36 |
| PREVALENCE ACCORDING TO GENDER | |
| GENDER | NO OF CASES |
| MALE | 29 (80.56%) |
| FEMALE | 7(19.44%) |
| TOTAL | 36 |
| PREVALENCE ACCORDING TO ANATOMIC SITE | |
| LOCATION | NO OF CASES |
| MAXILLA | 12(33.33%) |
| MANDIBLE | 24(66.67%) |
| TOTAL | 36 |
| PREVALENCE ACCORDING TO TEETH INVOLVED | |
| TEETH INVOLVED | NO OF CASES |
| ANTERIOR | 9 (25.00%) |
| POSTERIOR | 25 (69.44%) |
| ANTERIOR AND POSTERIOR | 2 (5.55%) |
| TOTAL | 36 |
| TREATMENT MODALITIES IN INDIA | |
| TREATMENT DONE | NO OF CASES |
| ENUCLEATION + EXTRACTION OF DECIDUOUS TEETH | 3 (8.33%) |
| ENUCLEATION + EXTRACTION OF DECIDUOUS AND AFFECTED PERMANENT TEETH | 19(52.78%) |
| MARSUPIALIZATION WITH PRESERVATION OF DECIDUOUS AND PERMANENT TEETH | 1 (2.78%) |
| MARSUPIALIZATION + EXTRACTION OF DECIDUOUS TEETH | 13 (36.11%) |
| REHABILITATION PROCEDURES | |
| ENUCLEATION ALONG WITH PERMANENT TEETH DONE | 19 |

| | |
|---|-------------|
| REHABILITATION PROCEDURES PLANNED | 11 (57.89%) |
| TYPES OF REHABILITATION PROCEDURES | |
| REMOVABLE FUNCTIONAL SPACE MAINTAINER | 6(31.58%) |
| TEMPORARY OBTURATOR | 2(10.52%) |
| ORTHODONTIC MANAGEMENT | 2(10.52%) |
| IMPLANT-SUPPORTED CROWNS | 1(5.26%) |

RESULT

A total of 36 case reports that met the inclusion and exclusion criteria and had minimal risk of bias were included in this systematic review after an extensive search of various electronic databases. After a thorough analysis of the obtained data, the prevalence of dentigerous cysts was found to be common among children aged 6–10 years (72.22%), followed by those aged 11–15 years (25.00%). It was the least common among children aged 0–5 years (2.78%). Males were found to be predisposed to dentigerous cysts, accounting for 80.56% of the cases, whereas females accounted for 19.44%. Single dentigerous cysts (83.34%) were more prevalent than multiple dentigerous cysts (16.67%). The most prevalent site in the oral cavity was the mandible posterior, followed by the anterior maxillary teeth. Enucleation and marsupialization were considered the treatment modalities for dentigerous cysts. Complete enucleation of the dentigerous cyst with the deciduous and permanent teeth was performed in 19 patients (52.78%); enucleation with the removal of deciduous dentition and preservation of the succedaneous tooth bud in 3 patients (8.33%); marsupialization with conservation of both deciduous and permanent teeth in 1 patient (2.78%); and marsupialization with the removal of primary dentition and preservation of permanent tooth bud in 13 patients (36.11%). Rehabilitation was planned only in 11 patients, in which complete enucleation of the cysts was performed. Considering the patient's age, a functional removable space maintainer was the most preferred rehabilitation procedure for better mastication efficiency. [12,37,40] Immediately after the procedure, a removable acrylic plate was given to act as an obturator. [14,15,22,33] In some cases, later,

orthodontic treatment or implant-supported crowns were planned for the missing permanent teeth. [25,26, 35]

DISCUSSION

Dentigerous cysts are the most prevalent type of developing odontogenic cyst among the pediatric population in India, occurring frequently in decades two and three of life [44-53], which is contradictory to the results of our systematic review that reported that DCs are the most common among the age group of 6–10 years as we reviewed only cases reported for the pediatric population aged 0–15 years. Dentigerous cysts have a male predilection [46,47,49,50, 53] and the results of our review are consistent with the gender predilection towards males by 80.56% of cases. Dentigerous cysts are the most prevalent in the mandibular posterior area, [49 50 52 54] which agrees with the results of our review. In contrast, some studies have stated that the maxillary anterior teeth are the most prevalent site of dentigerous cysts. [46,51,53] Bilateral dentigerous cysts are frequent in mucopolysaccharidosis (type VI) and cleidocranial dysplasia syndromic conditions. However, the bilateral involvement of dentigerous cysts has a prevalence of 16.67%, which is not associated with any syndromic conditions. [55]

If left untreated, dentigerous cysts can progress into carcinoma, including squamous cell carcinoma, mucoepidermoid carcinoma, and ameloblastoma. Primary intraosseous cancer develops in a dentigerous cyst in 16%-51% of patients, despite being a rare observation. [56] The incidence of mucoepidermoid carcinoma from dentigerous cysts constitutes approximately 42%. [57] Overall, 15%–20% of all unicystic ameloblastomas originate in dentigerous cyst margins. [58]

The management of dentigerous cysts depends on factors such as age and the anatomic site. Surgical techniques reported to date include enucleation of the cysts, marsupialization, and decompression, followed by fenestration.^[59]

Enucleation of the cyst is the most common treatment modality for surgically removing a dentigerous cyst, which carries the risk of sacrificing the developing permanent tooth buds.^[59] An alternative treatment is a marsupialization, which is considered when a large cyst is near the vital structures; the cystic lining is removed such that the cyst reduces in size gradually and the permanent tooth bud can be preserved. The only disadvantage of this procedure is the ectopic eruption of the permanent teeth, which sometimes requires orthodontic treatment.^[60] Decompression preserves developing tooth buds, alveolar bone, and nerves without significant complications. Fenestration allows decompression of the cyst while preserving the permanent teeth. Additionally, as decompression progresses, time permits ongoing root growth and partial bone fill.^[59]

Although several conservative surgical procedures are available for the removal of dentigerous cysts, enucleation of the cysts along with the affected tooth bud is preferred in 52.78% of cases. This can cause premature loss of succedaneous teeth and malocclusion. Hence, rehabilitation procedures should be planned accordingly. The various rehabilitation procedures listed in our review provide a functional space maintainer or obturator until an orthodontic therapy for aligning the teeth in the missing space or an implant-supported prosthesis is established. Considering the age of the children and the importance of maintaining the succedaneous tooth bud, pediatric dentists should work to provide a treatment plan that is less invasive and preserves the permanent tooth. Even when complete cyst enucleation and the affected dentition (permanent dentition) are required, pediatric dentists must plan the rehabilitation procedure effectively such that the

functional ability for mastication, speech, and deglutition is not hampered.

The limitations of this systematic review are that it included only observational studies, which do not have a higher level of evidence but since there are limited studies regarding the treatment modalities of Dentigerous cysts among the Pediatric population in India this would serve.

CONCLUSION

This systematic review provides a comprehensive analysis of dentigerous cysts in the pediatric population of India, focusing on prevalence, treatment modalities, and post-surgical rehabilitation. Through a meticulous review of 36 case reports, it was found that dentigerous cysts predominantly affect children aged 6-10 years, with a notable male predilection. The mandibular posterior region was identified as the most common site of occurrence. Enucleation and marsupialization were the primary treatment approaches, with a preference for enucleation despite its potential impact on succedaneous tooth buds. Rehabilitation strategies varied, with functional space maintainers and obturators being the preferred options immediately post-procedure, followed by orthodontic interventions or implant-supported prostheses for long-term dental rehabilitation.

Despite the valuable insights gained, this review acknowledges limitations inherent in the predominantly observational nature of the included studies. Nonetheless, these findings highlight the importance of tailored treatment plans that balance effective cyst removal with the preservation of dental structures crucial for oral function and development in pediatric patients. Further research employing higher-level evidence is warranted to validate these observations and optimize clinical outcomes in managing dentigerous cysts among children.

Declaration by Authors

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