

Treatment options of periapical cysts of deciduous teeth: report of a case and systematic review of the literature



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Abstract

Aims Periapical cysts of primary teeth are pathologic entities which are seldom encountered in the clinical practice. Most frequently, these lesions arise in correspondence with primary teeth presenting previous pulp therapy, severe carious lesions, or a history of previous trauma. The aim of the present study is to systematically review the treatment modalities of periapical cysts of the deciduous, along with the reporting of a clinical case.

Materials and methods A case of periapical cyst treated with marsupialization occurring in an 11-year-old patient is described. A literature search was devised to retrieve studies reporting the treatment of periapical cysts, and involved papers published in the Cochrane Oral Health Group specialist trials, MEDLINE via PubMed, and EMBASE up to March 2023. A total of 39 articles were retrieved. Following title and abstract analysis, 27 articles were selected for full-text analysis, with the final inclusion of 24 articles.

Results In total, 52 lesions were described in 50 subjects. Dental caries were more frequently associated with lesions' development, and primary molars were most frequently affected. The treatment approach was either marsupialization (8 studies) or enucleation (16 studies). Mean follow-up was 12 months. Recurrence of the lesions was not observed in any of the studies.

Conclusion Periapical cysts of primary teeth present an overall good prognosis irrespective of the treatment option adopted. The performance of a prompt diagnosis appears of utmost importance, as the extraction of the primary teeth involved implies the management of the residual space for the correct positioning of the corresponding permanent teeth.

Introduction

Radicular cysts develop in adults as a consequence of the stimulation of epithelial cell rests of Malassez following an inflammatory process. While the occurrence of such lesions has been frequently reported in permanent teeth, the occurrence of radicular cysts affecting primary teeth has been seldom reported [Nagata et al., 2008; Lustmann & Shear, 1985]. It is in fact estimated that radicular cysts account for 0.5%-3.3% of cystic lesions in primary teeth [Shear, 1985], with roughly more than

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120 cases described to date in the literature [Shetty et al., 2010]. Periapical cystic lesions in deciduous teeth most frequently occur as a consequence of extensive dental caries leading to pulp necrosis, although it appears that these lesions can also develop following pulp therapy of primary teeth [Mass et al., 1995]. In terms of histology, these lesions are similar to radicular cysts of permanent teeth, except for the presence of hyalin inclusions and the rarity of cholesterol crystal slits in the cysts of primary dentition [Mass et al., 1995]. Development is often asymptomatic, as swelling, tooth displacement and delayed eruption of permanent teeth are observed when the lesion is already well-established [Garg et al., 2022]. In some cases, diagnosis can occur in course of radiographic examinations for other purposes as an incidental finding [Bittencourt et al., 2021]. The treatment options include either enucleation or marsupialization, although always combined with the extraction of the primary tooth involved [Bodner, 2002]. While enucleation represents a more radical approach which often can cause damage or lead to the removal of the unerupted successor, marsupialization is a more conservative approach leading to the lesion's resolution with the preservation of the surrounding anatomical structures [Anavi et al., 2011].

The aim of the present study is to report a case of radicular cyst affecting an inferior primary molar and to review the cases of radicular cysts associated with primary teeth reported in the literature and the therapeutic approaches employed, in order to give insight into this uncommon pathological entity.

Case report

An 11-year-old boy was referred to the Unit of Dentistry and Oral Surgery of the University Hospital of Pisa for the development of swelling in correspondence with the lower right deciduous molars. The patient did not refer symptoms associated with the swelling. The first lower right deciduous molar had been endodontically treated two months before,

following the development of a severe carious lesion with loss of tooth vitality. A panoramic x-ray was prescribed and revealed the presence of a large osteolytic lesion with ill-defined margins involving both the deciduous molars and the gems of the premolars. In particular, the germ of the first premolar appeared dislocated apically. Cone Beam Computed Tomography was then performed, highlighting a large defect in the buccal mandibular cortex. Surgical intervention was scheduled. The deciduous molars were both removed along with the lower deciduous right canine, and the lesion marsupialised. A space retainer was positioned to protect the access cavity and to avoid tooth displacement. At 12-months follow-up, a panoramic x-ray revealed total remission of the lesion and correct positioning of the permanent premolars and canine. Clinically, complete wound closure was observed. No signs of recurrence nor symptoms were present.

Literature search: materials and methods

Search methods and eligibility criteria

The protocol for this study was developed according to the Preferred Reporting Items Systematic review and Meta-Analyses Extension Statement for Reporting of Systematic Reviews Incorporating Network Meta-analyses of Health Care Interventions [Liberati et al., 2009; Moher et al., 2009]. The following focused question was phrased: "What are the treatment options for radicular cysts of primary teeth arising following the development of dental caries, the performance of pulp therapy or trauma?" A literature search was performed to retrieve studies reporting on primary teeth radicular cysts. Only articles in English published between 2000 and 2023 were included. Review articles and systematic reviews were not included. An electronic search of the literature was performed in the Cochrane Oral Health Group specialist trials, MEDLINE via PubMed, and EMBASE up to March 2023 using a combination of MeSH terms and free text words ("Tooth, Deciduous"[Mesh]) AND "Radicular Cyst"[Mesh]. Trials databases such as clinicaltrials.gov and other relevant sites were searched. Hand search was also performed and bibliographies of all relevant papers and review articles were checked to detect additional studies.

Study selection and data collection

Two calibrated reviewers (RI and MN, κ -score > 0.8) screened the articles for possible inclusion in the review. Title and abstract analysis was performed to screen the articles retrieved from the literature search. In cases of unclear abstracts, full-text analysis was performed not to exclude any potentially relevant articles. Data from the articles included following full-text analysis were extracted and synthesized.

Statistical analysis

Data synthesis was conducted through evidence tables reporting on study characteristics and main study conclusions. The age of patients, the sites of occurrence of the lesions, the type of treatment and follow-up without signs of recurrence were registered. Data analysis was performed using IBM SPSS Statistics (Version 27).

Results

Study selection

A total of 27 citations were retrieved. Three articles were excluded following title and abstract analysis. Full-text analysis was performed on 24 articles [Takiguchi et al., 2001; Yakawa et al., 2002; Bhat et al., 2003; Smith & Cowpe, 2005; Johann et al.,

2006; Lenzi & Medeiros, 2006; Ramakrishna & Verna, 2006; Chiu et al., 2008; Gandhi et al., 2008; Nagata et al., 2008; Shetty et al., 2010; Toomarian et al., 2011; Gaynor 2012; Narsapur et al., 2012, Subramanya et al., 2012; Nagarathna et al., 2013; Uloopi et al., 2015; Mahesh et al., 2017; Sevekar et al., 2018; Tanaka et al., 2019; Truong-Nhu-Ngoc et al., 2019; Chybicki et al., 2020; Taludakar et al., 2020; Pei et al., 2022] (Figure 1). A kappa score >0.8 was obtained among the reviewers in all the phases of title and abstract selection, full-text analysis, and data extraction.

Population characteristics

The study population consisted of 52 lesions described in 50 subjects (15 females). Mean age was 7.95 (SD 2.44). The lesions were located predominantly in the mandible (40 lesions versus 12 in the maxilla). In 27 cases the lesions were associated to dental caries, in 17 cases pulp therapy of the deciduous tooth was present, while in 7 cases the development of the lesion was subsequent to a trauma. Mean follow-up was 12.67 months (SD 10.03).

Lesions treated with marsupialization

Eight studies described 35 lesions occurring in 33 patients and treated with marsupialization. In 28 cases the lesion involved the mandible. The lesion was most frequently localized in correspondence with the primary molars (85.71%), followed by incisors (4 cases), while only one case was reported to involve the canine. Mean patient age was 8.54 years (SD 2.51 years). Mean follow-up was 15.5 months (SD 11.56 years) (Table 1).

Yawaka et al. [2002] described a case of periapical cyst occurring in a 13-year-old girl and associated to a mandibular primary molar affected by recurrent carious lesions. The treatment included the extraction of the primary molar and the marsupialization of the lesion. The repositioning of the impacted permanent premolar was obtained through the application of a traction for two years following surgery.

Johann et al. [2006] reported a case occurring in a 9-year-old boy. The patient developed the lesion in correspondence with a residual root stump of the primary molar, which was extracted and the lesion marsupialised. In this case, the authors described recurrence of the lesions ten months after initial treatment due to a lack of compliance in the follow-up, and the re-insertion of drainage was required for additional four months, eventually leading to lesion's resolution.

Lenzi & Medeiros [2006] treated with marsupialization a radicular cyst occurring in correspondence with the upper right deciduous incisor in a 5-year-old boy. The patient had experienced trauma and displacement of the incisor two years before. The treatment involved primary incisor extraction, lesion exposure, and application of periodontal dressing for 10 days. At two years, correct positioning of tooth 1.1 was observed, in the absence of signs of recurrence.

Nagarathna et al. [2013] described a case of radicular cyst in a 8-year-old boy. The lesion involved a mandibular primary molar with incomplete pulp therapy, which was treated with extraction of the primary molars and lesion's marsupialization. The post-operative period was uneventful, and resolution was observed at six months.

Uloopi et al. [2015] described four cases of cystic lesions associated to primary teeth, with various etiology. Three cases affected two 11-year-old boys and one 8-year-old boy at the level of primary maxillary incisors following a history of trauma. In all the cases, treatment included extraction of the deciduous incisors and lesion's marsupialization, followed by orthodontic treatment for permanent teeth repositioning. The fourth case of a 10-year-

Author	Year	Patients characteristics	No. of lesions	Site	History	Follow-up
Yawaka et al.13	2002	1 female patient, 13 years of age	1	Mandibular deciduous molar	Dental caries	24 months
Johann et al. 14	2006	1 male patient, 9 years of age	1	Maxillary deciduous molar	Dental caries	14 months
Lenzi et al.15	2006	1 male patient, 5 years of age	1	Maxillary deciduous incisor	Trauma	24 months
Nagarathna et al.16	2013	1 male patient, 8 years of age	1	Mandibular deciduous molar	Pulp therapy	6 months
Uloopi et al.17	2015	3 male patients, aged 11 (2 patients) and 8 (1 patient) 1 female patient, 10 years of age	4	Maxillary deciduous incisor (3 male patients) Mandibular deciduous molar (1 female patient)	Trauma (for the lesions involving the incisors) Dental caries (for the lesion involving the primary molar)	12 months
Tanaka et al.18	2019	1 male patient, , 9 years of age	1	Maxillary deciduous molar	Dental caries	36 months
Truong-Nhu-Ngoc et al.19	2019	1 female patient, 5 years of age	1	Maxillary deciduous molar	Pulp therapy	3 months
Pei et al. 20	2022	13 male patients, 10 female patients, mean age 7.9 years	25	24 mandibular deciduous molars, 1 mandibular canine	Pulp therapy in 9 cases	5 months

TABLE 1 List of papers reporting lesions’ treatment with marsupialization

old girl described a lesion involving a mandibular decayed primary molar, which was treated with extraction of the involved deciduous tooth and marsupialization and the application of a space maintainer for the guidance of premolar eruption.

Tanaka et al. [2019] described the development of a large periapical cyst in correspondence with the severely decayed maxillary primary molars. Due to the dimensions of the lesion, primary molars and first permanent molar were extracted, and the lesion marsupialised. At three years follow-up no signs of recurrence were observed, and optimal occlusion was achieved through orthodontic treatment.

Truong-Nhu-Ngoc et al. [2019] reported a case in a 5-year-old girl that occurred following a pulp therapy of a maxillary primary molar with the application of non-resorbable material. The deciduous molar was removed and the lesion marsupialised,

along with the application of an acrylic obturator.

Pei et al. [2022] retrospectively analysed 25 cases of radicular cysts occurring in 23 patients. The lesions were all localized in the mandible and mostly involved the primary molars, as only one case was described in correspondence with the canine. Treatment involved tooth extraction and decompression, with regular follow-up once a month until complete healing, with a mean decompression duration of 5 months.

Lesions treated with enucleation

Sixteen studies described 17 lesions treated with enucleation occurring in 17 patients. In 12 cases the lesion involved the mandible. The lesion was most frequently localized in correspondence with the primary molars (82.35%), followed by incisors. The mean patient age was 7.31 years (SD 2.39 years).

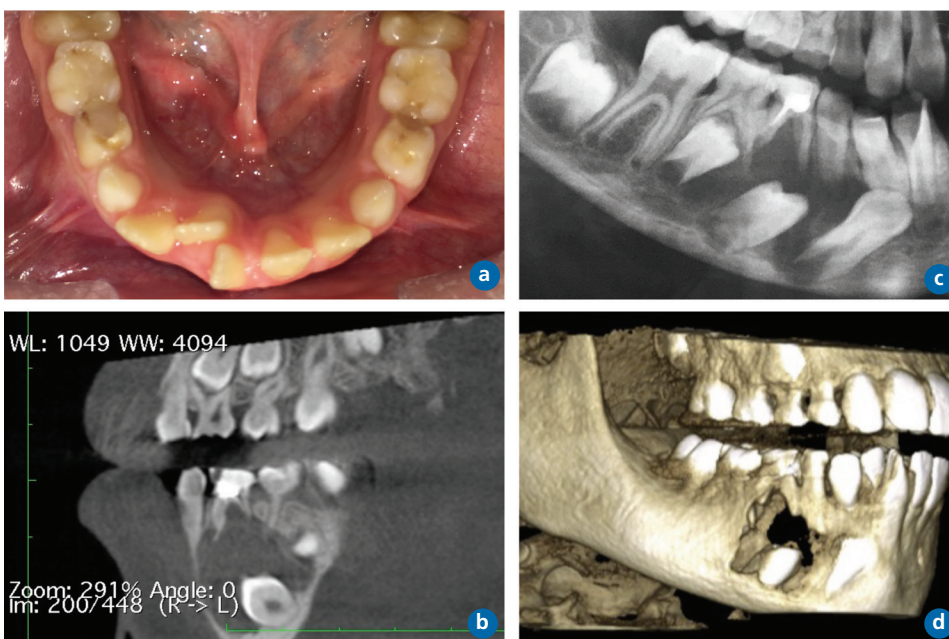


FIG. 1 preoperative evaluation of the patient. A) Clinical photograph showing swelling of the mandibular buccal cortical bone in correspondence with the lower right primary molars; B) Panoramic radiograph showing a large osteolytic area involving the first primary molar and causing displacement of the germs of the premolars; C) CBCT scan of the lesion; D) 3D volume rendering showing a large fenestration of the mandibular buccal cortical bone.

Author	Year	Patients characteristics	No. of lesions	Affected tooth	History	Follow-up
Tagikuchi et al.21	2001	1 male patient, 7.5 years of age	1	Mandibular deciduous molar	Pulp therapy	6 months
Bhat et al.22	2003	1 male patient, 9 years of age	1	Mandibular deciduous molar	Pulp therapy	Not reported
Smith & Cowpe23	2005	1 male patient, 3 years of age	1	Maxillary incisor	Trauma	2 weeks
Ramakrishna & Verma24	2006	1 male patient, 9 years of age	1	Mandibular deciduous molar	Pulp therapy	Not reported
Chiu et al.25	2007	2 male patients, 10.5 mean age	2	Mandibular deciduous molar	Pulp therapy	24 months
Gandhi & Franklin26	2008	1 female patient, 9 years of age	1	Mandibular deciduous molar	Pulp therapy	12 months
Nagata et al.27	2008	1 male patient, 6 years of age	1	Mandibular deciduous molar	Dental caries	1 month
Shetty et al.4	2010	1 male patient, 11 years of age	1	Maxillary deciduous molar	Dental caries	Not reported
Toomarian et al.28	2011	1 male patient, 5 years of age	1	Mandibular deciduous molar	Pulp therapy	20 months
Gaynor29	2012	1 male patient, 8 years of age	1	Mandibular deciduous molar	Dental caries	12 months
Narsapur et al.30	2012	1 male patient, 9 years of age	1	Mandibular deciduous molar	Dental caries	Not reported
Subramanya31	2012	1 male patient, 8 years of age	1	Maxillary incisor	Trauma	Not reported
Mahesh et al.32	2017	1 male patient, 7 years of age	1	Maxillary incisor	Trauma	Not reported
Sevekar et al.33	2018	1 male patient, 3 years of age	1	Mandibular deciduous molar	Dental caries	12 months
Chybicki et al.34	2020	1 male patient, 5 years of age	1	Maxillary deciduous molar	Dental caries	1 month
Taludakar et al.35	2020	1 male patient, 7 years of age	1	Mandibular deciduous molar	Dental caries	Not reported

TABLE 2 List of papers reporting lesions' treatment with enucleation

The mean follow-up was 9.83 months (SD 8.49 months) (Table 2). Tagikuchi et al. [2001] described a case of a 7.5-year-old boy developing a radicular cyst in correspondence with the mandibular left primary second molar. The tooth had been treated with pulp therapy 17 months prior to lesion diagnosis. The patient underwent tooth extraction and lesion enucleation. At 6-month follow-up, complete healing was observed. Bhat et al. [2003] reported a case in a 9-year-old boy involving the mandibular primary molar treated with pulp therapy. The lesion caused displacement of the permanent successor and expansion of the buccal cortical bone. The lesion was surgically removed along with the extraction of the involved tooth under general anesthesia. Smith & Cowpe [2005] described the case of a 3-year-old boy developing a swelling of the premaxilla which arose in the previous 4 months. The patient had a history of trauma on the upper primary incisors at 1.5 years of age. The occlusal radiograph revealed the presence of a large osteolytic lesion, which was enucleated along with the involved deciduous teeth. At two weeks complete healing was observed, although no further follow-up was performed.

Ramakrishna & Verma [2006] reported a case of radicular cyst localized in the mandible in correspondence with right primary molars, which occurred in a 7-year-old girl. The lesion was treated with enucleation, with the positioning of an intraoral drainage.

Chiu et al. [2008] described two cases of radicular cysts of the mandibular primary molars occurring in two male patients aged 10 and 11 years, respectively. The first patient developed the lesion in correspondence with the mandibular right primary molar treated with pulpectomy and filled with zinc oxide eugenol. The lesion was enucleated along with the involved tooth, and at 9-month follow-up complete eruption of the premolars was observed. The second case involved the left mandible with displacement of the gems of the permanent canine and premolars. The lesion was localized in correspondence with the left primary mandibular molars, which had been treated with formocresol pulpotomy and restored with intermediate restorative material

(IRM). At two years, alignment of the permanent canine and premolars was observed. Gandhi & Franklin [2008] described a case of a 9-year-old girl developing a periapical cystic lesion of a previously endodontically treated mandibular deciduous molar. The lesion was removed along with the involved primary tooth. Follow-up x-rays at 12 months showed correct positioning of the corresponding premolar.

Nagata et al. [2008] reported a case in a 6-year-old boy affecting the mandibular right primary second molar. Treatment involved primary molar extraction along with the lesion. The patient was then lost to follow-up after one month.

Shetty et al. [2010] described a case occurring in correspondence with the maxillary primary molars, which both presented severe carious lesions. the lesion was enucleated along with the primary teeth, while the permanent premolars were preserved. No data on follow-up were reported by the authors.

In the case described by Toomarian et al. [2011] in a 5-year-old boy, the cystic lesion developed in correspondence with the mandibular left primary first molar treated with pulpotomy one year before. The lesion was enucleated and the involved primary molar was removed. At 20 months follow-up, no signs of recurrence were observed.

Gaynor [2012] reported a case of bilateral radicular cyst occurring in an 8-year-old boy which involved the mandibular second primary molars. In the right mandible, both the primary molars were extracted with the cyst. In the left mandible, only the second primary molar was removed. At 12 months, complete healing was observed in both surgical sites.

Narsapur et al. [2012] described a radicular cyst arising periapically to the mandibular left primary second molar which presented a severe carious lesion. The treatment involved cyst enucleation and primary molar removal. No data on follow-up were reported.

The case reported by Subramanya et al. [2012] referred to an 8-year-old boy presenting a cystic lesion periapical to the maxillary primary incisor, which presented a history of trauma. Primary

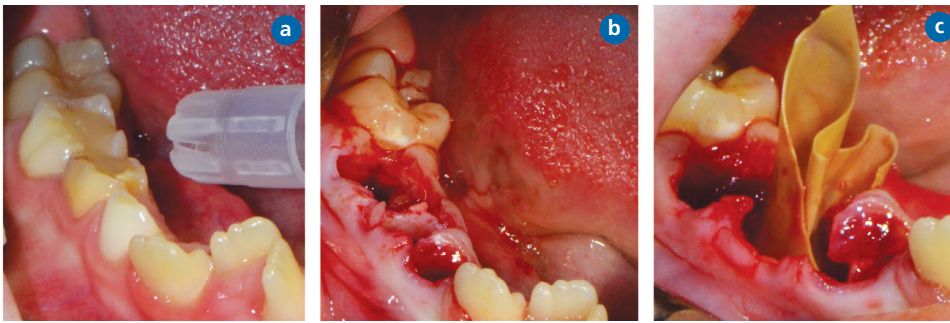


FIG. 2
Surgical intervention. A) Preoperative photograph; B) Extraction of the primary molars; C) Drainage positioning.

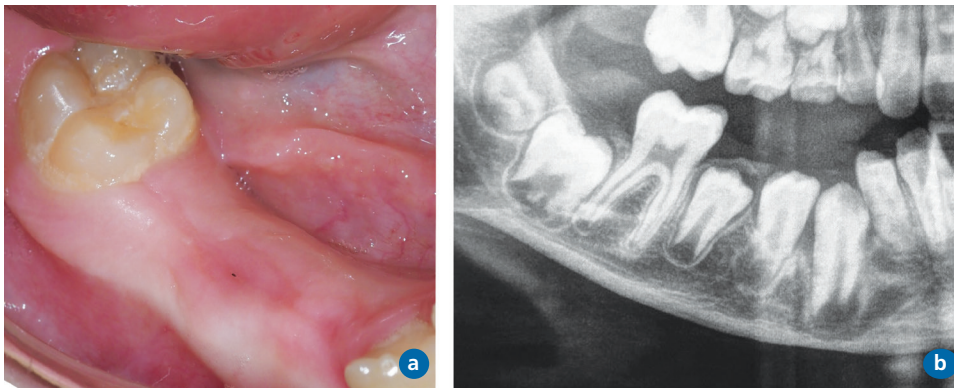


FIG. 3
Follow-up evaluation. A) Complete healing of the surgical site can be observed. B) Panoramic radiograph showing correct positioning of the germs of the premolars in the absence of signs of recurrence.

incisor extraction and enucleation of the cyst were performed.

Mahesh et al. [2017] reported a case involving the primary maxillary right incisor in a 7-year-old boy with a previous history of trauma of the premaxillary region. Treatment included enucleation and primary incisor extraction, with preservation of the germ of the permanent right incisor.

In the case by Sevekar et al. [2018] the development of a radicular cyst periapical to a severely decayed mandibular second primary molar in a 5-year-old boy was described. The primary tooth was removed with the lesion, and a space maintainer was positioned to avoid the mesial inclination of the permanent first molar. At 3.5 years follow-up, correct positioning of the second premolar was observed.

Chybicki et al. [2020] described a case developing in correspondence with the right maxillary primary molars, which had both been treated for carious lesions and did not respond to vitality tests. The teeth involved were both removed along with the cystic lesion. At three weeks, complete wound healing was observed. No further follow-up was performed.

Taludakar et al. [2020] reported the case of a 7-year-old boy developing a radicular cyst periapical to the decayed left mandibular first primary molar. The lesion was enucleated with the primary molar and the corresponding permanent premolar. No data on follow-up after the first week post-operative were reported.

Discussion

The present review highlights the importance of correct diagnosis of radicular cysts of primary dentition, as these progressively enlarging lesions may also affect permanent teeth positioning. It appears noteworthy that collecting patient history is of utmost importance in order to retrieve information on previous treatments of the deciduous teeth, as well as identifying previous history of trauma especially when incisors are involved. The surgical approach is the treatment of choice, either through

marsupialization or enucleation. Both techniques appear effective in the management of these lesions, as in none of the studies included in the present review lesions recurrence was observed, suggesting an overall good prognosis. In the last 20 years, several cases have been described in the literature, suggesting an increasing occurrence probably due to the higher number of dental procedures performed even at an early age.

It should be noted that trauma represented the cause only in a minority of cases. Most frequently, severe carious lesions and previous pulp therapy could be observed in relationship to radicular cysts. This fact calls for an increasing attention of the clinicians when performing pulp therapy of deciduous teeth, as monitoring the treatment in order to identify early signs of periapical osteolysis appears of utmost importance in both preventing and providing early treatment of these cysts [Schwendicke et al., 2021]. Early detection is crucial as in most cases permanent tooth germs may show displacement following lesion enlargement. This fact accounts also for the need for orthodontic treatment following lesion removal and tooth extraction, as the positioning of space maintainers is often needed to avoid the migration of the surrounding teeth while waiting for the eruption of the permanent teeth [Wagner et al., 1999; Yao et al., 2015]. Indeed, it must be also kept in mind that dental caries have been demonstrated to have a negative impact on masticatory performance and oral health-related quality of life in children, affecting both emotional and social well-being [Kaya et al., 2017; Martins et al., 2018]. Indeed, timely intervention and management of caries in children appear of utmost importance in the prevention of further complications and in reducing the invasiveness of treatment [Hazar Bodrumlu et al., 2018].

However, the clinical diagnosis is performed only when symptoms, such as swelling and pain, have already developed. Diagnostic imaging may be of support in identifying the teeth involved, the margins of the lesion, and the positioning of the germs of the underlying permanent elements. Among the most commonly employed techniques, a panoramic x-ray is the most

suitable for the initial investigation of the lesion. Panoramic x-ray has several advantages, including reduced radiation dose, the possibility to obtain a comprehensive view of the maxilla and mandible, and relative ease of performance [Izzetti et al., 2021]. Panoramic radiography is routinely employed in children to evaluate the presence of dental anomalies, including abnormalities in the size and number of teeth, and eruption pattern disruptions [Wagner et al., 2020]. Importantly, diagnostic capacity is strictly dependent on the quality of the panoramic examination, which should be balanced between the diagnostic need of the clinician and the radiation dose exposure to the patient [Wagner et al., 2020].

In cases of lesions of greater dimensions, three-dimensional imaging may be necessary to clarify the actual extent of the lesion, its margins, and its relationship with the surrounding anatomical structures [Izzetti et al., 2020]. From this perspective, cone beam computed tomography (CBCT) appears to be the most effective technique, especially considering that paediatric patients are involved. CBCT allows high spatial resolution imaging at lower radiation doses compared to multi-slice computed tomography. Importantly, the scan may be collimated only to the area of interest, further reducing the radiation dose to the patient, which becomes crucial when relating to X-ray exposure in paediatric patients [Izzetti et al., 2022]. Moreover, the possibility to perform post-processing of volumetric data may improve the pre-surgical planning [Izzetti et al., 2019]. Indeed, it should be always borne in mind that diagnostic imaging prescription should always be subjected to a careful evaluation in order to guarantee exposure to the lowest radiation possible [Pauwels et al., 2015]. As a matter of fact, studies on phantoms highlight that the effective doses

of a CBCT examination range between 79 and 116 mSv, which are similar to adult doses, although with considerably higher attributable lifetime mortality risk in the long term [Theodorakou et al., 2012]. Indeed, the performance of 3D diagnostic imaging should be always justified in children and adolescents, but most importantly the acquisition parameters should be optimized to reduce to a minimum the radiation exposure [Paglia et al., 2022].

Although the diagnosis of these lesions appears relatively straightforward, differential diagnosis should always be performed, as well as histopathology of the lesion removed. Other diagnoses may include dentigerous cysts, odontogenic tumours or even less common conditions [Ferrazzano et al., 2018; Izzetti et al., 2019]. However, radicular cysts are generally associated with either treated or decayed primary teeth, or the patient refers to a history of trauma.

Concluding, radicular cysts of primary teeth are pathologic entities which should be promptly recognized by the clinician, in order to provide adequate treatment. Apart from the therapeutic approach chosen between enucleation and marsupialization, the extraction of the primary teeth involved should be always performed, although requiring subsequent management by the orthodontist to maintain the spaces and favour the correct positioning of the corresponding permanent teeth in the dental arch.

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