Systematic Review

What is the survival rate of deciduous molars in cases with agenesis of premolar successors?
A systematic review

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ABSTRACT

Objectives: To systematically review the literature on the survival rate of deciduous molars in cases of agenesis of premolar successors.

Materials and Methods: Four electronic databases and partial grey literature were searched up to November 2020. The PECOS eligibility criteria included (P) second deciduous molar (E) exposed to agenesis of a premolar successor (O) evaluated by the survival rate in the oral cavity, infraocclusion, and root resorption through (S) observational studies. Risk of bias (RoB) was assessed using the checklists from the Joanna Briggs Institute and the level of evidence was assessed using the GRADE (Grading of Recommendations, Assessment, Development and Evaluations) tool.

Results: Three studies were included: one with low, one with moderate, and one with high RoB. Synthesis methods included the frequency of persistent deciduous second molars during the follow-up. Approximately 82% to 89% remained in the oral cavity after 5 to 13 years. The incidence of root resorption was 11%, and the infraocclusion was 1 mm. The level of evidence was considered low for each outcome. There was considerable RoB regarding the observational studies and a need for clinical and radiographic monitoring of the deciduous molars.

Conclusions: Maintaining a deciduous molar in the oral cavity in patients with agenesis of the premolar successor is a viable clinical choice since 82% to 89% of the retained molars evaluated were in good condition over a follow-up ranging from 5 to 13 years. Infraocclusion and root resorption did not seem to increase considerably. The level of evidence was considered low for each outcome. (*Angle Orthod.* 2022;92:110–117.)

KEY WORDS: Deciduous tooth; Anodontia; Survival rate; Root resorption

INTRODUCTION

The prevalence of premolar agenesis ranges from 1.5% to 3.1%, and patients with this condition have routinely need orthodontic treatment and high-cost future interventions.[1] The diagnosis is typically established when the patient is 8 to 10 years old,[2] and treatment must consider the individual's level of growth and development,[3] the integrity of the deciduous molar,[4] the type of malocclusion and facial profile,[5] and the duration of treatment required.[6]

Possible therapies include dental implants,[7] extraction[8] or controlled slicing[9] of the deciduous molar, orthodontic space closure,[5] dental autotransplantation,[3] and maintenance of the deciduous molar.[4] The failure rate in rehabilitation cases of agenesis was reported in a systematic review to be 3.3% for implants, 1% for autotransplantations, and 0.9% for maintenance of deciduous molars in a 2- to 3-year follow-up period.[10]

Keeping deciduous molars is a low cost and less invasive option that postpones the need for implants or orthodontic space closure[8,11] depending on the condi-

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Table 1. Search Strategies in the Database

Database	Keywords	Result
Pubmed	((((((((Tooth, Deciduous[MeSH Terms]) OR (Premolar[Title/Abstract])) OR (Deciduous Tooth[Title/Abstract])) OR (Primary retention of deciduous teeth[Title/Abstract])) OR (Dentition*, Deciduous[Title/Abstract])) OR (Deciduous molar*[Title/Abstract])) OR (Primary molar*[Title/Abstract])) OR (Retained deciduous[Title/Abstract])) AND (((((((Anodontia[MeSH Terms]) OR (Tooth Abnormalities[MeSH Terms])) OR (Tooth Agenesis, familial[Title/Abstract])) OR (Hypodontia[Title/Abstract])) OR (Tooth Abnormality[Title/Abstract])) OR (Teeth Abnormalities[Title/Abstract])) AND (((((((Survival Rate[MeSH Terms]) OR (Prognosis[MeSH Terms])) OR (time[MeSH Terms])) OR (survival rates[Title/Abstract])) OR (Mean Survival Time[Title/Abstract])) OR (Long-term[Title/Abstract])) OR (Longterm effects[Title/Abstract]))	168
Scopus	(TITLE-ABS-KEY (“Tooth, Deciduous” OR “Premolar” OR “Deciduous Tooth” OR “Primary retention of deciduous teeth” OR “Dentition*, Deciduous” OR “Deciduous molar*” OR “Primary molar*” OR “Retained deciduous”)) AND (TITLE-ABS-KEY (anodontia OR “tooth agenesis”)) AND (TITLE-ABS-KEY (“tooth impacted” OR “survival rate*” OR “prognosis” OR “survival time”))	105
Web of Science	TOPIC (“Tooth, Deciduous” OR “Premolar” OR “Deciduous Tooth” OR “Primary retention of deciduous teeth” OR “Dentition*, Deciduous” OR “Deciduous molar*” OR “Primary molar*” OR “Retained deciduous” AND TOPIC (“tooth impacted” OR “survival rate*” OR “prognosis” OR “survival time”)	379
LILACS	(“diente primario” OR “dente decíduo” “molar deciduo” OR “premolar” OR “Pré molar”) AND (“Taxa de sobrevivência” OR “taxa de sobrevida” OR “tasa de supervivencia” OR “anodoncia” OR “agenesia”)	139
Google Scholar	allintitle: “primary molar” OR “agenesis”	200
Open Grey	Primary molar	17

tion of root resorption, infraocclusion, dental caries, and periodontal disease.[4,11] Root resorption progression tends to decrease with age in retained teeth and stabilizes after the third decade of life.[4] Infraocclusion is often associated with ankylosis,[12] and it is estimated that progression is slow after the growth phase.

The orthodontist must know the treatment options and limitations, especially regarding keeping the primary molars, in terms of clinical viability, low cost, and good patient tolerance. This review was undertaken to summarize the information available regarding the survival of deciduous molars in cases of agenesis of premolar successors and possible complications related to root resorption and infraocclusion.

MATERIALS AND METHODS

Protocol and Registration

This systematic review was registered in the PROSPERO database (https://www.crd.york.ac.uk/prospero/#myprospero) under protocol ID CRD42020182851, and it was carried out according to the PRISMA guidelines (http://prisma-statement.org/).

Eligibility and Inclusion Criteria

Observational studies that assessed the survival rate of second deciduous molars in cases of agenesis of premolar successors were included. No restrictions on the publication period or language were applied. The population sample included both sexes, and there were no restrictions in terms of age, growth stage, or type of malocclusion.

Exclusion Criteria

Studies in patients with systemic diseases, craniofacial malformations, premature loss of deciduous molars and previous orthodontic treatment were excluded. Also excluded were studies that did not correspond to the PECOS as a guide strategy: (P) Population: deciduous molars; (E) Exposure: agenesis of premolar successor; (C) Comparison: not applicable; (O) Outcome: the primary outcome was survival time in the oral cavity; the secondary outcomes were the incidence of root resorption and infraocclusion; (S) Studies: observational studies.

Information Sources

Searches were conducted in the PubMed, LILACS, Scopus, Web of Science, and partial grey literature (eg, Open Grey and Google Scholar) databases. The search strategies are shown in Table 1 and included studies were published before November 10, 2020. The references of the included studies were searched manually. An alert was created for new studies compatible with the search in the databases.

Study Selection

Two independent examiners (CS and DM) performed the searches. In cases of disagreement, a third examiner (DN) was consulted. The search strategy was developed from a combination of the National Library of Medicine’s medical subject headings (MeSH), entry terms, and key words related to the PECOS strategy using Boolean operators. The selected articles were exported to a reference manager (EndNote, Clarivate Analytics, Philadelphia, Pa) to remove dupli-

Table 2. Summary of the Data From Included Studies[a]

1. Authorship			2. Material				
Author	Country, Publication Year	Study Design	Sample Size (n)	Male/ Female	Mean Age at T0 (y)	Tooth (n), Location Mx/Md	Follow-up (TF–T0)
Bjerklin et al[4]	Sweden, 2008	Observational prospective	99	37/62	12 to 13	149 Md	13 y
Ith-Hansen et al[16]	Denmark, 2000	Retrospective observational	18	13/13	Not included	26 Mx: 7 Md: 19	Mixed dentition until 16 y
Rune et al[17]	Sweden, 1984	Observational prospective	77	29/48	11	123 Mx: 19 Md: 104	M: 5.3 y F: 5.4 y

[4] Abbreviations: F indicates Female; M, Male; Md, mandibular; Mx, maxillary; RR, root resorption.

cates and to exclude those that did not meet the pre-established inclusion criteria. Finally, the relevant articles were read for final selection, and a third examiner (DN) was consulted to resolve discrepancies.

Data Collection Process and Summary Measures

The same reviewers performed data extraction independently. Data were collected based on the following items: authorship, including author names, country, year of publication, and study design; sample characteristics, including sample size, distribution by sex and age, number of teeth and location, and follow-up; exposure in terms of the tooth affected by successor agenesis; and results, including incidence or prevalence of infraocclusion and root resorption, mean survival of deciduous molar, and conclusions (Table 2).

Risk of Bias in Individual Studies

The analysis of the risk of bias was carried out through the checklists for critical evaluation from the Joanna Briggs Institute for Quasi-experimental studies.[13] The critical analysis includes the completion of checklists with nine questions with the answers “yes”, “no”, “not clear”, and “not applicable”. The evaluators agreed on the scoring criteria before conducting the critical analysis. Thus, the studies were characterized as high risk of bias (RoB) when up to 49% of the answers were “yes”, moderate risk when 50% to 69% of the answers were “yes”, and low when more than 70% of the answers were “yes”, regardless of the question asked. Two examiners independently evaluated RoB of the selected studies (CS and DM) and, in case of discrepancies, a third examiner was consulted (DN).

Synthesis of Results

The studies described frequency as the percentage of persistent second deciduous molars during the clinical follow-up, performed in populations with different age groups and follow-ups. This would have added heterogeneity to the meta-analysis and generated unreliable information. Therefore, a meta-analysis was not performed.

Level of Evidence

The outcomes evaluated using the GRADE (Grading of Recommendations, Assessment, Development and Evaluations)[14] tool were classified based on the survival rate of deciduous molars in cases of agenesis of premolar successors, root resorption, and infraocclusion. The studies were evaluated based on study design, RoB, inconsistency, indirect evidence, and imprecision.

RESULTS

Study Selection

A total of 791 references were found in the PubMed (168), LILACS (139), Scopus (105), and Web of Science (379) databases. After removing duplicate references in EndNote manager, 740 articles remained. Six potential studies remained after title and abstract screening, and grey literature and reference list searches. Applying the eligibility criteria, three studies were excluded due to absence of relevant data.[8,11,15] Three studies were selected for qualitative analysis.[4,16,17] The process of identifying, selecting, and excluding studies is shown in the PRISMA flow chart (Figure 1).

Study Characteristics

Three observational studies were included: two prospective[4,17] and one retrospective.[16] The mean age of the samples at the beginning of follow-up were 11 year[17] and 13 years.[4] One study did not report these data.[16] The mean follow-up periods were 5 years,[17] 10

Table 2. Extended

3. Exposure	4. Results			
Tooth Affected by Successor Agenesis	Incidence or Prevalence of Infraocclusion (mm or %)	Incidence or Prevalence of RR (mm or %)	Mean Survival of Deciduous Molar	5. Conclusion
Lower deciduous second molars	Incidence: 1.00 mm	44% unchanged	89%	Survival: 89% after 13 y Infraocclusion: incidence: 1 mm RR: 44% unchanged
Deciduous second molars	Incidence: 11%	Incidence: 11%	88.46%	Survival: 88.46% until 16 y Infraocclusion: incidence: 11% RR incidence: 11%
Deciduous second molars	70% unchanged (Md)	51.92% unchanged (Md)	82.92%	Survival: 82.92% after 5 y Infraocclusion: 70% unchanged RR incidence: 51.92% unchanged

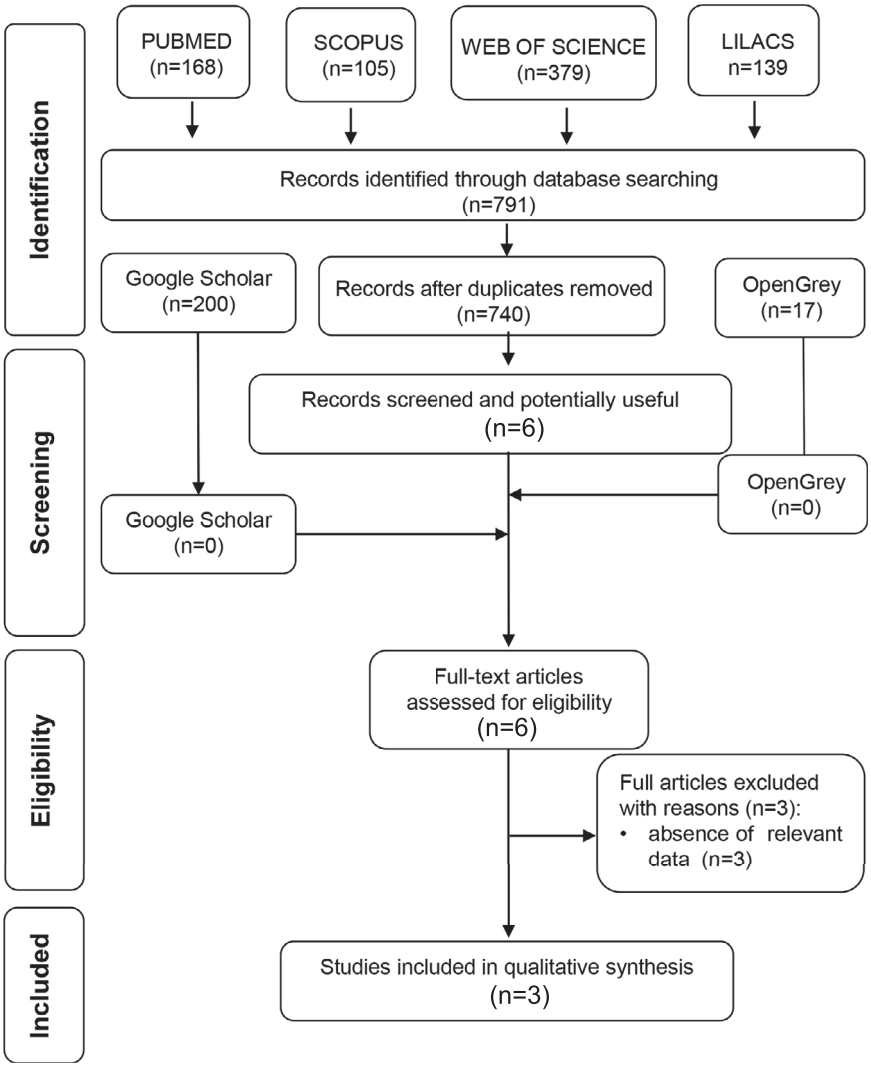

Figure 1. PRISMA flow diagram of article retrieval.

Table 3. Risk of Bias of Included Studies Evaluated by JBI Critical Appraisal Checklist for Quasi-Experimental Studies[a]

Question	Bjerklin et al. 2008[4]	Ith-Hansen et al. 2000[16]	Rune et al. 1984[17]
1. Is it clear in the study what is the 'cause' and what is the 'effect' (ie, there is no confusion about which variable comes first)?	Y	Y	Y
2. Were the participants included in any comparisons similar?	Y	Y	Y
3. Were the participants included in any comparisons receiving similar treatment/care, other than the exposure or intervention of interest?	Y	Y	Y
4. Was there a control group?	N	N	N
5. Were there multiple measurements of the outcome both before and after the intervention/exposure?	Y	N	N
6. Was follow-up complete and, if not, were differences between groups in terms of their follow-up adequately described and analyzed?	Y	N	Y
7. Were the outcomes of participants included in any comparisons measured in the same way?	Y	Y	U
8. Were outcomes measured in a reliable way?	Y	U	Y
9. Was appropriate statistical analysis used?	Y	N	N
Total	88.8%	44.4%	55.5%
Risk of bias	Low	High	Moderate

[a] N indicates no; U, undecided; Y, yes.

years,[16] and 13 years.[4] The smallest study sample included 18 individuals,[16] followed by 77 participants[17] and 99 participants.[4]

Results of Individual Studies

Three case series studies evaluated the survival rate of second deciduous molars with premolar agenesis and a prognosis related to infraocclusion and root resorption.[4,16,17] One prospective study with moderate RoB and a sample of 77 individuals with an initial age of 11 years reported a survival rate of 82.9% with a follow-up of 5 years.[4] A survival rate of 89% was reported for the low RoB study with an average follow-up of 13 years.[17] One high RoB retrospective study spanning approximately 10 years with a sample of 18 individuals reported survival rates of 88.4% for deciduous molars.[16]

The levels of infraocclusion and root resorption for the second deciduous molars were evaluated before and after orthodontic treatment using periapical radiographs[16,17] associated with bitewings.[4] Infraocclusion was confirmed when the deciduous molar did not reach the crown height of the adjacent tooth[17] or through measurement, in millimeters, from the occlusal plane to the occlusal surface of the deciduous molars.[4] One study did not report the evaluation method for infraocclusion.[16] The level of infraoccluson was 1 mm,[4] affecting 10%[12] to 11%[16] of the evaluated teeth. One study reported no changes related to infraocclusion concerning 70% of the teeth.[17] Two studies assessed root resorption on scales of 4 points[17] or 6 points.[4] One study reported the incidence of root resorption as 11% but did not report the evaluation method.[16] Two studies reported no changes in the level of root resorption in 44%[4] and 51.92%[17] of the samples.

Risk of Bias Within Studies

In the RoB analysis, one study was found to have low,[4] one moderate[17] and one high RoB.[16] The studies showed weakness in statistical analysis regarding the absence of a sample calculation,[17] a statistical regression model to adjust for confounding factors and a control group,[16,17] and in the initial data related to the root resorption and infraocclusion.[17] The high RoB study[16] did not describe how infraocclusion and root resorption were measured (Table 3).

Assessment of the Certainty of Evidence

The evidence that 82% to 89% of second deciduous molars with premolar successor agenesis remain in good condition in the oral cavity during a follow-up from 5 to 13 years was considered low. Also, infraocclusion and root resorption did not seem to affect the longevity of the deciduous molar based on low evidence (Table 4).

Observational studies reduce the certainty of evidence. They have methodologic weaknesses related to the RoB, as one study was classified as moderate[17] and one as high RoB.[16] Infraocclusion and root resorption were measured in a heterogeneous way, including evaluation of periapical radiographs[16,17] and bitewings.[4] Also, the studies were different regarding sample sizes; the smallest study had a sample of 18 individuals[16] and the study with the largest sample evaluated 99 patients.[4]

DISCUSSION

Summary of Evidence

Approximately 82% to 89% of deciduous molars in cases of agenesis of the premolar successor remained

Table 4. Evaluation of the Level of Certainty of the Evidence by the GRADE Pro Tool

Certainty Assessment									
Number of Studies	Study Design	Risk of Bias	Inconsistency	Indirectness	Imprecision	Outher Considerations	Impact	Certainty	Importance
Survival of deciduous molars in cases of agenesis of premolar successor.									
3	Observational studies	Serious[a]	Not serious	Not serious	Serious[b]	None	All studies reported an average survival from 82% to 89% of second deciduous molars in the oral cavity after 5 to 13 years of follow-up. Only one study was evaluated as low risk of bias,[4] one with moderate,[17] and one with high.[16] The imprecision between the studies reflects the different samples, where the smallest study had a sample of 18,[16] and the larger study had 99 participants.[4]	⊕⊕◯◯ Low	Important
Root resorption associated to deciduous molars without premolar successor.									
2	Observational studies	Serious[a]	Serious[c]	Not serious	Serious[b]	None	Three studies did not point out significant changes related to the level of root resorption. However, two studies used subjective scales,[4,17] and one high risk of bias study did not report the measurement method.[16] Therefore, it was not included in the evaluation.	⊕⊕◯◯ Low	Important
Infraocclusion associated to deciduous molars without premolar successor.									
2	Observational studies	Serious[a]	Not serious	Not serious	Serious[b]	None	None of the studies reported a considerable increase in infraocclusion. The evaluation method differed between studies, being considered present when the deciduous molar did not reach the crown height of the adjacent tooth[17] or from the measurement in millimeters from the occlusal plane to the occlusal surface of the deciduous molar.[4]	⊕⊕◯◯ Low	Important

[a] High (15), moderate (16), and low (4) risk of bias study.
[b] Great variability of sample size.
[c] Heterogeneity between the methods for measuring resorption.

in the oral cavity for 5 to 13 years. Based on a low evidence level, it is reasonable to consider the maintenance of a retained primary molar in cases with favorable crown, roots, and alveolar bone.[11,18]

The survival rate of implants over a period of 10 years of follow-up was approximately 94%.[19] However, during the growth phase of children and adolescents, implants are susceptible to surgical and prosthetic complications, also related to mandibular growth, which can lead to changes in the position of the implant in both the vertical and horizontal directions.[7] Considering that, on average, 82% to 89% of second deciduous molars remained in the oral cavity during the individual's growth period, it seems reasonable to maintain and postpone implants until the growth period has ended.

Autotransplanted teeth have the potential to adapt to vertical growth and the development of the periodontium.[20] A study carried out 26 years after autotransplant surgery in 11-year-old children found a 90% survival rate.[3] However, as a surgical procedure, it can trigger discomfort to the patient. This is an important factor for the patient's guardian to consider when choosing a rehabilitation therapy.

One of the main dilemmas when a decision is made to retain the lower second deciduous molars, especially when orthodontic treatment is needed, is whether to reduce tooth width so that, in the long term, an optimally sized prosthetic replacement can be placed at the right time to achieve good buccal interdigitation. Alternatively, the mesiodistal width of the lower second molars can be maintained, accepting a compromised buccal segment relationship anteroposteriorly. However, in addition to the inherent pain and discomfort,[9] size reduction by slicing the tooth may stimulate the resorption process. Resorption of primary teeth can be accelerated by inflammatory processes or occlusal trauma and, consequently, cause tooth loss.[21]

Orthodontic treatment with a plan to retain deciduous molars is feasible if they are not included in the

mechanotherapy to prevent root resorption. Clinical monitoring is required concerning mobility, the progression of infraocclusion, associated with radiographic examination every 6 months to assess root resorption. Orthodontic space closure is justified if extraction of the deciduous molar favors orthodontic treatment, as in cases where there is a lack of space in the dental arch or protrusion of the incisors.[22,23] However, this therapy can result in retrusion of the lower incisors and hinder the establishment of a normal overjet and overbite,[20] which can be harmful in patients with a flat or concave facial profile, anterior mandibular rotation, deep overbite, and narrow teeth.[16]

Progressive changes in infraocclusion and root resorption were clinically insignificant. An increase of 1 mm of infraocclusion was observed after a 13-year follow-up.[4] For infraocclusion to compromise the occlusion, teeth would have to be affected between 0.5 mm to 4.5 mm.[24,25] Regarding root resorption, most of the evaluated individuals did not present clinically significant values. A low RoB study[4] reported that 44% of the sample of 99 individuals did not experience a change in the level of root resorption. Similarly, a study with moderate RoB found that 51% of the sample did not show changes.[17]

Dental caries is the main reason for loss of deciduous molars. A study with a high RoB[16] reported that 11.53% were lost due to caries, and a low RoB study[4] reported 18.18%. The maintenance of oral health is a key factor for conserving deciduous molars in the oral cavity. Complementing the adverse effects, autotransplanted teeth have a percentage of ankylosis from 4.2% to 18.2%, and root resorption from 3% to 10%.[26] Patients with dental implants evaluated retrospectively over a 10-year period showed mucositis in 77% of cases, initial peri-implantitis in 41.2%, and moderate/severe peri-implantitis in 16%.[20] All therapies demonstrate some undesirable effects, which must be evaluated along with the biological and financial cost of the procedure, the patient's emotional well-being, and the impact on quality of life, such as biological conditions related to craniofacial growth and the malocclusion present. The decision regarding what clinical procedure to recommend must consider (1) skeletal characteristics, since extraction can be harmful when facial height is reduced, especially if there is no crowding; (2) biological conditions related to root resorption, infraocclusion, caries, and extensive restorations; (3) chronological and dental age because dental implants are not indicated in younger patients; and (4) preference of the patient for the type of treatment.[4]

The GRADE tool was used to classify the evidence as low level. The studies showed methodologic deficiencies, such as the absence of a sample calculation[16,17] and a statistical model that could consider confounding variables, except for the only study with a low RoB.[4] Also, there was a discrepancy in the sample sizes[4,16,17] and in the data analysis of the variables,[16] which generated inaccuracy in the interpretation of the results. However, it is important for the clinician to understand the viability of maintaining second deciduous molars in the absence of premolar successors due to the low incidence of adverse effects and pain/discomfort to the patient as well as the low cost.

Limitations

Observational studies with better methodology and statistical analysis that adjusts for confounding factors are necessary. Also, measurement of variables and data collection must be performed homogeneously and must be standardized using periapical, panoramic, and/or tomographic radiographs. The samples chosen and follow-up periods should be similar for all individuals evaluated, considering that loss within the sample may occur over the study period. For this review, the heterogeneity and considerable risk of bias in the studies included discouraged the performance of meta-analysis. In cases in which a deciduous tooth is maintained, management will require clinical and radiographic monitoring, especially if there is extensive caries or restorations, root resorption, or infraocclusion.

CONCLUSIONS

- Although level of evidence is low, maintaining a second deciduous molar in the oral cavity in cases associated with agenesis of the successor tooth is a viable clinical alternative. Of the retained molars evaluated, 82% to 89% were in good condition over a follow-up from 5 to 13 years.
- Infraocclusion and root resorption did not seem to affect the longevity of the retained deciduous molar, especially in older patients, according to a low level of evidence.
- It is relevant to emphasize the need for studies with long-term follow-up and greater methodologic accuracy to increase the certainty of the evidence.

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