

# Establishing the association between nonnutritive sucking behavior and malocclusions

## A systematic review and meta-analysis

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Infants and young children may engage in nonnutritive sucking behavior (NNSB), that is, habitual sucking of digits, pacifiers, or other objects without deriving any nourishment from them. NNSB is a type of “comfort habit,”<sup>1</sup> affording the child a sense of security and calmness. Researchers have suggested the use of NNSB as a non-pharmacological intervention in the management of acutely painful procedures in preterm infants, neonates, and older infants,<sup>2</sup> and pacifier sucking is related to the reduced incidence of sudden infant death syndrome.<sup>3</sup> However, pacifier use also has been associated with shorter duration of breast-feeding<sup>3,4</sup> and otitis media.<sup>5</sup> Malocclusion, defined as “a deviation in intramaxillary and/or intermaxillary relations of teeth from normal occlusion [contact between teeth],”<sup>6</sup> is

### ABSTRACT

**Background.** The authors studied the effects of nonnutritive sucking behavior (NNSB) on malocclusions through a systematic review of association (etiology).

**Types of Studies Reviewed.** The authors performed a 3-step search strategy, including electronic searches. Studies of healthy participants with a history of active or previous NNSB, for whom specific malocclusion outcomes had been assessed, were eligible for inclusion. The authors considered before-and-after studies, prospective and retrospective (longitudinal) studies, case-control studies, and analytical cross-sectional studies. They excluded reviews, text- and opinion-based articles, conference abstracts, case reports, case-series, and descriptive cross-sectional studies. The authors, using standardized instruments, independently assessed methodological quality and extracted data from the included studies. In situations for which there were sufficient studies, the authors conducted meta-analyses using the random-effects model, supplemented with the fixed-effects model in situations for which statistical heterogeneity was less than 50%, which the authors assessed using the  $I^2$  statistic.

**Results.** The authors included 15 identified studies. They found that NNSB was associated with varying risks of developing malocclusions. Pacifier suckers are less likely to develop an increased overjet compared with digit suckers, although the results of a meta-analysis of 7 studies whose investigators had assessed posterior crossbite in the primary dentition demonstrated a significant association with pacifier sucking over digit sucking ( $n = 5,560$ ; risk ratio, 1.42; 95% confidence interval, 1.18-1.70;  $P = .0001$ ). Longer duration of NNSB was associated with an increased risk of developing malocclusions. Across-study heterogeneity likely resulted from methodological and sample size differences.

**Conclusions.** The authors of this study have confirmed the association between NNSB and the development of malocclusions. This study provides the highest level of evidence on this topic. Pacifiers were associated with a higher risk of developing most malocclusion features when compared with digit sucking.

**Practical Implications.** Though malocclusions are of multifactorial etiology, clinicians should inform parents and caregivers about the dental risks of NNSB, an environmental factor that is modifiable. NNSB should be discouraged in order to avoid the development of malocclusions. Future studies should adopt standardized, universally agreed and accepted definitions and classifications when measuring and reporting orthodontic outcome measures. This will help achieve across-study homogeneity.

**Key Words.** Evidence-based dentistry; finger sucking; malocclusion; meta-analysis; orthodontics; pacifiers; pediatric dentistry; sucking behavior; sucking habits; systematic review.

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another recognized outcome related to NNSB.<sup>7</sup> NNSBs are said to contribute specifically to the development of increased overjet (“horizontal projection of maxillary teeth beyond the mandibular anterior teeth”<sup>6</sup>), posterior crossbite (“an abnormal relationship of a tooth or teeth to the opposing teeth, in which normal buccolingual or labiolingual relationships are reversed”<sup>6</sup>), anterior open bite (“lack of [anterior] tooth contact in an occluding position”<sup>6</sup>), and incorrect sagittal relationship of teeth.<sup>7</sup>

Clinicians should not interpret the presence of a malocclusion as always needing treatment, as the spectrum of malocclusions ranges from those that are associated with minimal or no functional, dental health-related, or esthetic impairment, to those that are severe and can predispose a patient to traumatic dental injury<sup>8,9</sup> or impaction resorption,<sup>10</sup> both of which can cause tooth loss, as well as those that can elicit unfavorable social responses.<sup>11</sup> Clinicians may use reliable and validated indexes, such as the Index of Orthodontic Treatment Need,<sup>12</sup> to stratify patients’ need for orthodontic treatment according to the severity of their malocclusions.

Malocclusions also have multifactorial etiology; they are determined by a complex interaction of both genetics and environment. Whether malocclusions can be corrected by “therapeutic environmental intervention” may be determined by correctly diagnosing the extent to which genetics and the environment play a part in the expression of the phenotype.<sup>13</sup> If the features of a patient’s malocclusion have limited genetic origin, a clinician may suggest that the patient’s parents attempt to modify environmental factors that can induce malocclusion during the patient’s growth and development.<sup>14</sup> The clinician may suggest withdrawing pacifiers from the infant, or the clinician may advise interceptive orthodontic devices for digit suckers.<sup>15</sup> In patients who have ceased NNSB but for whom features of malocclusion have persisted and are severe, the clinician may encourage orthodontic intervention.

Orthodontic treatment carries significant implications for patients and their caregivers or families with respect to absenteeism from school or work and travel to attend appointments, pressure on health service providers to rationalize the use of limited resources, and society as a whole. “Prevention or interception of harmful behaviors may prevent the development of malocclusions, minimize their psychosocial impact, and reduce the demand for orthodontic treatment and the associated economic burden.”<sup>16</sup> Although a large body of literature exists, largely composed of retrospective cohort studies, case reports, case series, and opinion or review articles whose authors have reported on the relationship between NNSB and malocclusions, to our knowledge,

no investigators previously have undertaken a study to reveal high-level evidence, in the form of a systematic review of association (etiology). We conducted this review with the objective of assessing the association between NNSB and malocclusions.

## METHODS

We registered the title of this review and prospectively archived the protocol with the Joanna Briggs Institute (JBI) before commencing the review; we followed the JBI methodology for systematic reviews of association (etiology).<sup>17</sup>

**Review questions.** The objective of this review was to identify the relationship of NNSB on the development of malocclusions. We addressed the following specific review questions:

- What is the risk of developing malocclusions in participants with NNSB compared to those without NNSB?
- What is the risk of developing malocclusions between participants with different types of NNSB?
- What is the risk of developing malocclusions in participants with longer duration of NNSB compared with those having a shorter duration of NNSB?

**Inclusion criteria.** The usual population, intervention, comparator, and outcome approach to generate review questions for systematic reviews does not align with reviews related to etiology. Therefore, we used the population, exposure, and outcome approach to generate the review questions.<sup>17</sup>

**Population.** We conducted a search for studies of healthy participants with a history of active or previous NNSB and no previous orthodontic or surgical treatment. We set no restrictions on the basis of participants’ ages or sex. We excluded studies of participants who had a cleft lip, palate, or both; other craniofacial deformities; any syndrome; or a history of maxillofacial trauma.

**Exposures of interest.** We considered for inclusion studies whose investigators had evaluated the orthodontic impact of pacifier and digit sucking.

**Types of outcomes.** We assessed the following outcomes: increased overjet, sagittal relationship, posterior crossbite, and anterior open bite.

**Types of studies.** In this review, we considered for inclusion before-and-after studies, prospective and retrospective cohort (longitudinal) studies, case-control studies, and analytical cross-sectional studies. We excluded reviews, text- and opinion-based articles, conference abstracts, case reports, case-series, and descriptive cross-sectional studies.

**ABBREVIATION KEY.** AOB: Anterior open bite. CINAHL: Cumulative Index to Nursing and Allied Health Literature. CR: Canine relationship. JBI: Joanna Briggs Institute. MR: Molar relationship. NNSB: Nonnutritive sucking behavior. OJ: Overjet. X-bite: Posterior crossbite.



Supplemental material is available online.

**Search strategy for the identification of studies.** We used a 3-step search strategy. We conducted an initial limited search of MEDLINE and Scopus, followed by an analysis of the text words contained in the titles and abstracts and the index terms used to describe the articles. Next, we conducted a search using all identified key words and index terms across MEDLINE (Ovid), Embase (Ovid), Scopus, and the Cumulative Index to Nursing and Allied Health Literature (CINAHL) (EBSCO) from the inception date of each database up to the end of May 2016 (eTable, available online at the end of this article). After we removed duplicate records, we screened, independently and in duplicate, the title and abstract (or summary, where available) and the descriptor or Medical Subject Headings terms of the identified records to identify potentially relevant articles for full-text assessment. Finally, we performed a citation search of the reference lists of all included articles. We placed no restrictions on the language or the year of publication.

**Assessment of methodological quality.** As JBI-trained reviewers, we both independently assessed the methodological quality of the full-text articles by means of using standardized critical appraisal instruments from JBI SUMARI.<sup>18</sup> This process aims to identify sources of bias by means of using criteria that the reviewer can score as being met, not met, unclear, or not applicable to the particular study.<sup>19</sup> An a priori decision stated that a cutoff for the inclusion of a study would be a score of 8 (maximum score = 10). We resolved any disagreements that arose through discussion until we reached a decision by consensus.

**Data extraction.** Using standardized data extraction tools from JBI SUMARI,<sup>18</sup> we independently extracted data from studies included in the review. The data extracted included authors' names, year of publication, study setting, study design, population details, exposures, and outcomes of significance to the review questions. We contacted authors for clarification or to request further information as required.

**Data analysis and synthesis.** In situations for which there was a sufficient number of studies whose authors had reported comparable exposures and outcomes, we performed a meta-analysis. We used the random-effects model, which we supplemented by using the fixed-effects model in situations for which statistical heterogeneity was low ( $\leq 50\%$ ); we assessed this by using the  $I^2$  statistic, as a means of sensitivity analysis. We calculated risk ratios (RR) with 95% confidence intervals (CI) for each study, as well as for the pooled results of all component studies. We performed all analyses using Review Manager (RevMan), Version 5.3 (The Nordic Cochrane Centre, The Cochrane Collaboration, Copenhagen, Denmark).

## RESULTS

**Search strategy results.** We identified 569 records through electronic database searches; 268 were

duplicates. Of the remaining 301 records, we discarded 228 records after screening the titles and abstracts or summaries. A full-text assessment for methodological quality of the remaining 73 articles eliminated 48 articles. We excluded 1 article because some of the study participants had received orthodontic treatment. We noted that data from 1 study whose authors had published repeated and different outcomes were identified in 2 articles; we included only the more comprehensive article. We contacted the authors of 8 articles to clarify data, provide additional information relevant to the review that was not apparent in the identified article, or both. One author replied that the data were no longer accessible, the authors of 2 articles had not collected the information we sought, 1 author did not return with clarification of data published in 4 articles, and the authors of 1 other study did not reply. Therefore, we excluded those articles. We included 15 studies in the systematic review (Figure). A citation search of the included articles did not reveal additional records.

**Included studies.** Table 1 shows the main characteristics of the 15 studies,<sup>20-34</sup> all of which were published in English.

**Characteristics of the study settings.** Investigators conducted 7 of the included studies in Brazil,<sup>25-28,30,33,34</sup> 2 each in Finland,<sup>21,23</sup> Italy,<sup>24,32</sup> and Sweden,<sup>20,29</sup> and 1 each in Saudi Arabia<sup>22</sup> and the United Kingdom.<sup>31</sup>

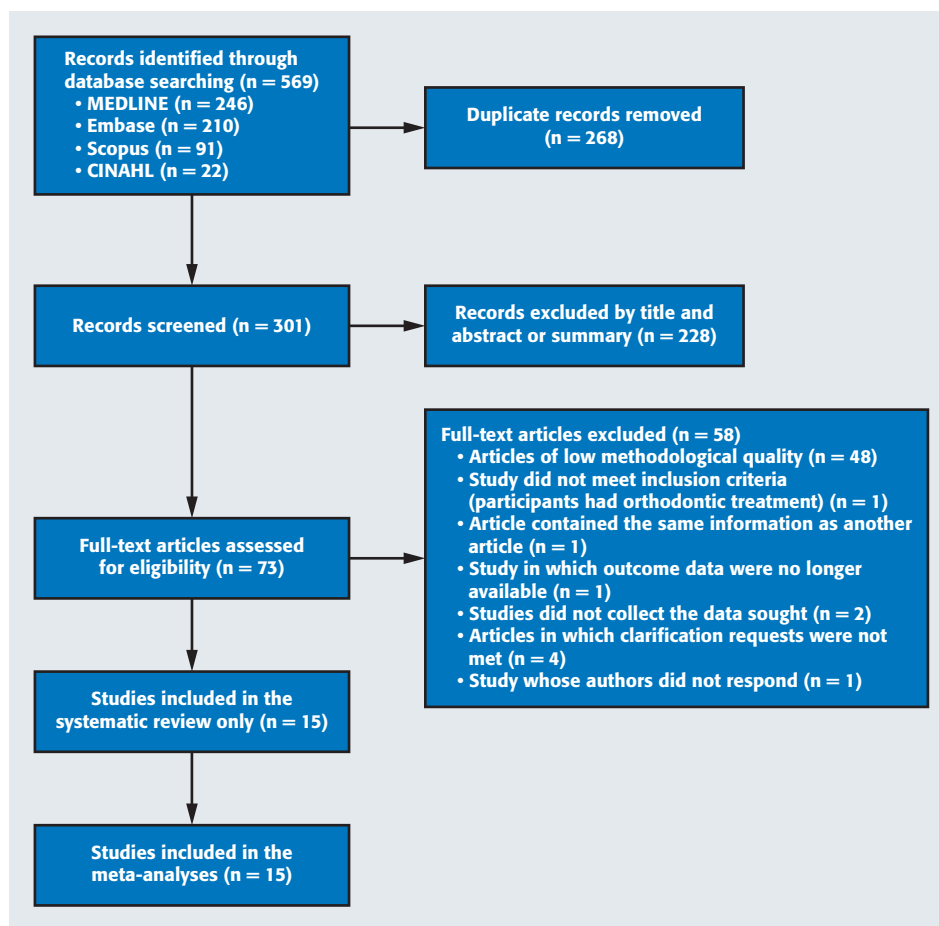
**Characteristics of the participants.** The investigators of all of the studies had investigated children; the investigators of 13 examined the primary dentition,<sup>20-30,33,34</sup> the investigators of 1 examined the mixed dentition,<sup>32</sup> and the investigators of another examined the mixed and secondary dentitions.<sup>31</sup>

**Characteristics of the exposures.** The investigators of 9 studies looked at both pacifier and digit sucking,<sup>20,22,25,26,28,29,32-34</sup> the authors of 1 study investigated only digit sucking,<sup>31</sup> and the investigators of 6 studies examined the combined effect of all types of NNSB as a single exposure.<sup>21,23,24,27,29,30</sup>

**Characteristics of the outcomes.** With the available data, it was possible to perform meta-analyses on 4 malocclusion outcomes in the primary dentition and 3 in the mixed dentition. eFigures 1 and 2<sup>20-34</sup> (available online at the end of this article) show forest plots of all of the meta-analyses.

**Primary dentition: NNSB versus no NNSB.** Children with NNSB were at risk of developing a class II canine relationship, posterior crossbite, anterior open bite, or a combination of these (Table 2, eFigure 1A [available online at the end of this article]).

**Primary dentition: pacifier sucking versus digit sucking.** Children with a pacifier sucking habit were 32% less likely to develop an increased overjet, although they were at risk of developing a class II canine relationship. The results of a meta-analysis of 7 studies whose investigators had assessed posterior crossbite demonstrated a



**Figure.** Flowchart of the screening and study selection process. CINAHL: Cumulative Index to Nursing and Allied Health Literature.

significant association with pacifier sucking ( $n = 5,560$ ;  $RR = 1.42$ ; 95% CI, 1.18-1.70;  $P = .0001$ ). Both random-effects and fixed-effects models gave similar results. The results related to anterior open bite were inconclusive (Table 2, eFigure 1B [available online at the end of this article]).

*Primary dentition: pacifier sucking versus no pacifier sucking habit, and digit sucking versus no digit sucking habit.* We found an association between pacifier sucking and the development of posterior crossbite and anterior open bite. We could not establish a difference in the risk of developing posterior crossbite with digit sucking, although digit suckers were more likely to develop an anterior open bite (Table 2, eFigure 1C and D [available online at the end of this article]). These results should be viewed with caution, as there was evidence of a moderate to high level of heterogeneity, meaning the effect sizes may not be accurate. In addition, the investigators of the component studies included in these meta-analyses conducted the studies in a single country. Therefore, the

generalizability of the findings may be limited only to the populations studied.

*Mixed dentition: digit sucking versus no sucking habit.* We found that no overall difference was demonstrated in the development of a class II molar relationship, although an association existed between digit sucking and posterior crossbite. There was a significant association between digit sucking and anterior open bite; the results were not significantly heterogeneous (Table 2, eFigure 2 [available online at the end of this article]).

*Secondary dentition.* It was not possible to provide narrative syntheses into the outcomes in this dentition owing to the relatively few participants in the single identified study.<sup>31</sup> A lack of other studies with results related to secondary dentition precluded meta-analyses.

*Longer- versus shorter-duration NNSB.* Longer duration of pacifier sucking was associated with anterior open bite<sup>25</sup> and a class II canine relationship<sup>33</sup> in the primary dentition. Longer duration of NNSBs was associated with anterior open bite in the primary dentition.<sup>27</sup>

## DISCUSSION

To our knowledge, we are the first to conduct a systematic review that examined the association between NNSB and malocclusions. The results of our review provide the highest level of evidence on this topic, confirming the important role of NNSB in the development of specific features of malocclusions, which has been shown previously in the literature.<sup>20-34</sup> The nature of a systematic review allows investigators to pool the results of studies, which, with the increased sample sizes of the populations and the number of “events” within them, provide the best possible estimates of effect, compared with results of individual studies viewed in isolation. This

TABLE 1

Characteristics of the included studies.							
STUDY	COUNTRY	STUDY DESIGN	SAMPLE SIZE	AGE OF PARTICIPANTS	DENTITION	EXPOSURE(S)	OUTCOME MEASURES
Larsson, <sup>20</sup> 1975	Sweden	Prospective cohort	3,214	4 y	Primary	Pacifier Digit sucking	X-bite* AOB <sup>†</sup>
Paunio and Colleagues, <sup>21</sup> 1993	Finland	Cross-sectional nested within a cohort	938	3 y	Primary	NNSB <sup>‡</sup>	X-bite AOB
Farsi and Salama, <sup>22</sup> 1997	Saudi Arabia	Cross-sectional	583	3-5 y	Primary	Pacifier Digit sucking	OJ <sup>§</sup> CR <sup>¶</sup> X-bite AOB
Karjalainen and Colleagues, <sup>23</sup> 1999	Finland	Cross-sectional nested within a cohort	148	3 y	Primary	NNSB	OJ X-bite AOB
Viggiano and Colleagues, <sup>24</sup> 2004	Italy	Cross-sectional	1,130	3-5 y	Primary	NNSB	X-bite AOB
Peres and Colleagues, <sup>25</sup> 2007	Brazil	Cross-sectional nested within a cohort	359	6 y	Primary	Pacifier Digit sucking	X-bite AOB
Hebling and Colleagues, <sup>26</sup> 2008	Brazil	Cross-sectional	728	5 y	Primary	Pacifier Digit sucking	X-bite AOB
Heimer and Colleagues, <sup>27</sup> 2008	Brazil	Prospective cohort	287	4-6 y	Primary	NNSB	X-bite AOB
Macena and Colleagues, <sup>28</sup> 2009	Brazil	Cross-sectional	2,750	18-59 mo	Primary	Pacifier Digit sucking	X-bite
Dimberg and Colleagues, <sup>29</sup> 2010	Sweden	Cross-sectional	457	3 y	Primary	Pacifier Digit sucking NNSB	OJ CR X-bite AOB
Mistry and Colleagues, <sup>31</sup> 2010	United Kingdom	Cross-sectional	75	7-13 y	Mixed secondary	Digit sucking	OJ MR <sup>#</sup> X-bite AOB
Jabbar and Colleagues, <sup>30</sup> 2011	Brazil	Cross-sectional	911	3-6 y	Primary	NNSB	OJ CR
Montaldo and Colleagues, <sup>32</sup> 2011	Italy	Cross-sectional	1,451	7-11 y	Mixed	Pacifier Digit sucking	MR X-bite AOB
Caramex da Silva and Colleagues, <sup>33</sup> 2012	Brazil	Cross-sectional nested within a cohort	153	3-5 y	Primary	Pacifier Digit sucking	CR
dos Santos and Colleagues, <sup>34</sup> 2012	Brazil	Cross-sectional	1,385	5-6 y	Primary	Pacifier Digit sucking	OJ CR X-bite AOB

\* X-bite: Posterior crossbite.  
† AOB: Anterior open bite.  
‡ NNSB: Nonnutritive sucking behavior.  
§ OJ: Overjet.  
¶ CR: Canine relationship.  
# MR: Molar relationship.

contributes to clinicians' ability to have greater confidence in the results of meta-analyses, for instance, when compared with reflecting on the results of individual studies, for which clinicians also need to consider whether the results are generalizable to the population with which they work and whether they can draw any meaningful and practical implications from the results. In this study, we did not aim to report on the appropriate management of malocclusions induced by NNSB, nor on the effectiveness of interventions, as these topics already had been addressed by the authors of a different

systematic review and meta-analysis.<sup>15</sup> Our findings of increased risk of developing a class II canine relationship, posterior crossbite, and anterior open bite related to pacifier sucking versus digit sucking in the primary dentition are consistent with the findings of a longitudinal study whose investigators examined sucking habits in childhood,<sup>35</sup> which suggested that digit sucking is "... a preferable habit to dummy sucking..." However, because of a patient's greater risk of developing an increased overjet that we identified with digit sucking, we are unable to support this statement.

TABLE 2

<b>Summary of meta-analyses* comparing different exposures against specific malocclusion features in the primary and mixed dentitions.</b>				
DENTITION	EXPOSURE	INCREASED OVERJET		
		No. of Studies	Pooled Risk Ratio (95% CI <sup>†</sup> )	P Value
Primary	NNSB <sup>‡</sup> versus no NNSB	Insufficient number of studies for meta-analysis		
	Pacifier versus digit sucking	2	0.68 (0.36-1.29)	.24
	Pacifier versus no pacifier sucking habit	Insufficient number of studies for meta-analysis		
	Digit versus no digit sucking habit	Insufficient number of studies for meta-analysis		
Mixed	Digit versus no sucking habit	Insufficient number of studies for meta-analysis		

\* All meta-analyses were conducted using the random-effects model.  
<sup>†</sup> CI: Confidence interval.  
<sup>‡</sup> NNSB: Nonnutritive sucking behavior.  
<sup>§</sup> Results of meta-analyses that were conducted with the fixed-effects model in addition to the random-effects model.  
<sup>¶</sup> Please note that these values represent class II molar relationship rather than class II canine relationship.

The JBI methodology has a “broader definition of what constitutes research evidence for practice”<sup>36</sup> and is not restricted to considering only a specific type of study design as a source of evidence. This approach allows for evaluation of the literature on the basis of the study design, the methodological quality, and the rigor of evidence, and these are not necessarily related to the strength of the findings.<sup>36</sup> Another important feature of reviews whose investigators use JBI methodology is distinguishing the included studies according to methodological quality. Only high-quality studies are included, as they provide scientifically sound and clinically relevant results in relation to the review question. This in turn gives strength and reliability to the results and, therefore, validity to the meta-analysis.<sup>37</sup> The JBI methodology accounts for different types of study designs by means of using study-specific and standardized critical appraisal and data extraction tools.<sup>18</sup> In this way, the variability in design of the component studies, and their observed effect estimates, are unlikely to contribute to unrealistic or inaccurate estimates of effect. It may be argued that the exclusion of poorly designed studies goes against the spirit of inclusiveness of meta-analysis. Furthermore, analyses of studies that were associated with lower quality methodology could be undertaken and reported separately. However, results emanating from such studies may be questionable and perhaps invalid, owing to bias in their design, conduct, analysis, or a combination of these.<sup>19</sup> Therefore, reporting the results of meta-analyses that include poorly designed studies may be considered problematic, and the usefulness of the results for informing health care providers, researchers, and policy makers is doubtful.

During the course of the review, we encountered across-study heterogeneity in participant characteristics, clinical definitions, and classification of outcome

measures. Many of the studies we included were sub-studies with participant characteristics that had been determined by the broader aims and objectives of a parent study. The World Health Organization recommends the index ages of 5 and 12 years for population oral health surveys<sup>38</sup>; however, not all study investigators adopt these recommendations. To circumvent age-related heterogeneity, we investigated the different exposures and outcomes with respect to the dentition that was present.

Investigators have varying definitions of increased overjet. Two studies defined this as greater than 2 millimeters,<sup>30,34</sup> another considered values greater than 3 mm,<sup>23</sup> whereas others accepted 4 mm as the critical value.<sup>22,29</sup> Those study investigators who adopted a lower threshold may have overestimated the prevalence compared with the study investigators who adopted a higher threshold, and vice versa. Given the variability, we were able to perform only a single meta-analysis on this outcome, prioritizing studies whose investigators had reported the highest threshold. Orthodontic treatment, in the context of where it is rationed or subsidized by the state, is offered to those whose overjet is stratified as being severe and, therefore, in great or very great need for treatment.<sup>12</sup> Heterogeneity of this particular clinical definition likely stems from differences in identifying when an overjet should be considered to be “increased.” Authors of an internationally respected orthodontic textbook stated that normal overjet was 2 to 3 mm,<sup>7</sup> leading to the inference that values greater than 3 mm were increased. The Index of Orthodontic Treatment Need,<sup>12</sup> used extensively in the United Kingdom as well as in other countries, distinguishes any overjet greater than 3.5 mm as being increased.<sup>12</sup> Study investigators who adopted 2 mm as being the “increased” reference point cited an article published in 1969<sup>39</sup> about a nested

**TABLE 2 (CONTINUED)**

CLASS II CANINE RELATIONSHIP			POSTERIOR CROSSBITE			ANTERIOR OPEN BITE		
No. of Studies	Pooled Risk Ratio (95% CI)	P Value	No. of Studies	Pooled Risk Ratio (95% CI)	P Value	No. of Studies	Pooled Risk Ratio (95% CI)	P Value
2	5.27 (0.72-38.39)	.10	5	2.32 (1.39-3.88)	.001	5	10.33 (5.29-20.15)	< .00001
3	1.80 (0.61-5.32)	.29	7	1.42 (1.18-1.70)	.0001	6	1.15 (0.87-1.53)	.32
				1.45 <sup>§</sup> (1.21-1.74)	< .0001 <sup>§</sup>			
Insufficient number of studies for meta-analysis			3	2.53 (1.68-3.81)	< .00001	3	3.03 (0.95-9.72)	.06
Insufficient number of studies for meta-analysis			3	1.01 (0.58-1.77)	.96	3	1.42 (1.10-1.84)	.007
2 <sup>¶</sup>	1.09 (0.63-1.90) <sup>¶</sup>	.76 <sup>¶</sup>	2	2.06 (0.95-4.48)	.07	2	6.37 (1.17-34.86)	.03
							4.11 <sup>§</sup> (3.31-5.10)	< .00001 <sup>§</sup>

study whose investigators used a sample that was admittedly not representative of the target population and in which the outcomes of only “white” children were preferentially reported.<sup>30,34</sup>

We also found differences in the classification of outcome measures, specifically sagittal relationships. Most study investigators used the classification by Foster and Hamilton.<sup>39</sup> With a single exception, all studies using this classification were Brazilian, with most authors having a dental public health background. We suggest that Angle’s<sup>40</sup> classification be the preferred outcome measure because it is widely recognized and used internationally by dentists, orthodontists, and other clinical dental specialists, who, for the most part, are responsible for identifying, intercepting, or treating malocclusions.

Despite limiting meta-analyses to be used only when there were a sufficient number of studies whose populations were homogenous in terms of participant characteristics, exposures, and outcomes, we encountered moderate statistical heterogeneity (50-90%) in most of the results of the meta-analyses. Methodological differences, such as categorization of participants, differences in the sample sizes and number of events, as well as individual variation in the expression of malocclusions, are possible explanations. Some participants may have engaged in both pacifier and digit sucking. Rather than being allocated into a unique group, they may have been considered according to the predominant or most recent habit, or even double-counted in both the pacifier-sucking and digit-sucking groups. We attempted to find explanations for the heterogeneity, but a lack of sufficient information prevented subgroup analyses; not all authors we approached for clarification replied. We nonetheless endeavored to avoid the problems presented by statistical heterogeneity by applying a random-effects model in all meta-analyses, supplemented by applying a fixed-effects model when heterogeneity was low.

In light of the across-study heterogeneity, which can hamper the conduct of meta-analyses, we recommend

that investigators adopt uniform, standard, and widely accepted definitions and classifications when measuring and reporting orthodontic outcome measures. This facilitates precise communication between clinicians and researchers. Also, we urge researchers to allocate study participants to exposure-specific groups in future studies, which may help investigators using meta-analyses to reach clearer directions of effect, rather than identifying inconclusive results or reporting an absence of differences. Adoption of these recommendations can help achieve across-study homogeneity, which can ensure validity of the overall conclusions of reviews and meta-analyses, so that the clinical interpretations are not misleading and can be relied on to inform the decision-making process. The authors of a seminal article<sup>41</sup> written more than 40 years ago raised the importance of this issue.

Some of the component studies had inconsistent findings when compared with the results of the meta-analyses. dos Santos and colleagues<sup>34</sup> found pacifier sucking to be favorable for anterior open bite in the primary dentition compared with digit sucking, as did Farsi and Salama,<sup>22</sup> who also found that pacifier sucking favored class II canine relationships and posterior crossbite. Random error and small-study effect may explain these differences. Opposing results concerning the sagittal relationship in the mixed dentition also was evident.<sup>31,32</sup> Confounding factors, such as the early loss of primary molars allowing mesial drift of secondary molars into the leeway space to establish a class II sagittal relationship, might be responsible, and we suggest that investigators consider and adjust for these factors in future studies. We were unable to assess publication bias in this review as none of our meta-analyses had more than 10 component studies.

NNSB is 1 of several variables influencing malocclusions. Despite study investigators’ attempts to avoid or minimize bias by means of controlling for confounders or mediators, such as neonatal characteristics of the child, socioeconomic status of the child’s household,

dental behaviors, and infant feeding methods, individual variation nonetheless may exert sufficient influence to render any attempt to demonstrate the existence or significance of differences difficult.<sup>42</sup> Some of our results did not reach statistical significance; this should not be misinterpreted as evidence of no effect or difference. Statistical significance, often represented by a *P* value, is related to sample size and, therefore, could be improved by increasing the number of study participants.<sup>43</sup> It is also important to reflect that statistical significance does not necessarily equal clinical significance, the latter referring to “whether the [intervention] makes a real (for example, genuine, palpable, practical, noticeable) difference in the everyday life” of people.<sup>44</sup> Aside from validated qualitative research methods that can help health care professionals better appreciate the impact of different features of malocclusions on study participants, investigators also may determine clinical significance by examining the effect size, such as the relative risk, which helps “estimate the magnitude of effect or association between 2 or more variables.”<sup>45</sup> Investigators should consider effect size in relation to the size of the 95% CI (influenced by sample size and number of events) and its upper and lower boundaries, because the effect size may not be accurate in the presence of statistical heterogeneity. We found that the upper and lower boundaries of the 95% CIs of nearly one-half of our meta-analyses clearly favored an exposure or a control over the other. In other words, there was consistency in the direction of the effect. Such findings help health care professionals give appropriate advice to patients and their caregivers or families. For example, the highest effect size estimate in our systematic review related to NNSB and anterior open bite, which had a moderate degree of statistical heterogeneity (Table 2, eFigure 1). As both boundaries of the CI favored no sucking habit, the correct clinical advice would be to refrain from NNSB in the primary dentition so as to avoid the development of an anterior open bite.

The results of our systematic review confirm the association between NNSB and malocclusions and offer a higher level of evidence than was previously available. The investigators of future systematic reviews and meta-analyses on this topic may benefit from the inclusion of prospective and high-quality research on the effects of NNSB on malocclusions that are scarce. Although infants may determine whether they will suck their digits, parents and caregivers have the right to choose whether to introduce a pacifier. It is important that they are properly informed about the facts to make the most appropriate decisions.

## CONCLUSIONS

In the primary dentition, NNSB should be discouraged to avoid the development of malocclusions. Pacifier and digit sucking both present varying risks of developing features of malocclusions. When comparing pacifier with

digit sucking, children are less likely to have an increased overjet if they use a pacifier, however, they are at greater risk of developing other malocclusions such as a class II canine relationship and posterior crossbite. In the mixed dentition, a history of digit sucking carries an increased risk of developing posterior crossbite and anterior open bite. Longer duration of NNSB is associated with increased risk of developing a malocclusion. ■

## SUPPLEMENTAL DATA

Supplemental data related to this article can be found at <http://dx.doi.org/10.1016/j.adaj.2016.08.018>.

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eTABLE

<b>Electronic database search strategies.</b>				
<b>SEARCH TERM ID/STEP</b>	<b>MEDLINE (OVID)</b>	<b>EMBASE (OVID)</b>	<b>SCOPUS</b>	<b>CINAHL* (EBSCO)</b>
1	Sucking Behavior/	Sucking behavior.mp	((( TITLE-ABS-KEY ( sucking behavior ) OR TITLE-ABS-KEY ( suck\$ AND ( habit\$ OR behav\$ OR routine\$ ) ) OR TITLE-ABS-KEY ( non nutritive suck\$ OR non-nutritive suck\$ OR nonnutritive suck\$ ) ) AND ( ( TITLE-ABS-KEY ( pacifiers ) OR TITLE-ABS-KEY ( fingersucking ) OR TITLE-ABS-KEY ( pacifier\$ OR digit\$ OR dummy OR dummies OR soother\$ OR blanket\$ OR finger\$ OR thumb\$ ) ) ) AND ( ( TITLE-ABS-KEY ( malocclusion ) OR TITLE-ABS-KEY ( malocclusion, angle class i ) OR TITLE-ABS-KEY ( malocclusion, angle class ii ) OR TITLE-ABS-KEY ( malocclusion, angle class iii ) OR TITLE-ABS-KEY ( dental occlusion ) OR TITLE-ABS-KEY ( tooth occlusion ) OR TITLE-ABS-KEY ( occlusion ) OR TITLE-ABS-KEY ( distocclusion OR disto-occlusion OR distocclusion ) OR TITLE-ABS-KEY ( mesiocclusion OR mesio-occlusion OR mesioocclusion ) AND TITLE-ABS-KEY ( canine relationship ) AND TITLE-ABS-KEY ( molar relationship ) ) ) OR ( ( TITLE-ABS-KEY ( retrognathia ) OR TITLE-ABS-KEY ( prognathism ) ) ) OR ( ( TITLE-ABS-KEY ( open bite ) OR TITLE-ABS-KEY ( anterior open bite OR asymmetric anterior open bite OR symmetric anterior open bite ) ) ) OR ( ( TITLE-ABS-KEY ( overbite ) OR TITLE-ABS-KEY ( overjet OR crossbite OR deep bite ) OR TITLE-ABS-KEY ( centerline discrepancy ) OR TITLE-ABS-KEY ( Index of orthodontic treatment needs ) OR TITLE-ABS-KEY ( dental arch ) OR TITLE-ABS-KEY ( palate ) OR TITLE-ABS-KEY ( growth, development AND ageing ) ) ) ) AND ( LIMIT-TO ( EXACTKEYWORD , "Human" ) ) )	MH "Sucking Behavior"
2	(suck\$ and (habit\$ or behav\$ or routine\$)).mp	(suck* and (habit* or behave* or routine*)).mp		(( (suck* and habit*) ) OR ( (suck* and behav*) ) OR ( (suck* and routine*) ) )
3	("non nutritive suck\$" or "non-nutritive suck\$" or "nonnutritive suck\$").mp	("non nutritive suck*" or "non-nutritive suck*" or "nonnutritive suck*").mp		"Non nutritive suck*" OR "nonnutritive suck*" OR "non-nutritive suck*"
4	1 or 2 or 3	1 or 2 or 3		MH "Pacifiers"
5	Pacifiers/	pacifier.mp		pacifier* OR digit* OR dummy OR dummies OR soother* OR blanket* OR finger* OR thumb*
6	Fingersucking/	fingersucking.mp		S1 or S2 or S3
7	(pacifier\$ or digit\$ or dummy or dummies or soother\$ or blanket\$ or finger\$ or thumb\$).mp	(pacifier* or digit* or dummy or dummies or soother* or blanket* or finger* or thumb*).mp		S4 or S5
8	5 or 6 or 7	5 or 6 or 7		S6 and S7
9	4 and 8	4 and 8		MH "Malocclusion" OR "Malocclusion, Angle Class I" OR "Malocclusion, Angle Class II" OR "Malocclusion, Angle Class III"
10	Malocclusion/ or Malocclusion, Angle Class I/ or Malocclusion, Angle Class II/ or Malocclusion, Angle Class III/	Malocclusion.mp		dental occlusion OR tooth occlusion
11	Dental occlusion/ or tooth occlusion.mp	Malocclusion, Angle Class I.mp		occlusion
12	occlusion.mp	Malocclusion, Angle Class II.mp		distocclusion OR disto-occlusion OR distocclusion
13	(distocclusion or disto-occlusion or distocclusion).mp	Malocclusion, Angle Class III.mp		mesiocclusion OR mesio-occlusion OR mesiocclusion
14	(mesiocclusion or mesio-occlusion or mesiocclusion).mp	10 or 11 or 12 or 13		canine relationship
15	canine relationship.mp	((dental or tooth) and occlusion).mp		molar relationship
16	molar relationship.mp	occlusion.mp		S9 or S10 or S11 or S12 or S13 or S14 or S15
17	11 or 12 or 13 or 14 or 15 or 16	15 or 16		retrognathia
18	Retrognathia/	(distocclusion* or disto-occlusion* or distocclusion*).mp		prognathism
19	Prognathism	(mesiocclusion* or mesio-occlusion* or mesiocclusion*).mp		S17 or S18
20	18 or 19	retrognath*.mp		open bite
21	Open Bite/	prognath*.mp		anterior open bite OR asymmetric anterior open bite OR symmetric anterior open bite
22	(Anterior open bite or asymmetric anterior open bite or symmetric anterior open bite).mp	((canine or molar) and relationship).mp		S20 or S21
23	21 or 22	18 or 19 or 20 or 21 or 22		overbite
24	Overbite/	(posterior adj3 occlusion*).mp		overjet or crossbite or deep bite
25	(Overjet or crossbite or deep bite).mp	(anterior adj3 occlusion).mp		centerline discrepancy

\* CINAHL: Cumulative Index to Nursing and Allied Health Literature.

eTABLE (CONTINUED)

SEARCH TERM ID/STEP	MEDLINE (OVID)	EMBASE (OVID)	SCOPUS	CINAHL* (EBSCO)
26	centerline discrepancy.mp	(overjet* or over jet* or over-jet*).mp		Index of Orthodontic Treatment Need
27	"Index of Orthodontic Treatment Need"/	(crossbite* or cross bite* or cross-bite*).mp		dental arch
28	Dental Arch/	(deep bite* or deepbite* or deep-bite*).mp		palate
29	Palate/	(overbite* or over bite* or over-bite*).mp.		growth, development and aging
30	(Growth, development and aging).mp	(open bite* or openbite* or open-bite*).mp.		S16 or S19 or S22 or S23 or S24 or S25 or S26 or S27 or S28 or S29
31	10 or 17 or 20 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30	(anterior open bite* or anterior openbite* or anterior open-bite*).mp.		S8 and S30
32	9 and 31	((asymmetric or symmetric) and anterior).mp		
33	limit 32 to humans	30 and 32		
34		Index of Orthodontic Treatment Need.mp		
35		dental arch*.mp		
36		palate*.mp		
37		(growth, development and aging).mp		
38		14 or 17 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 31 or 33 or 34 or 35 or 36 or 37 or 38		
39		9 and 38		
40		limit 39 to human		
<b>Total Records</b>	<b>246</b>	<b>210</b>		<b>91</b>

**i. Class II canine relationship**



**ii. Posterior crossbite**



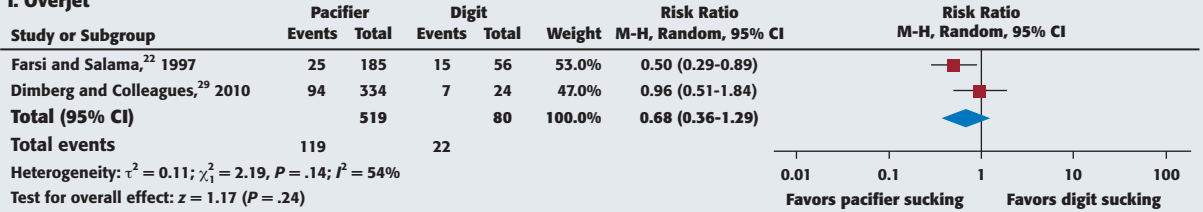
**iii. Anterior open bite**



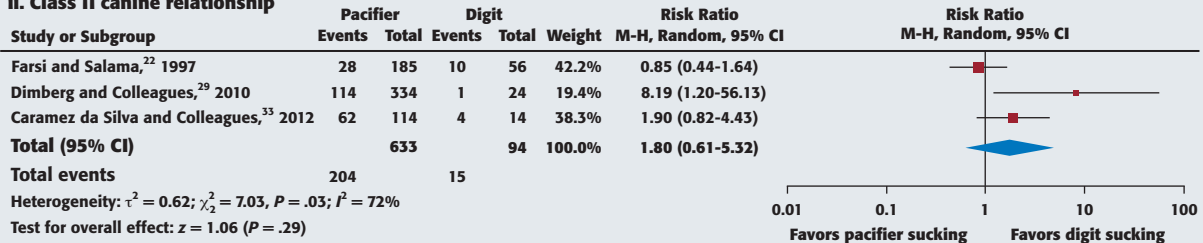
**A**

**eFigure 1.** Forest plots of meta-analyses investigating the effects of nonnutritive sucking behavior (NNSB) on malocclusion outcomes in the primary dentitions. **A.** NNSB versus no NNSB. **B.** Pacifier sucking versus digit sucking. **C.** Pacifier sucking habit versus no pacifier sucking habit. **D.** Digit sucking habit versus no digit sucking habit. \*: Data from initial examination in 2002 used in this analysis. CI: Confidence interval. M-H: Mantel-Haenszel test.

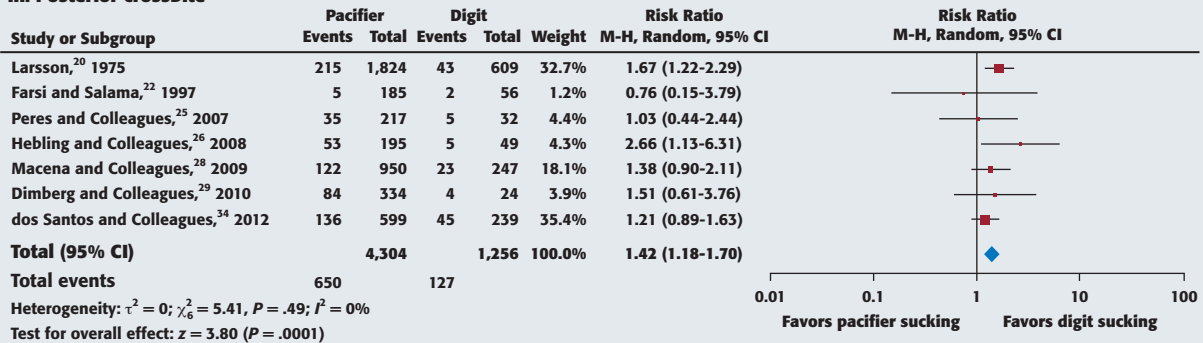
**i. Overjet**



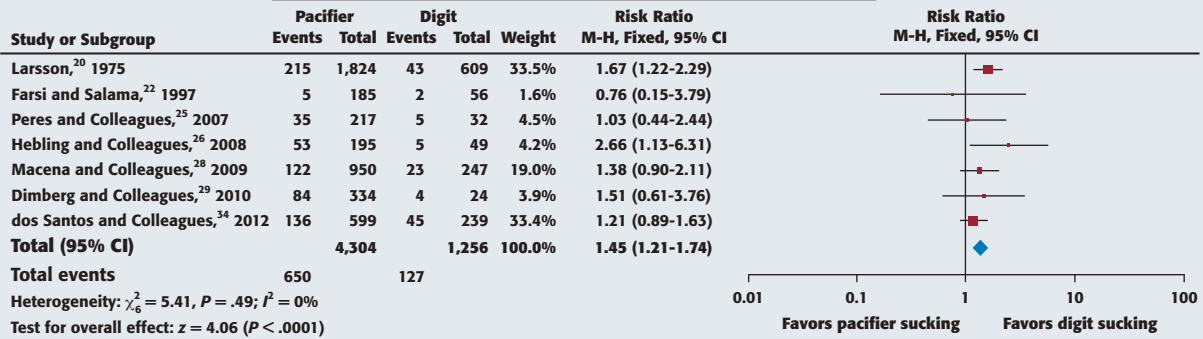
**ii. Class II canine relationship**



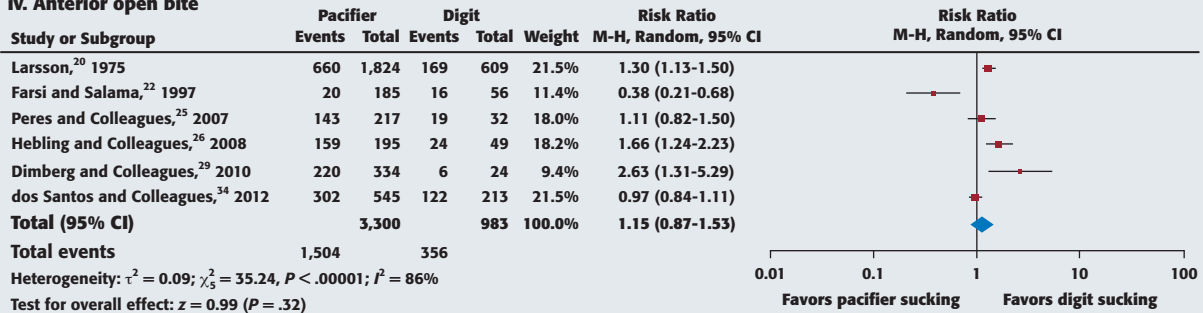
**iii. Posterior crossbite**



Supplementation with fixed-effects model owing to low statistical heterogeneity



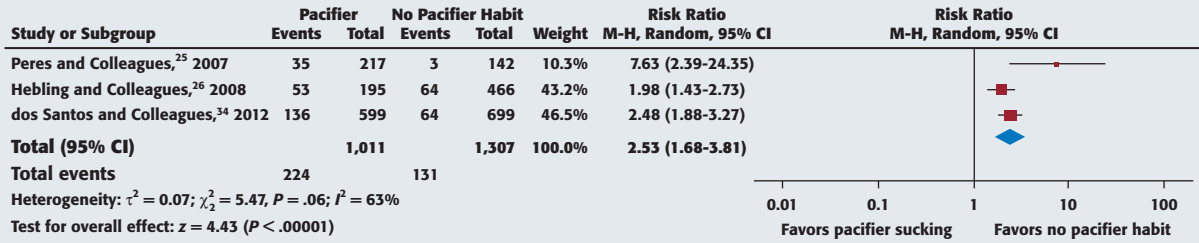
**iv. Anterior open bite**



**B**

**eFigure 1.** Continued

**i. Posterior crossbite**

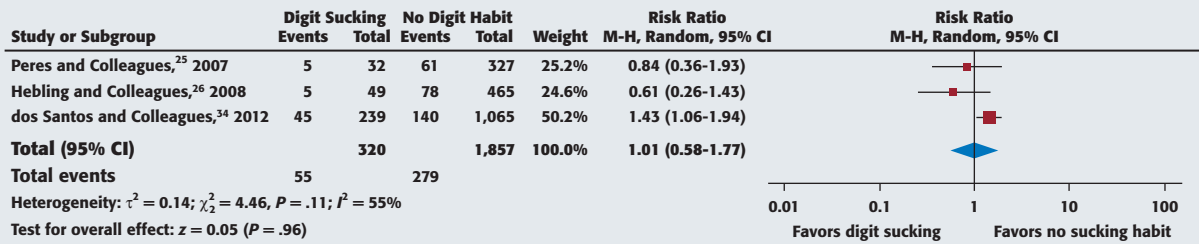


**ii. Anterior open bite**



**C**

**i. Posterior crossbite**



**ii. Anterior open bite**



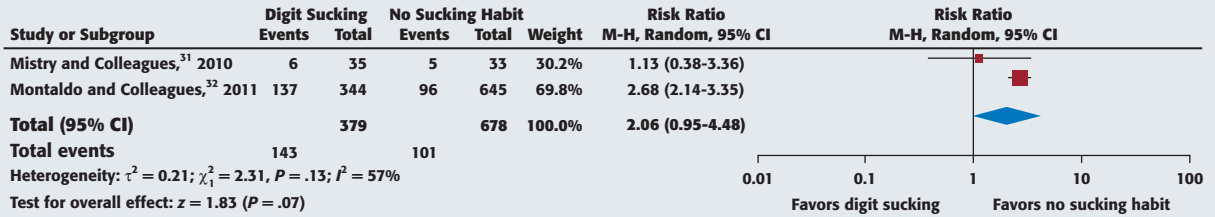
**D**

**eFigure 1.** Continued

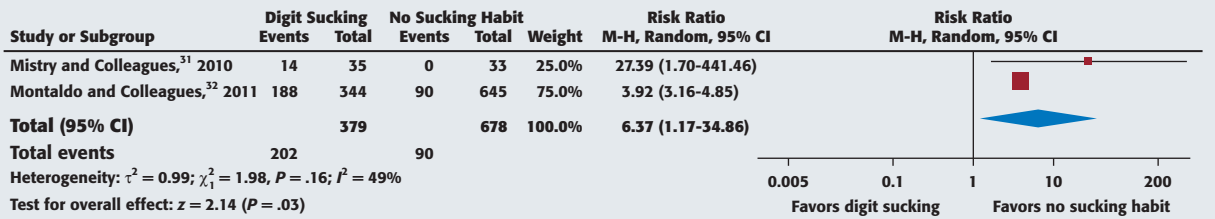
**i. Class II molar relationship**



**ii. Posterior crossbite**



**iii. Anterior open bite**



Supplementation with fixed-effects model due to low statistical heterogeneity



**eFigure 2.** Forest plots of meta-analyses investigating the effects of nonnutritive sucking behavior (NSB) on malocclusion outcomes in mixed dentitions.