Cover Story

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Prevalence of ankyloglossia according to different assessment tools

A meta-analysis

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ABSTRACT

Background. Prevalence of ankyloglossia may vary depending on the assessment tool. This systematic review aimed to evaluate the prevalence of ankyloglossia in distinct age groups according to different assessment tools.

Types of Studies Reviewed. Nine electronic databases were searched from inception through November 2021 without restrictions of language or year of publication. Paired independent reviewers selected cross-sectional and cohort studies reporting the diagnosis of ankyloglossia, extracted data, and assessed methodological quality. The number of patients with ankyloglossia and the sample were extracted to calculate the overall prevalence of ankyloglossia and 95% CI. The authors calculated the prevalence of ankyloglossia per assessment tool, age group, and sex. They assessed the certainty of evidence using the Grading of Recommendations Assessment, Development and Evaluation approach.

Results. Seventy-one studies were included. Seven different diagnostic tools were used. The overall prevalence of ankyloglossia was 5% (95% CI, 4.0% to 5.0%) and ranged from 2% (using an unspecific tool) to 20% (Coryllos classification). The prevalence per age group was higher in infants (7%). The prevalence ratio was 1.34 (95% CI, 1.17 to 1.54) for boys, with very low certainty of evidence.

Practical Implications. The prevalence of ankyloglossia is higher among infants and differs depending on the assessment tool used for the diagnosis. It is uncertain whether boys are more affected by ankyloglossia than girls.

Key Words. Lingual frenulum; tongue; systematic review; ankyloglossia; prevalence.

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Ankyloglossia, or tongue-tie, is a congenital anomaly characterized by a short lingual frenulum or frenulum highly adhered to the genioglossus muscle, which restricts the movements of the tongue and causes motor and functional problems.[1] The restricted tongue movement due to ankyloglossia may be associated with breast-feeding problems,[2,3] can affect speech and swallowing, and can cause orthodontic problems, such as malocclusion, open bite, and separation of the mandibular incisors.[4,5]

The literature reports several assessment tools for diagnosing ankyloglossia. Some common tools are the Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF), which evaluates tongue appearance and function[6]; the Bristol Tongue Assessment Tool (BTAT), which was developed on the basis of the 4 most important aspects of the HATLFF[7]; the Coryllos classification[8] and Kotlow classification,[9] both comprising 4 categories of ankyloglossia severity; and the Lingual Frenulum Protocol for Infants and its short version, the Neonatal Tongue Screening Test.[10] Owing to the particularities of each instrument, the prevalence of ankyloglossia can vary, and there is no consensus on the best assessment tool for diagnosing this condition.[11]

This article has an accompanying online continuing education activity available at: http://jada.ada.org/ce/home.

Two previous systematic reviews summarized the prevalence of ankyloglossia in infants, ranging from 4.2% through 10.7%.[11,12] The prevalence varied according to the assessment tool used for the diagnosis.[11] However, the evidence has limitations. Both systematic reviews only considered studies with infants, and few studies were included (5 in the first systematic review and 15 in the second).[11,12] Most importantly, neither systematic review summarized the prevalence in age groups other than infants, and there is no information regarding the overall prevalence of ankyloglossia in the general population.

To date, there is no evidence indicating the prevalence of ankyloglossia according to the assessment tool used to diagnose the condition. Different assessment tools resulting in diverging diagnoses can lead to undertreatment or overtreatment. Our review can help clinicians know what prevalence of ankyloglossia they might expect when using a specific tool for the diagnosis in clinical practice. Moreover, this review can provide an estimated rate of ankyloglossia that can serve as the basis for the calculation of the sample size in future epidemiologic studies according to the assessment tool that the researchers have selected for use. Therefore, the aim of our systematic review was to evaluate the prevalence of ankyloglossia in the general population of infants, children, and adolescents according to different assessment tools.

METHODS

Registration and protocol

The protocol for our review was registered a priori in the International Prospective Register of Systematic Reviews database (CRD42021224934). There was 1 change from the original protocol: the exclusion of case-control studies from the sample of studies included.

Eligibility criteria

This review was conducted and reported following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses 2020 statement.[13]

The research question was: What is the prevalence of ankyloglossia among the general population of infants, children, and adolescents according to different assessment tools (population: infants, children, and adolescents; condition: ankyloglossia; context: diagnosis according to different assessment tools)?

The inclusion criteria were observational studies (cross-sectional, cohort) that evaluated the prevalence of ankyloglossia using any assessment tool. We excluded studies with adults, patients with syndromes and conditions that could affect craniofacial development (for example, cleft palate and microcephaly), studies including only people with breast-feeding or speech problems, patients who underwent frenectomy or frenotomy, studies not reporting the prevalence of ankyloglossia, studies using the same sample as another included study, case-control studies, interventional studies, case reports or series, reviews, letters, and editorials.

Search strategy

The following databases were searched from the inception through November 1, 2021: MEDLINE (Ovid), Embase (Ovid), Scopus, Web of Science, Cochrane Database of Systematic Reviews, Latin American and Caribbean Health Sciences Literature, and the Brazilian Library of Dentistry through the Virtual Health Library (Bireme, Latin America). The gray literature was searched through OpenGrey and ProQuest Dissertation and Abstracts. No restrictions were imposed regarding language or year of publication. The reference lists of the studies selected were hand searched in an attempt to find further studies not retrieved during the electronic search. Specific search strategies were created for each database, which were verified by an expert in systematic reviews (C.C.M.). The search strategies are presented in Appendix 1, available online at the end of this article.

Selection process

The studies retrieved during the electronic searches were exported to EndNote software Version 20 (Clarivate), and all duplicates were removed. Five independent reviewers (P.V.C., A.C.S.-O., S.Q.N., I.G.P.O.-A., R.M.M.) organized in pairs selected studies. The studies were screened initially via title and abstract. If an article met the inclusion criteria in the title or abstract, the full text was

ABBREVIATION KEY

BTAT: Bristol Tongue Assessment Tool.
HATLFF: Hazelbaker Assessment Tool for Lingual Frenulum Function.

retrieved for further analysis. Disagreements in all phases were resolved by means of discussion and consensus. If disagreements persisted, the senior author (C.C.M.) made the final decision. Before each phase, the reviewers underwent a training process conducted by the principal investigator (P.V.C.) using a sample of 10% of the studies.

Data extraction

The 5 paired independent reviewers extracted data using a spreadsheet created on Microsoft Excel. Before data extraction, the reviewers underwent training and calibration exercises conducted by the principal investigator and senior author. Disagreements were solved by means of discussion and consensus. The senior author cross-checked all extracted data: language of publication, geographic region of the authors and sample, year of publication, data collection setting, sample, sex distribution, age group, funding, conflicts of interest, assessment tool, and number of patients with ankyloglossia.

Methodological quality

The independent reviewers assessed the methodological quality of studies using the Joanna Briggs Institute Critical Appraisal Tool checklist for prevalence studies.[14] This tool has 9 domains, each of which is judged as "yes," "no," "does not apply," or "unclear." The reviewers previously underwent training and calibration exercises conducted by the principal investigator and senior author. Disagreements were discussed until a consensus was reached. If a disagreement persisted, the senior author was consulted to make the final decision.

Synthesis of results

We used Stata statistical software Version 12 (StataCorp) for meta-analysis of proportion, using a random-effects model owing to the inherent heterogeneity among different populations.[15] We used the I^2 statistic to measure heterogeneity among the studies and calculated τ^2. One study reported prevalence of ankyloglossia equal to 0.[16] Therefore, we used the Metaprop command in Stata with double arcsine method to allow the inclusion of studies with prevalence equal to 0.[17]

For the meta-analysis of pooled crude prevalence data, we extracted the final sample and number of patients with ankyloglossia. One cohort study collected data at baseline and after 1 month of follow-up.[18] As the aim was to report the prevalence rather than incidence of ankyloglossia, we only considered the baseline data when the sample was larger. We calculated overall pooled crude prevalence estimates and corresponding 95% CIs. To explain the heterogeneity in the model, we ran a random-effects meta-regression. The independent variable was the year of publication. The dependent variable was the prevalence of ankyloglossia of pooled crude effect estimates.

We subgrouped the prevalence of ankyloglossia by assessment tool, geographic region, and age group (infants, children, adolescents) as reported by the authors using a mixed-effects model. We categorized studies reporting their own criteria and studies that did not clearly describe the diagnosis as using an unspecific tool. We considered other assessment tools as specific tools and reported them separately. Coryllos and Kotlow classifications classify all cases as a certain degree of ankyloglossia.[8,9] To be consistent among studies, we considered as "ankyloglossia cases" the moderate and severe degrees of ankyloglossia (anterior ankyloglossia), which was possible for some assessment tools (Coryllos and Kotlow classification). The interference in tongue movements was also considered as an "ankyloglossia case" (for example, HATLFF) unless the authors did not categorize ankyloglossia. In this case, we extracted the total number of patients with ankyloglossia as reported by the authors. We also extracted the total number of cases reported by the authors in the following instances: studies using unspecific tools that did not describe criteria, studies that did not agree as severe and moderate cases, or studies that only reported the overall prevalence. Therefore, we followed the authors' classification to avoid selective bias. The list of reported assessment tools and detailed descriptions are displayed in Appendix 2, available online at the end of this article.

We treated studies with a large epidemiologic database, such as national or province sample, separately as a subgroup because the prevalence of ankyloglossia could differ from that of studies involving samples from local universities, hospitals, schools, or single cities. To test the robustness of the overall prevalence of ankyloglossia, we performed a further subgroup analysis by geographic region of the sample, followed by a sensitivity test excluding studies using specific tools.

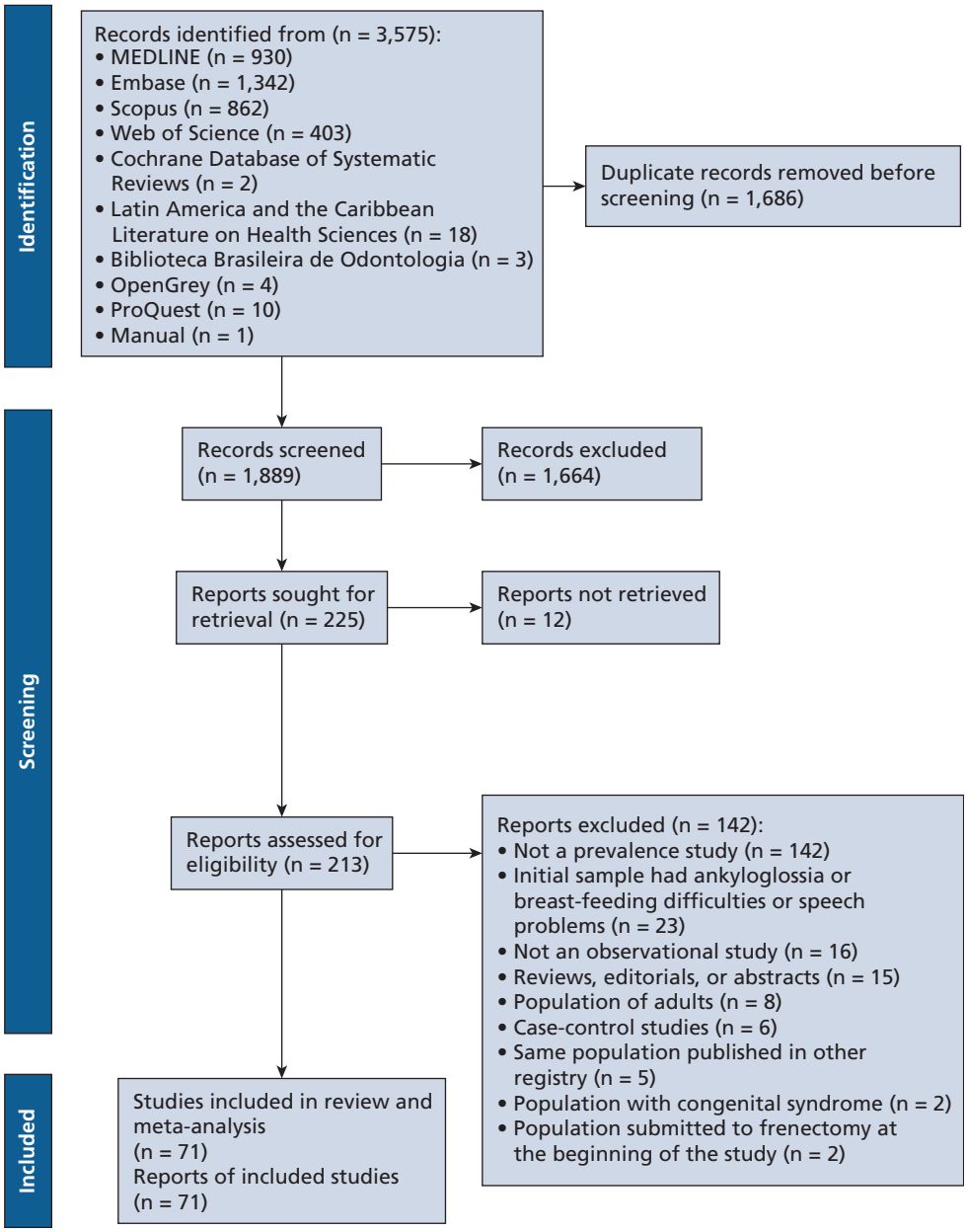

Figure 1. Flowchart showing the screening process, according to Preferred Reporting Items for Systematic Reviews and Meta-Analyses.[13]

We also investigated the occurrence of ankyloglossia stratified by sex. We calculated the pooled crude prevalence of ankyloglossia for boys and girls using a random-effects model. We used the number of patients with ankyloglossia per sex and the total number of boys and girls to calculate the effect estimates. We used the total number of boys and girls and the number of patients with ankyloglossia per sex to calculate the prevalence ratio and corresponding 95% CI. Lastly, we evaluated publication bias using the estimated log of the effect estimate in a funnel plot and Egger test.

Certainty of evidence

Two reviewers (P.V.C., C.C.M.) assessed the certainty of the evidence for the prevalence ratio of ankyloglossia between boys and girls using the Grading of Recommendations Assessment, Development and Evaluation approach. The certainty of evidence from observational studies starts low and can be downgraded owing to problems related to risk of bias, indirectness, imprecision, inconsistency, and publication bias or upgraded on the basis of magnitude of the effect, a dose-response effect, and residual confounding variables.[19] The results are displayed in the summary of

Table 1. Summary of characteristics of included studies.

CHARACTERISTIC	STUDIES, NO. (%)*
Language	
English	66 (93.0)
Spanish	3 (4.2)
Portuguese	2 (2.8)
Geographic Region of Authors	
Latin America	19 (26.8)
North America	14 (19.7)
Asia	12 (16.9)
Europe	8 (11.3)
Middle East	8 (11.3)
Collaboration between > 1 country	7 (9.8)
Africa	3 (4.2)
Geographic Region of Sample	
Latin America	22 (31.0)
North America	14 (19.7)
Asia	13 (18.3)
Europe	9 (12.7)
Middle East	9 (12.7)
Africa	3 (4.2)
Oceania	1 (1.4)
Year of Publication	
2011 and after	47 (66.2)
2000-2010	17 (23.9)
1999 and earlier	7 (9.9)
Setting	
Hospital or dental school	42 (59.2)
School	16 (22.5)
Large or national database	5 (7.1)
Dental clinic	3 (4.2)
Residence	3 (4.2)
Not reported	2 (2.8)
Funding	
Government or university grant	15 (21.1)
No funding	13 (18.3)
Industry	2 (2.8)
Not reported	41 (57.8)
Conflict of Interest	
Authors declare no conflicts of interest	28 (39.4)
Potential conflicts of interest	1 (1.4)
Not reported	42 (59.2)

* Total number of studies, 71 (100%). † Not all studies reported sample distribution per sex.

Table 1. Continued

CHARACTERISTIC	STUDIES, NO. (%)*
Final Sample	
Minimum (absolute number)	21
Maximum (absolute number)	37,990,863
Total (absolute number)	41,865,393
Sex†	
Total number of girls (absolute number)	40,606
Total number of boys (absolute number)	45,062

findings table (eTable 1, available online at the end of this article) created using the GRADEpro software for binary outcomes. We did not assess the certainty of the evidence for prevalence of ankyloglossia as there was no comparison between groups.[20]

RESULTS

Study selection

Figure 1 shows the screening process for the selection of articles based on the titles, abstracts, and full texts. We included 71 studies in the systematic review (70 cross-sectional studies, 1 cohort study).[3,16,18,21-88] The list of studies included is presented in Supplementary Reference List, available online at the end of this article. Excluded studies and the reasons for exclusion are described in eTable 2, available online at the end of this article.

Study characteristics

The characteristics of the 71 studies are summarized in Table 1. Ninety-three percent of the studies were published in the English language, and 66.2% were published from 2011 and after. The samples were from all geographic regions: Latin America (31.0%), North America (19.7%), Asia (18.3%), Europe (12.7%), Middle East (12.7%), Africa (4.2%), and Oceania (1.4%). The samples were recruited from hospitals or dental schools (59.2%), schools (22.5%), large or national databases (7.1%), dental clinics (4.2%), and residences (4.2%). A total of 21.2% of the studies were funded by a university or governmental grant, and 39.4% declared no potential conflict of interests. eTable 3, available online at the end of this article, shows the detailed study characteristics.

Methodological quality

The methodological quality of the studies is reported in Table 2 and eFigure 1, available online at the end of this article. The major methodological problems were related to item 1 (appropriateness of the sample to address the target population: 52.1% judged as "no"), item 3 (49.3% of studies did not have an adequate sample size), and item 6 (60.6% of studies did not use validated methods for the diagnosis of ankyloglossia). There were also several unclear domains, such as item 2 (unclear recruitment of participants, 49.2%), item 5 (coverage bias, 54.9%), and item 7 (52.1% of studies did not report whether the condition was measured in a standard, reliable way for all participants).

Meta-analysis

Seventy-one studies were included in the meta-analysis. The analysis included 77, as some studies stratified the prevalence of ankyloglossia into more than 1 assessment tool or different age groups. Table 3 and Figure 2 show the prevalence of ankyloglossia according to the assessment tool used. The overall prevalence of ankyloglossia was 5% (95% CI, 4.0% to 5.0%; eFigure 2, available online at the end of this article). In the subgroup analyses, the prevalence of ankyloglossia was lower when studies used an unspecific tool (3%; 95% CI, 2.0% to 4.0%) than when some types of tools were used. The prevalence was highest when using the Coryllos classification (20%; 95% CI, 9.0% to 35.0%), followed by HATLFF combined with the Coryllos classification (18%; 95% CI, 6.0% to 34.0%). The prevalence was 9% using Kotlow classification (95% CI, 5.0% to 14.0%). The prevalence of ankyloglossia was approximately 11% through 9% for BTAT and Neonatal Tongue

Table 2. Summary of methodological quality using Joanna Briggs Institute Critical Appraisal Tool[14] checklist for prevalence studies.

DOMAIN	YES, NO. (%)	UNCLEAR, NO. (%)	NO, NO. (%)
1. Was the Sample Frame Appropriate to Address the Target Population?	16 (22.5)	18 (25.4)	37 (52.1)
2. Were Study Participants Recruited in an Appropriate Way?	18 (25.4)	35 (49.2)	18 (25.4)
3. Was the Sample Size Adequate?	28 (39.4)	8 (11.3)	35 (49.3)
4. Were the Study Subjects and Setting Described in Detail?	37 (52.2)	17 (23.9)	17 (23.9)
5. Was Data Analysis Conducted With Sufficient Coverage of the Identified Sample?	17 (24.0)	39 (54.9)	15 (21.1)
6. Were Valid Methods Used for the Identification of the Condition?	26 (36.6)	2 (2.8)	43 (60.6)
7. Was the Condition Measured in a Standard, Reliable Way for All Participants?	18 (25.4)	37 (52.1)	16 (22.5)
8. Was There Appropriate Statistical Analysis?	68 (95.8)	0 (0.0)	3 (4.2)
9. Was the Response Rate Adequate, and if Not, Was the Low Response Rate Managed Appropriately?	16 (22.5)	45 (63.4)	10 (14.1)

Screening Test, respectively, used separately. The prevalence was less than 6% using other tools. The prevalence of ankyloglossia in studies comprising a large database was 2% (95% CI, 1.0% to 3.0%).

The meta-regression model showed that the year of publication had a positive effect on the crude prevalence of ankyloglossia. The prevalence increased by 0.003 points with each year (adjusted R^2, 6.21%; τ^2, 0.01439; coefficient, 0.0032498; SE, 0.0013592; $P = .019$).

Table 4 shows the subgroup analysis for geographic region. Considering all tools, the prevalence of ankyloglossia ranged from 7% (95% CI, 2.0% to 14.0%) in Europe to 0% in Africa. For the sensitivity analysis, we kept large database studies in the model, as the overall prevalence was similar regardless of whether the study included a large database (2%) or local sample (3%). Considering only unspecific tools in the meta-analysis, the prevalence decreased a little, ranging from 6% (95% CI, 0.0% to 17.0%) in Asia to 2% (95% CI, 1.0% to 3.0%) in Europe (no studies from Africa and Oceania were excluded, and the estimates remained the same).

Table 5 shows the meta-analyses per sex and age group. Thirty-one studies reported the prevalence of ankyloglossia by sex and were included in the meta-analysis. The overall crude prevalence of ankyloglossia was similar for boys (6%) (95% CI, 3.0% to 8.0%) and girls (4%; 95% CI, 2.0% to 6.0%). The prevalence ratio indicated that the prevalence of ankyloglossia was 34% higher among boys than girls (95% CI, 1.17% to 1.54%; very low certainty of evidence, eTable 1, available online at the end of this article). There were very serious problems related to the risk of bias and inconsistency. The funnel plot showed slight asymmetry, and Egger test was not significant regarding publication bias ($P = .062$, eFigure 3, available online at the end of this article).

The subgroup analysis by age group revealed the prevalence of ankyloglossia to be 7% (95% CI, 6.0% to 8.0%) for infants, 1% (95% CI, 0.0% to 3.0%) for children, and 2% (95% CI, 0.0% to 4.0%) for adolescents. eFigures 2 and 4-12, available online at the end of this article, show all forest plots.

DISCUSSION

Our findings on the overall prevalence of ankyloglossia are in agreement with the findings of a previous systematic review, which reported the prevalence to be 8%.[12] However, the authors of that review included only 15 studies involving infants up to 1 year of age.[12] In contrast, we included all age groups. Nonetheless, the prevalence of ankyloglossia among infants reported in our review (7%) is close to that reported in the previous systematic review.[12] As no other systematic reviews, to our knowledge, have reported the prevalence of ankyloglossia among children and adolescents, there is no evidence to compare with our data. The prevalence of ankyloglossia among children, adolescents, or mixed-age groups is lower than that among infants. Some reasons can explain the higher rate of ankyloglossia among infants. We excluded studies in which the participants underwent

Table 3. Pooled prevalence of ankyloglossia according to different assessment tools.

ASSESSMENT TOOL FOR DIAGNOSING ANKYLOGLOSSIA	STUDIES, NO (%)	I^2, *P* VALUE	EFFECT ESTIMATE (PREVALENCE),* % (95% CI)	MIXED-EFFECTS† *z* TEST; *P* VALUE
1 Assessment Tool Used				
Coryllos classification	3 (3.9)	Not estimated	20 (9.0 to 35.0)	5.12; < .001†
Kotlow classification	6 (7.8)	95.4%, < .001	9 (5.0 to 14.0)	6.71; < .001†
BTAT‡	1 (1.3)	Not estimated	11 (9.0 to 13.0)	19.02; < .001†
Neonatal Tongue Screening Test	9 (11.7)	98.8%, < .001	9 (3.0 to 19.0)	4.22; < .001†
HATLFF§ modified	1 (1.3)	Not estimated	6 (4.0 to 9.0)	8.15; < .001†
Lingual Frenulum Protocol for Infants	1 (1.3)	98.7%, < .001	5 (3.0 to 8.0)	6.94; < .001†
HATLFF	2 (2.6)	Not estimated	4 (4.0 to 5.0)	32.04; < .001†
Unspecific tool¶	42 (54.5)	99.4%, < .001	3 (2.0 to 4.0)	8.25; < .001†
Unspecific tool, populational#	6 (7.8)	99.9%, < .001	2 (1.0 to 3.0)	6.48; < .001†
Academy of Breastfeeding Medicine	1 (1.3)	Not estimated	0 (0.0 to 0.0)	15.07; < .001†
Combination of > 1 Tool				
HATLFF and Coryllos classification	3 (3.9)	Not estimated	18 (6.0 to 34.0)	4.35; .01†
HATLFF modified and BTAT	1 (1.3)	Not estimated	4 (1.0 to 8.0)	4.12; .01†
Lingual Frenulum Protocol for Infants and BTAT	1 (1.3)	Not estimated	3 (2.0 to 5.0)	6.65; < .001†
Overall Prevalence**	77 (100)	99.9%, < .001; τ^2, 0.01††	5 (4.0 to 5.0)	31.73; < .001

* Prevalence. † Mixed-effects model used for subgroup analysis. ‡ BTAT: Bristol Tongue Assessment Tool. § HATLFF: Hazelbaker Assessment Tool for Lingual Frenulum Function. ¶ Unspecific tool when the authors did not describe the diagnostic tool or used their own criteria for the study. # Unspecific tool used in population-based studies. ** Random-effects model for overall prevalence. The number of studies is greater than 71 because some studies described the prevalence for more than 1 assessment tool. †† For overall prevalence.

frenectomy or frenotomy. Therefore, the higher rate among infants is probably not due to treatment. Studies that included exclusively infants tend to use more specific tools than studies including children and adolescents. In analyzing our data, from 42 studies that included only infants, 19 studies used unspecific tools, and 23 used a specific tool. Among these, some assessment tools were responsible for higher rates of ankyloglossia among infants: Coryllos classification (20%), HATLFF and Coryllos (18%), and BTAT (11%). Contrarily, of the 8 studies that included only children, only 1 study used the Kotlow classification, showing a prevalence of 5%. One of 5 studies including only adolescents showed a prevalence of 3% using the Kotlow classification. The other studies including children and adolescents used unspecific tools (prevalence approximately 1%-4%, eFigures 10-12, available online at the end of this article). Moreover, we raise the question whether ankyloglossia is a condition that remains as a person becomes older or whether the tongue frenulum undergoes a change in its position with craniofacial development. A cohort study found limited evidence to answer this question, as the infants were reexamined after 1 through 3 months; the prevalence was 2.2% at baseline and 2.3% at follow-up.[18] We found no cohort studies that followed infants through to childhood or adolescence.

Although year of publication had a positive effect on the prevalence of ankyloglossia, it is unlikely that this result is due to an increased occurrence of ankyloglossia in the last decade. Moreover, the clinical relevance of the coefficient (0.0031569) is questionable. We could assume, however, an increased interest in diagnosing ankyloglossia over the years and its possible consequences with regard to breast-feeding. This theory is supported by our data, as 66.2% of the studies included in our review were published in 2011 or after.

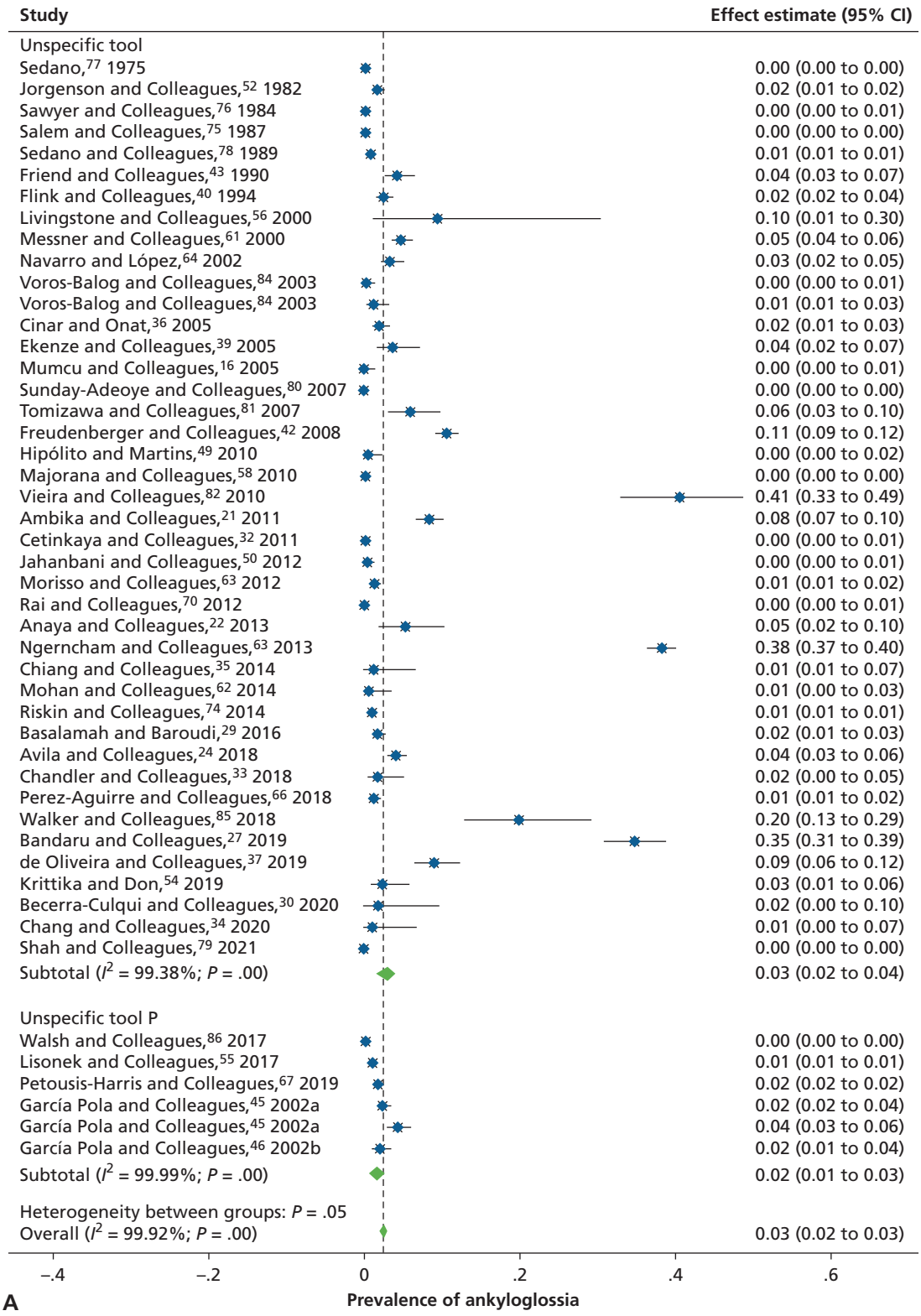

Figure 2. Mixed-effects meta-analysis of ankyloglossia subgrouped by assessment tool. **A.** Prevalence: effect estimate. Unspecific criteria: authors used their criteria or did not describe the diagnosis. Unspecific criteria P (populational study). Unspecific tool: $\chi^2_{41} = 6{,}607.06$; $z = 8.25$; $P = .000$; $I^2 = 99.38\%$. Unspecific tool P: $\chi^2_5 = 49{,}467.55$; $z = 6.48$; $P = .000$; $I^2 = 99.99\%$. Overall: $\chi^2_{47} = 57{,}003.96$; $z = 19.03$; $P = .000$; $I^2 = 99.92\%$. Test for heterogeneity between subgroups: overall: $\tau^2 = 3.86$. $\chi^2_{(\text{degrees of freedom})} = \chi^2$ test for I^2 test. **B.** Prevalence: effect estimate. Specific criteria: any criteria already reported on literature. Lingual Frenulum Protocol for Infants and BTAT (Bristol Tongue Assessment Tool): $\chi^2_0 =$ not estimated; $z = 6.65$, $P = .000$; $I^2 =$ not estimated. Kotlow: $\chi^2_5 = 109.02$; $z = 6.71$; $P = 0.00$; $I^2 = 95.41\%$. HATLFF (Hazelbaker Assessment Tool for Lingual Frenulum Function): $\chi^2_1 =$ not estimated; $z = 32.04$, $P = .000$; $I^2 =$ not estimated. HATLFF and Coryllos: $\chi^2_2 =$ not estimated; $z = 4.35$; $P = .000$; $I^2 =$ not estimated. Neonatal Tongue Screening Test: $\chi^2_8 = 666.03$; $z = 4.22$, $P = .000$; $I^2 = 98.80\%$. BTAT: $\chi^2_0 =$ not estimated; $z = 19.02$, $P = .000$; $I^2 =$ not estimated. Coryllos: $\chi^2_2 =$ not estimated; $z = 5.12$, $P = .000$; $I^2 =$ not estimated. Academy of Breastfeeding Medicine: $\chi^2_0 =$ not estimated; $z = 15.07$, $P = .000$; $I^2 =$ not estimated. modified HATLFF: $\chi^2_0 =$ not estimated; $z = 8.15$, $P = .000$; $I^2 =$ not estimated. modified HATLFF and BTAT: $\chi^2_0 =$ not estimated; $z = 4.12$, $P = .000$; $I^2 =$ not estimated. Lingual Frenulum Protocol for Infants: $\chi^2_0 =$ not estimated; $z = 6.94$, $P = .000$; $I^2 =$ not estimated. Overall: $\chi^2_{28} = 5471.48$; $z = 32.04$, $P = .000$; $I^2 = 99.49\%$. Test for heterogeneity between subgroups: overall: $\tau^2 = 885.34$. $\chi^2_{(\text{degrees of freedom})} = \chi^2$ test for I^2 test.

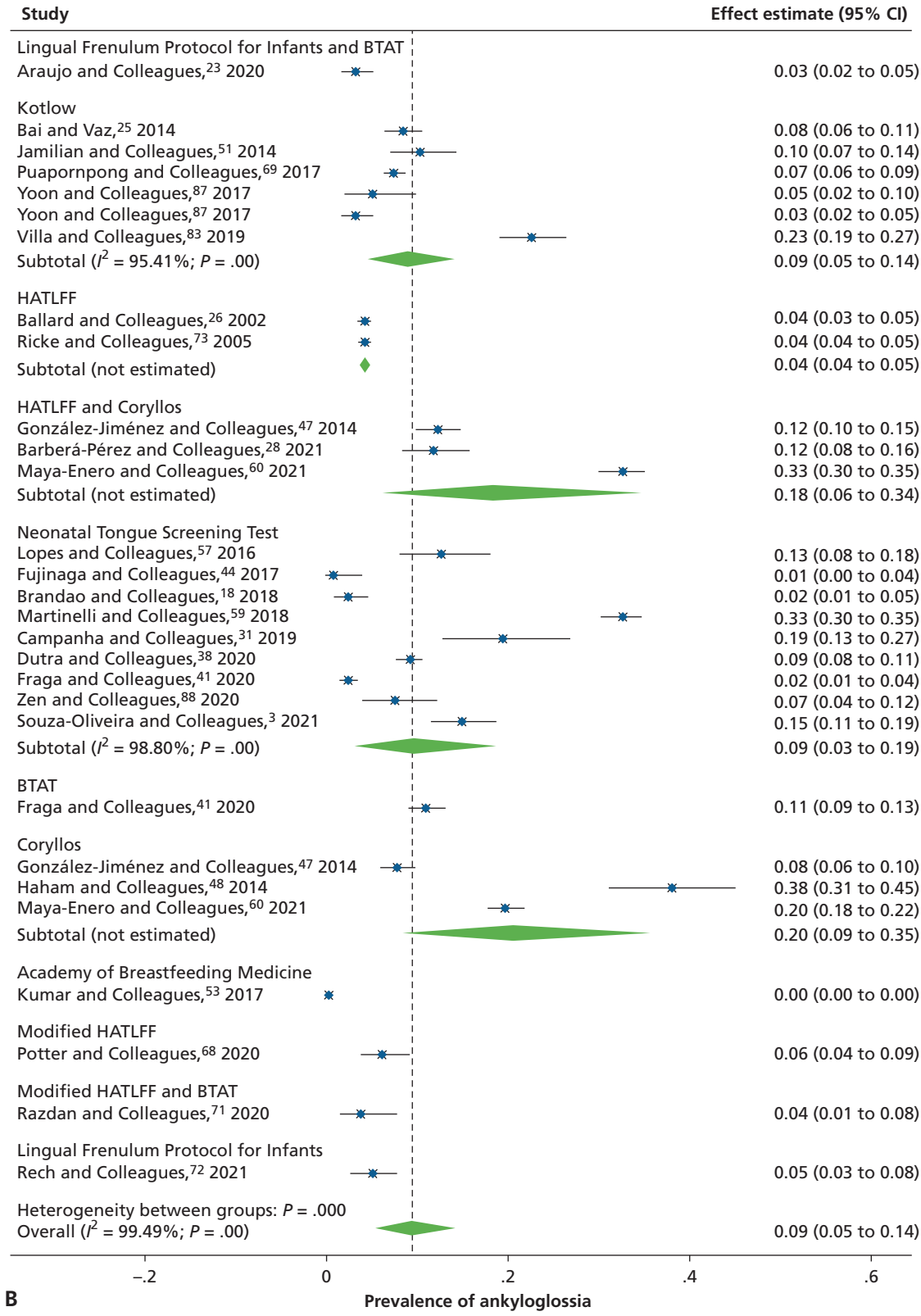

Figure 2. (Continued)

There are differences in grading systems among the specific tools. We considered anterior ankyloglossia for Coryllos and Kotlow classifications to make all data more consistent among studies. For Coryllos classification, anterior ankyloglossia is type 1 (frenulum attached from the tip

Table 4. Subgroup meta-analysis of pooled prevalence of ankyloglossia by geographic region of sample population.

GEOGRAPHIC REGION	STUDIES, NO. (%)	I^2, *P* VALUE	EFFECT ESTIMATE,* % (95%CI)	MIXED-EFFECTS† *z* TEST, *P* VALUE
Considering All Tools	77 (100)	NA‡	NA	NA
Latin America	23 (29.9)	98.2%, < .001	7 (4.0 to 11.0)	6.74, < .001†
North America	15 (19.5)	99.9%, < .001	3 (2.0 to 4.0)	10.45, < .001†
Asia	13 (16.9)	99.8%, < .001	5 (1.0 to 12.0)	3.42, < .001†
Europe	13 (16.9)	99.5%, < .001	7 (2.0 to 14.0)	4.14, < .001†
Middle East	9 (11.6)	97.9%, < .001	3 (1.0 to 5.0)	5.01, < .001†
Africa	3 (3.9)	Not estimated	0 (0.0 to 1.0)	2.12, .03†
Oceania	1 (1.3)	Not estimated	2 (2.0 to 2.0)	70.9, < .001†
Considering Unspecific Tools§	48 (100)	NA	NA	NA
Latin America	11 (22.9)	97.8%, < .001	5 (2.0 to 8.0)	5.68, < .001†
North America	9 (18.8)	99.9%, < .001	2 (1.0 to 3.0)	6.18, < .001†
Asia	10 (20.8)	99.7%, < .001	6 (0.0 to 17.0)	2.20, < .001†
Europe	7 (14.6)	96.2%, < .001	2 (1.0 to 3.0)	4.01, < .001†
Middle East	7 (14.6)	91.9%, < .001	1 (0.0 to 1.0)	4.50, < .001†
Africa	3 (6.3)	Not estimated	0 (0.0 to 1.0)	2.12, .24†
Oceania	1 (2.0)	Not estimated	2 (2.0 to 2.0)	70.90, < .001†

* Prevalence. † Mixed-effects model used for subgroup analysis. ‡ NA: Not applicable. § Sensitivity analysis considering only unspecific tools (when the authors did not describe the diagnostic tool or used their own criteria for the study; also included large database samples). The number of studies is greater than 71 because some studies described the prevalence for more than 1 assessment tool.

of the tongue to the alveolar ridge) and type 2 (frenulum attached up to 2-4 mm behind the tip of the tongue).[8] When adding type 3 (frenulum attached from the middle of the tongue to the middle of the floor of mouth) and type 4 (frenulum attached against the base of the tongue),[8] the prevalence would be higher (≈ 45%). Kotlow classification considers the length of the tongue and the tongue movement. We considered class II (moderate, 8-11 mm), class III (severe, 3-7 mm), and class IV (complete, < 3 mm).[9] However, there was a significant variation among studies when using the other tools. For example, for HATLFF, 1 study considered significant ankyloglossia when the appearance score was 8 or below or the function score was 11 or below.[26] Another study considered ankyloglossia as perfect function (score 14), acceptable function (score of 11, if appearance was 10), and impaired function (score of 11). In this case, the study did not report the numbers per category but the total number of cases.[73] The lack of agreement between studies was more remarkable for combined tools. Ankyloglossia could be any Coryllos type 1 through 4 and HATLFF score 6 or below,[73] Coryllos types 1 through 4, and HATLFF score of 8[60] or any type of Coryllos classification with HATLFF without specification.[28] Therefore, the prevalence rates reported in our review are estimates that can vary according to the assessment tool and the categorization used by the authors.

Five large epidemiologic studies were grouped apart, comprising large databases from an entire country or province.[45,46,55,67,86] These studies sought to diagnose tongue problems, and all used unspecific tools. Despite differences in representativeness of the sample, the prevalence rates generated from the database studies and studies comprising local samples using unspecific tools were similar. The subgroup meta-analysis allowed us to separate different tools in a way that the unspecific tools would not to alter the prevalence ratio of the specific tools. Moreover, many other studies that used unspecific tools evaluated several oral conditions besides ankyloglossia in children, such as Epstein pearls, Bohn nodules, mucocele,[43] and geographic tongue.[16] The ratio generated by the unspecific tools can give an idea of the prevalence to expect from studies evaluating several oral conditions rather than only ankyloglossia.

We found differences in the prevalence of ankyloglossia per geographic region even after the sensitivity analysis. Considering only unspecific tools, the prevalence of ankyloglossia decreased for most geographic regions except Asia, in which the prevalence increased slightly from 5% to 6%. Two studies reported high prevalence estimates of ankyloglossia and were responsible for the increased prevalence in Asia: Bandaru and colleagues[27] (35%) and Ngerncham and colleagues[65]

Table 5. Subgroup meta-analysis showing pooled prevalence of ankyloglossia by sex and age group.

DIAGNOSTIC CRITERIA	STUDIES, NO. (%)	I^2, *P* VALUE	τ^2	EFFECT ESTIMATE,* % (95% CI)	MIXED-EFFECTS† *z* TEST, *P* VALUE
Sex					
Girls	31	98.5%, < .001	0.06	4 (2.0 to 6.0)	7.50, < .001
Boys	31	98.1%, < .001	0.09	6 (3.0 to 8.0)	7.74, < .001
Girls‡	NA	NA	NA	Prevalence ratio,§ 1	NA
Boys‡	31	38.9%, .013	0.0342	Prevalence ratio, 1.34 (1.17 to 1.54)	4.24, < .001
Age Group¶					
Infants	42 (54.5)	99.6%, < .001	NA	7 (6.0 to 8.0)	16.85; < .001†
Children	8 (10.4)	93.4%, < .001	NA	1 (0.0 to 3.0)	4.34; < .001†
Adolescents	5 (6.5)	90.0%; < .001	NA	2 (0.0 to 4.0)	3.62; < .001†
Infants and children	2 (2.6)	Not estimated	NA	0 (0.0 to 0.0)	5.32; < .001†
Infants, children, and adolescents	2 (2.6)	Not estimated	NA	0 (0.0 to 0.0)	224.69; < .001†
Children and adolescents	18 (23.4)	98.9%, < .001	NA	4 (2.0 to 6.0)	6.56; < .001†
Overall#	77 (100)#	99.9%, < .001	NA	5 (4.0 to 5.0)	31.73; < .001

* Prevalence. † Mixed-effects model for subgroup analysis for age groups. ‡ Random-effects meta-analysis of occurrence of ankyloglossia comparing girls with boys. § Egger test for publication bias. $P = .048$. ¶ Infants were considered neonates to age 1 year; children were considered if the study included participants aged from 1 through 11 years; and adolescents were considered if the study included participants aged from 11 through 19 years. Eleven-year-old participants were not exclusively from 1 group. They could be grouped as children or adolescents according to the other predominant age groups. # Random-effects model for overall prevalence. The number of studies is greater than 71 because some studies stratified the prevalence per age groups.

(38%). The first study did not describe the diagnostic criteria,[27] and the other used simple diagnostic criteria with 3 categories: normal, moderate (frenulum attached up to two-thirds of the tip of the tongue), and severe (complete attachment of the frenulum to the tip of the tongue).[65] This shows that an unspecific tool also can generate an overestimate of the prevalence of ankyloglossia.

Diverse examiners performed the diagnosis of ankyloglossia, and the diagnosis can differ according to the experience and level of education of different examiners. The methodological quality in this item was mainly low (Table 2, item 7). Only 25.4% of the studies reported a calibration and training exercise of the examiners before diagnosing ankyloglossia, and 74.8% of studies did not provide adequate calibration and training for the examiners before the diagnosis. The examiners must know the anatomy and physiology of the lingual frenulum before making the diagnosis. There is vast variability of the lingual frenulum related to the midline attachment of the floor of mouth fascia and variability in how the mucosa, fascia, and genioglossus are drawn into the floor of the frenulum. The clinician should know these variations before making the diagnosis and, most importantly, before performing the frenectomy, given the risk of permanent injury of the lingual nerve branches.[89]

It is uncertain whether boys are more prone to ankyloglossia than girls. We found very serious problems of inconsistency owing to the statistically significant heterogeneity in the meta-analysis. eFigure 8 shows that some studies favored more ankyloglossia among boys, whereas others favored girls. This difference may be due to the varying assessment tools used, different age groups, and geographic regions of the sample. We also found very serious problems due to the low methodological quality of the studies, especially the representativeness of the cases and a lack of valid criteria for measuring ankyloglossia.

Strengths and limitations

To our knowledge, our review is the first complete review summarizing the prevalence of ankyloglossia, including several different tools for the diagnosis. The strengths of our review are the methodological rigor shown by the subgroup analyses, investigation of publication bias using funnel plots and Egger test, meta-regression, sensitivity analysis, and Grading of Recommendations Assessment, Development and Evaluation approach to assess the certainty of the evidence. As limitations, the prevalence ratio calculated for boys and girls could have resulted in different effect estimates if case-controls studies had been included. All studies had low methodological quality.

Thus, sensitivity analysis was not feasible, as all studies would have been removed from the meta-analysis. Instead, we opted to downgrade the certainty of the evidence by 2 levels owing to the risk of bias. Lastly, although the unspecific tools differed among the studies, we opted to create a single subgroup, risking the loss of information regarding these assessment tools.

Implications for clinical practice and research

This review can serve as the basis for sample size calculations in future epidemiologic studies, enabling authors to choose the prevalence according to each assessment tool. However, there is no reference standard tool for diagnosing ankyloglossia, and no tool was validated. Thus, future clinical studies should seek to determine the best assessment tool for diagnosing ankyloglossia as well as evaluate the reliability and reproducibility of each tool. Future prospective studies should also investigate whether ankyloglossia remains in infants as they become older.

CONCLUSIONS

The actual prevalence of ankyloglossia is unknown, as the prevalence ratio can vary according to the assessment tool. There is a wide range (20% prevalence using Coryllos classification to 2% using an unspecific tool). The prevalence is higher among infants than children and adolescents. Moreover, it is uncertain whether boys are more prone to ankyloglossia than girls. ■

SUPPLEMENTAL DATA

Supplemental data related to this article can be found at https://doi.org/10.1016/j.adaj.2022.07.011.

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APPENDIX 1. SEARCH STRATEGIES USED IN EACH ELECTRONIC DATABASE (FROM INCEPTION THROUGH NOVEMBER 21, 2020, UPDATED ON NOVEMBER 1, 2021).

MEDLINE through Ovid

#1. ankyloglossia.mp. or exp Lingual Frenum/ or exp Ankyloglossia/
#2. tongue-tie.mp.
#3. exp Labial Frenum/ or lip-tie.mp.
#4. oral mucosal lesions.mp.
#5. oral lesions.mp.
#6. 1 or 2 or 3
#7. 1 or 2 or 3 or 4 or 5
#8. exp Prevalence/ or prevalence.mp.
#9. exp Cross-Sectional Studies/ or cross-sectional.mp.
#10. exp Diagnosis/ or diagnosis.mp.
#11. epidemiology.mp. or exp Epidemiology/
#12. incidence.mp. or exp Incidence/
#13. 8 or 9 or 10 or 11 or 12
#14. child*.mp. or exp Child/
#15. newborn*.mp. or exp Infant, Newborn/
#16. infant*.mp. or exp Infant/
#17. bab*.mp.
#18. 14 or 15 or 16 or 17
#19. frenectomy.mp.
#20. exp Ankyloglossia/ or Frenotomy.mp.
#21. 1 or 2 or 3 or 4 or 5 or 19 or 20
#22. 13 and 18 and 21

Embase through Elsevier

#1. ankyloglossia
#2. "tongue-tie"
#3. "lip-tie"
#4. “oral mucosal lesions”
#5. “oral lesions”
#6. frenotomy
#7. frenectomy
#8. prevalence
#9. cross-sectional
#10. diagnosis
#11. epidemiology
#12. incidence
#13. child*
#14. newborn*
#15. infant*
#16. bab*
#17. #1 or #2 or #3 or #4 or #5 or #6 or #7
#18. #8 or #9 or #10 or #11 or #12
#19. #13 or #14 or #15 or #16
#20. #17 and #18 and #19

Scopus

TITLE-ABS-KEY (ankyloglossia OR "tongue-tie" OR frenotomy OR frenectomy OR "oral mucosal lesions" OR "oral lesions") AND TITLE-ABS-KEY (prevalence OR cross-sectional OR diagnosis OR epidemiology OR incidence) AND TITLE-ABS-KEY (child* OR newborn* OR infant* OR bab*)

Web of Science

TS=((ankyloglossia OR "tongue-tie" OR “oral mucosal lesions” OR “oral lesions” or frenectomy OR frenotomy) AND (prevalence OR cross-sectional OR diagnosis OR epidemiology OR incidence) AND (child* OR newborn* OR infant* OR bab*))

Cochrane Systematic Reviews

#1. “ankyloglossia”

#2. MeSH descriptor: [ankyloglossia] explode all trees

#3. frenectomy

#4. frenetomy

#5. #1 or #2 or #3 or #4

Latin American and Caribbean Health Sciences Literature and Brazilian Library of Dentistry through Bireme

(ankyloglossia) AND (diagnosis) AND (child* OR newborn*)

Proquest Dissertation and Abstracts

(ankyloglossia or "tongue-tie" or “lip-tie” or frenetomy or frenectomy)

OpenGrey: 1

Ankyloglossia

APPENDIX 2. DESCRIPTION OF ASSESSMENT TOOLS.

Hazelbaker Assessment Tool for Lingual Frenulum Function (HATLFF)

The HATLFF evaluates the appearance and function of the tongue according to the items described below. Score of 14 indicates perfect score; 11 is acceptable; and below 11 indicates impaired function and frenotomy might be considered if management fails; score below 8 indicates that frenotomy is considered necessary.[e1]

Appearance items:

Appearance of tongue when lifted

2: round or square

1: slight cleft in tip apparent

0: heart-shaped

Elasticity of frenulum:

2: very elastic (excellent)

1: moderately elastic

0: little or no elasticity

Length of the lingual frenulum when tongue is lifted:

2: more than 1 cm or embedded in the tongue

1: 1 cm

0: less than 1 cm

Attachment of lingual frenulum to tongue:

2: posterior to tip

1: at tip

0: notched tip

Attachment of lingual frenulum to inferior alveolar ridge:

2: attached to floor of mouth or well below ridge

1: attached just below ridge

0: attached at ridge

Functional items:

Lateralization:

2: complete

1: body of tongue but not tongue tip

0: none

Lift of tongue:

2: tip to mid-mouth

1: only edges to mid-mouth

0: tip stays at alveolar ridge or rises to mid-mouth only with jaw closure

Extension of tongue:

2: tip over lower lip

1: tip over lower gum only

0: neither of above or anterior or mid-tongue humps

Spread of anterior tongue:

2: complete

1: moderate or partial

0: little or none

Cupping:

2: entire edge, firm cup

1: side edges only, moderate cup

0: poor or no cup

Peristalsis:

2: complete, anterior or posterior (originates at the tip)

1: partial: originating posterior to tip

0: none or reverse peristalsis

Snapback:

2: none

1: periodic

0: frequent or with each suck

Bristol Tongue Assessment Tool (BTAT)

BTAT was developed from 4 most important aspects of the former HATLFF.[e2]

Tongue tip appearance

0: heart shaped

1: slight cleft/ notched

2: rounded

Attachment to the lower gum ridge

0: attached at top of gum ridge

1: attached to inner aspect of gum

2: attached to the floor of mouth

The lift of the tongue

0: minimal tongue lift

1: edges only to mid-mouth

2: full tongue lift to mid-mouth

Protrusion of the tongue

0: tip stays behind gum

1: tip over gum

2: tip can extend over lower lip

The scores for the 4 items range from 0 to 8, and scores 0 to 3 indicated more severe reduction of tongue function.

Coryllos Classification

Ankyloglossia can be classified into 4 types according to the distance between the tip of the tongue and the insertion of the lingual frenulum.[e3]

- *Type 1*: fine elastic frenulum; tongue is attached from the tip of the tongue to the alveolar ridge in the lower lip sulcus;
- *Type 2*: fine elastic frenulum; tongue is attached 2 to 4 mm behind the tip of the tongue to almost near the alveolar ridge;
- *Type 3*: thick fibrous less elastic frenulum; tongue is attached from middle of tongue to the middle of the floor of mouth;
- *Type 4*: the frenulum is thick, shiny and very inelastic, it is essentially against the base of the tongue.

Types 1 and 2 are the most common types, considered "classic" ankyloglossia. The incidence is approximately 75%. Types 3 and 4 are less common and hard to diagnose and, therefore, receive less treatment. Type 4 is more likely to cause difficulty in handling the bolus and dysphagia.

Kotlow Classification

The assessment is based on the length, in millimeters, of the tongue from the insertion of the lingual frenulum into the base of the tongue to the tip of the tongue. The severity of ankyloglossia is classified as follows:[e4]

- *Class I*: mild ankyloglossia, 12 to 16 mm;
- *Class II*: moderate ankyloglossia, 8 to 11 mm;
- *Class III*: severe ankyloglossia, 3 to 7 mm;
- *Class IV*: complete ankyloglossia, less than 3 mm.

A tongue with normal mobility is when the length of the frenulum is greater than or equal to 16 mm. Complete ankyloglossia (class IV) can restrict tongue movement; children with severe ankyloglossia (class III) can adapt to the short attachment. Children with moderate (class II) and mild (class I) are difficult to evaluate, and children might appear to have normal speech.

Neonatal Tongue Screening Test (NTST)

NTST is the anatomic and functional evaluation of the LFPI (for the clinical examination, see below). The authors recommend screening newborns for ankyloglossia using NTST within 48 hours after birth. According to the authors, the early assessment of ankyloglossia can diagnose severe cases that can be referred immediately to the frenotomy. In case of doubt (score 5-6), or when the lingual frenulum is not visible, the authors recommend an additional evaluation after 30 days of life using the full-protocol LFPI.[e5]

When the sum of anatomic and functional evaluation is greater than or equal to 7, there is an interference of the lingual frenulum with the movement of the tongue.

Academy of Breastfeeding Medicine (ABM)

The diagnosis was made in the presence of a sublingual frenulum that changed the appearance or function of the infant's tongue because of its decreased length, lack of elasticity, or attachment too distal beneath the tongue or too close to or onto the gingival ridge.[e6]

Lingual Frenulum Protocol for Infants (LFPI)

The authors recommend screening newborns for ankyloglossia using LFPI after 30 days of life for the cases in which NTST could not determine a precise diagnosis. The tool comprises a breast-feeding questionnaire given to the mother, a clinical examination, and an evaluation of nonnutritive and nutritive sucking habits.[e5]

PART I: CLINICAL EXAMINATION

Anatomo-functional evaluation:

Lip posture at rest:

( ): closed

( ): half-open

( ): open

Tongue posture during crying

( ): midline / elevated

( ): midline with the lateral elevated

( ): down

Tongue shape when elevated during crying

( ): round

( ): V-shaped

( ): heart-shaped

Lingual frenulum

( ): visible

( ): not visible

( ): visible with maneuver

If the lingual frenulum is not visible, go to part II (evaluation of the orofacial functions).

Frenulum thickness

( ): thin

( ): thick

Frenulum attachment to the tongue

( ): midline

( ): between midline to apex

( ): apex

Frenulum attachment to the floor of the mouth

( ): visible from the carundes

( ): visible from the crest

Maneuver: elevate and push back the tongue. If the frenulum is not visible, the infant must be seen by a speech-language pathologist every 2 months for periodic frenulum evaluation.

Best result: 0

Worst result: 12

PART II: EVALUATION OF NONNUTRITIVE SUCKING AND NUTRITIVE SUCKING

1. Nonnutritive sucking (little finger sucking wearing gloss)

1.1 Tongue movement

0: adequate: tongue protrusion, coordinated movements and efficient suction

1: inadequate: restricted tongue protrusion, uncoordinated movement and late suction start

2.Nutritive sucking during breast-feeding (when breast-feeding starts, observe infant sucking during 5 minutes)

2.1 Sucking rhythm

0: several suctions in a row followed by short pause

1: a few suctions followed by long pauses

2.2 Coordination among suction/swallowing/breathing

0: adequate: balance between feeding and suction-swallowing-breathing without stress

1: inadequate: cough, choking, dyspnea, regurgitation, hiccup, noises during deglutition

2.3 Nipple chewing

0: no

1: yes

2.4 Clicking during sucking

0: no

1: non-systematic

2: frequent

Non-nutritive sucking and nutritive sucking evaluation scores:

Best result: 0

Worst result: 7

History + clinical examination total scores:

Best results: 0

Worst results: 27

When the sum of history and clinical examination is equal to or more than 9, the authors suggest considering frenotomy.

	Domain								
Study	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**
Sedano,[e65] 1975	Yes	Unclear	Unclear	Yes	Yes	No	Yes	Yes	No
Jorgenson and Colleagues,[e39] 1982	No	Unclear	Unclear	Unclear	Unclear	No	No	Yes	Yes
Sawyer and Colleagues,[e64] 1984	Unclear	Unclear	No	No	Unclear	No	No	Yes	Unclear
Salem and Colleagues,[e63] 1987	Unclear	Unclear	Unclear	No	Unclear	Unclear	No	Yes	Yes
Sedano and Colleagues,[e66] 1989	Yes	Unclear	Yes	Unclear	Yes	No	Yes	Yes	No
Friend and Colleagues,[e30] 1990	No	Unclear	No	No	Unclear	No	Unclear	Yes	Unclear
Flink and Colleagues,[e27] 1994	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes
Livingstone and Colleagues,[e43] 2000	No	Yes	Yes	Unclear	No	No	No	Yes	Unclear
Messner and Colleagues,[e48] 2000	No	Unclear	No	Unclear	Unclear	No	Unclear	Yes	Unclear
Ballard and Colleagues,[e12] 2002	No	Unclear	No	No	Unclear	Yes	Unclear	Yes	Unclear
Garcia-Pola and Colleagues,[e32] 2002a	No	Unclear	No	No	Unclear	No	Unclear	Yes	Unclear
Garcia-Pola and Colleagues,[e33] 2002b	Yes	Yes	Yes	Yes	Yes	No	Unclear	Yes	No
Navarro and López,[e52] 2002	Unclear	Unclear	No	No	Unclear	No	Unclear	No	Unclear
Voros-Balog and Colleagues,[e73] 2003	No	No	No	Yes	Unclear	No	Unclear	Yes	Unclear
Cinar and Onat,[e23] 2005	No	Unclear	No	No	No	No	Unclear	Yes	Unclear
Ekenze and Colleagues,[e26] 2005	Unclear	No	No	No	No	No	Unclear	Yes	No
Mumcu and Colleagues,[e51] 2005	Yes	Yes	Yes	Yes	Unclear	No	Unclear	Yes	No
Ricke and Colleagues,[e61] 2005	Unclear	Unclear	Unclear	Yes	Unclear	Yes	Yes	Yes	Yes
Sunday-Adeoye and Colleagues,[e69] 2007	No	Yes	Yes	Unclear	No	No	Unclear	Yes	Unclear
Tomizawa and Colleagues,[e71] 2007	No	Unclear	No	No	Unclear	No	Unclear	Yes	Yes
Freudenberger and Colleagues,[e29] 2008	Unclear	Unclear	No	Unclear	No	No	No	Yes	Unclear
Hipólito and Martins,[e36] 2010	No	No	No	No	Unclear	No	Unclear	Yes	Unclear
Majorana and Colleagues,[e45] 2010	Unclear	Yes	Yes	Yes	Unclear	No	Yes	Yes	Unclear
Vieira and Colleagues,[e70] 2010	Unclear	Yes	No	Yes	Yes	Unclear	Unclear	Yes	Unclear
Ambika and Colleagues,[e7] 2011	No	No	No	Yes	Unclear	No	Unclear	Yes	Unclear
Çetinkaya and Colleagues,[e19] 2011	No	Unclear	Yes	Yes	Yes	No	Yes	Yes	Unclear
Jahanbani and Colleagues,[e37] 2012	Yes	Yes	Yes	Yes	Yes	No	Unclear	Yes	Unclear
Morisso and Colleagues,[e50] 2012	Unclear	Unclear	No	Unclear	Unclear	No	No	Yes	Unclear
Rai and Colleagues,[e58] 2012	No	No	No	No	No	No	Unclear	No	No

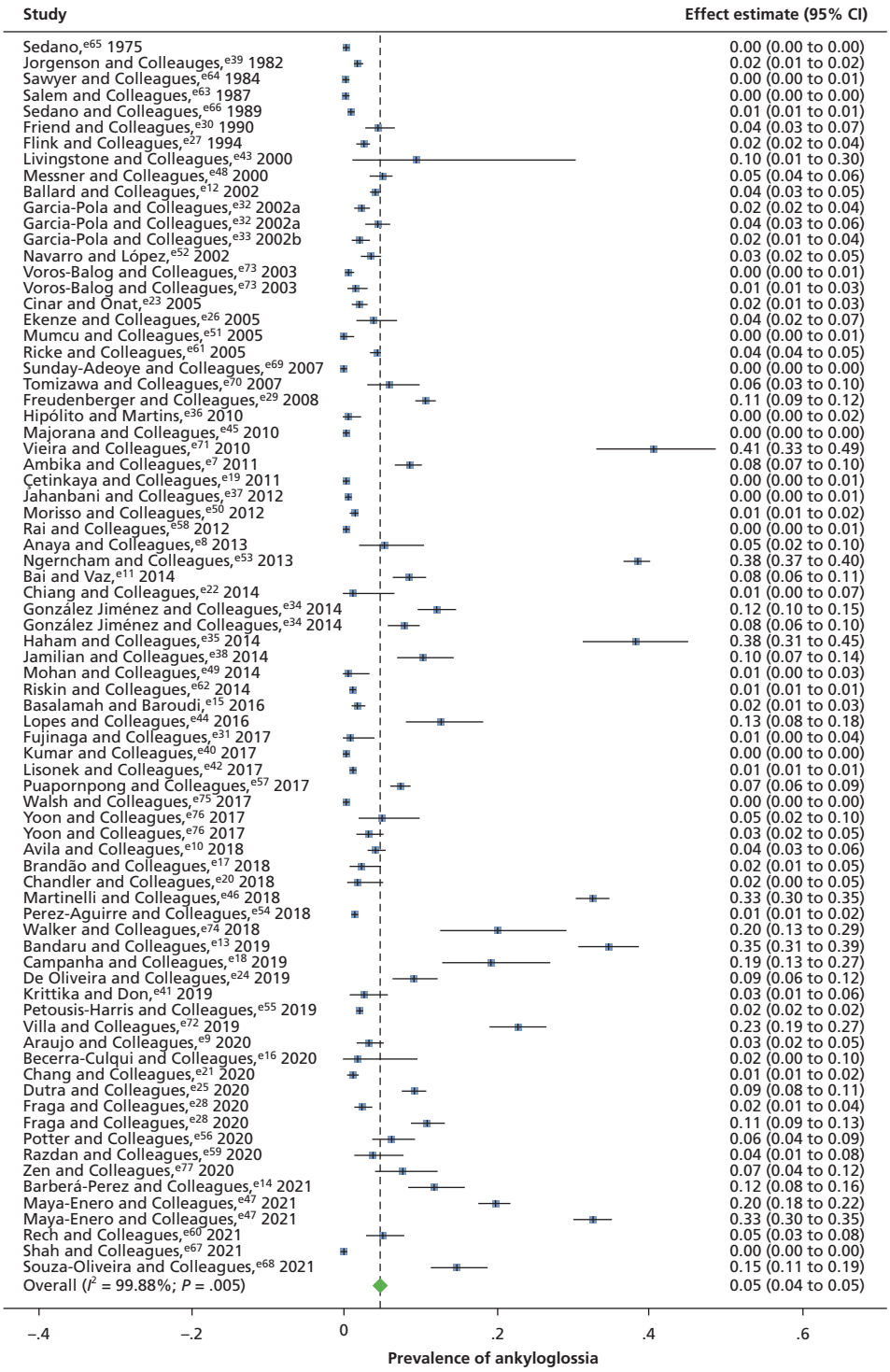
eFigure 1. Methodological quality using Joanna Briggs Institute Critical Appraisal Tool[14] for prevalence studies. Low methodological quality is represented in dark blue (no); unclear is represented in light blue; high quality is represented in green (yes). 1. Was the sample frame appropriate to address the target population? 2. Were study participants recruited in an appropriate way? 3. Was the sample size adequate? 4. Were the study subjects and setting described in detail? 5. Was data analysis conducted with sufficient coverage of the identified sample? 6. Were valid methods used for the identification of the condition? 7. Was the condition measured in a standard, reliable way for all participants? 8. Was there appropriate statistical analysis? 9. Was the response rate adequate, and if not, was the low response rate managed appropriately?

Anaya and Colleagues,[e8] 2013	No	No	Yes	Yes	Unclear	No	Yes	Yes	Yes
Ngerncham and Colleagues,[e53] 2013	No	Unclear	Yes	Yes	Yes	No	Unclear	Yes	Unclear
Bai and Vaz,[e11] 2014	No	No	No	No	Unclear	Yes	No	Yes	Unclear
Chiang and Colleagues,[e22] 2014	No	No	No	No	No	No	Unclear	Yes	Unclear
González Jiménez and Colleagues,[e34] 2014	Unclear	No	Yes	Unclear	No	Yes	Unclear	No	Unclear
Haham and Colleagues,[e35] 2014	No	Unclear	No	Unclear	Unclear	Yes	Unclear	Yes	Unclear
Jamilian and Colleagues,[e38] 2014	Yes	Yes	No	Yes	Unclear	Yes	Yes	Yes	Unclear
Mohan and Colleagues,[e49] 2014	Unclear	No	No	Yes	Unclear	No	Unclear	Yes	Unclear
Riskin and Colleagues,[e62] 2014	No	Yes	Yes	No	Yes	No	No	Yes	No
Basalamah and Baroudi,[e15] 2016	Yes	Yes	Yes	Unclear	Yes	No	Unclear	Yes	Unclear
Lopes and Colleagues,[e44] 2016	No	No	No	Yes	No	Yes	Yes	Yes	Unclear
Fujinaga and Colleagues,[e31] 2017	Yes	Unclear	Unclear	Yes	Unclear	Yes	Unclear	Yes	Unclear
Kumar and Colleagues,[e40] 2017	No	Yes	Yes	Yes	Yes	Yes	No	Yes	Unclear
Lisonek and Colleagues,[e42] 2017	Yes	Yes	Yes	Yes	Yes	No	No	Yes	Yes
Puapornpong and Colleagues,[e57] 2017	Yes	Unclear	Yes	Yes	Unclear	Yes	Unclear	Yes	Unclear
Walsh and Colleagues,[e75] 2017	Yes	Yes	Yes	Yes	Yes	No	Unclear	Yes	Unclear
Yoon and Colleagues,[e76] 2017	No	Unclear	No	Yes	No	Yes	Unclear	Yes	Unclear
Avila and Colleagues,[e10] 2018	No	No	No	Yes	Unclear	Yes	Unclear	Yes	Yes
Brandão and Colleagues,[e17] 2018	No	No	Yes	Yes	Unclear	Yes	Yes	Yes	Yes
Chandler and Colleagues,[e20] 2018	No	No	No	Unclear	Unclear	No	No	Yes	No
Martinelli and Colleagues,[e46] 2018	Unclear	Unclear	No	Unclear	Unclear	Yes	Yes	Yes	Unclear
Perez-Aguirre and Colleagues,[e54] 2018	No	Unclear	Unclear	Unclear	Unclear	No	Yes	Yes	No
Walker and Colleagues,[e74] 2018	No	Unclear	No	Yes	No	No	No	Yes	Unclear
Bandaru and Colleagues,[e13] 2019	Unclear	Unclear	No	Yes	Unclear	No	Unclear	Yes	Yes
Campanha and Colleagues,[e18] 2019	No	No	Yes	Unclear	Unclear	Yes	Yes	Yes	Unclear
de Oliveira and Colleagues,[e24] 2019	No	Unclear	Yes	Yes	Unclear	No	Unclear	Yes	Unclear
Krittika and Don,[e41] 2019	No	No	No	No	No	No	Unclear	Yes	Unclear
Petousis-Harris and Colleagues,[e55] 2019	Yes	Yes	Unclear	Yes	Yes	No	No	Yes	Yes
Villa and Colleagues,[e72] 2019	No	Unclear	No	No	Unclear	Yes	Unclear	Yes	Yes
Araujo and Colleagues,[e9] 2020	No	No	Yes	Yes	Yes	Yes	Yes	Yes	Unclear

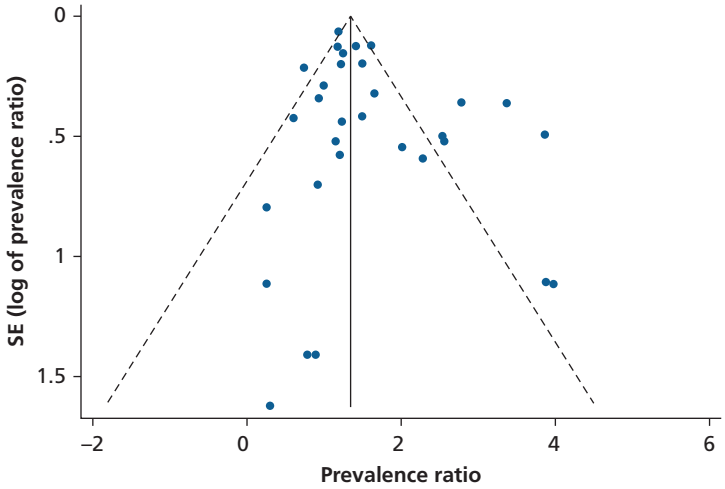
eFigure 1. (Continued)

Becerra-Culqui and Colleagues,[e16] 2020	Unclear	No	No	No	Unclear	No	No	Yes	Yes
Chang and Colleagues,[e21] 2020	Yes	Yes	Yes	Yes	Yes	No	Yes	Yes	Yes
Dutra and Colleagues,[e25] 2020	No	Unclear	No	Yes	No	Yes	No	Yes	Yes
Fraga and Colleagues,[e28] 2020	Unclear	Unclear	Yes	Yes	Unclear	Yes	Yes	Yes	Unclear
Potter and Colleagues,[e56] 2020	Unclear	Unclear	No	Unclear	Unclear	Yes	Unclear	Yes	Unclear
Razdan and Colleagues,[e59] 2020	No	Unclear	Unclear	Yes	Unclear	Yes	Yes	Yes	No
Zen and Colleagues,[e77] 2020	No	Unclear	Yes	Yes	No	Yes	Unclear	Yes	Unclear
Barberá-Perez and Colleagues,[e14] 2021	Unclear	Unclear	No	Unclear	Unclear	Yes	Unclear	Yes	Unclear
Maya-Enero and Colleagues,[e47] 2021	Yes	Yes	Yes	Yes	Yes	Yes	No	Yes	Unclear
Rech and Colleagues,[e60] 2021	Unclear	Unclear	Yes	Yes	Unclear	Yes	Unclear	Yes	Unclear
Shah and Colleagues,[e67] 2021	Yes	Unclear	No	Unclear	Unclear	No	Unclear	Yes	Unclear
Souza-Oliveira and Colleagues,[e68] 2021	No	Yes	Yes	Yes	No	Yes	Yes	Yes	Yes

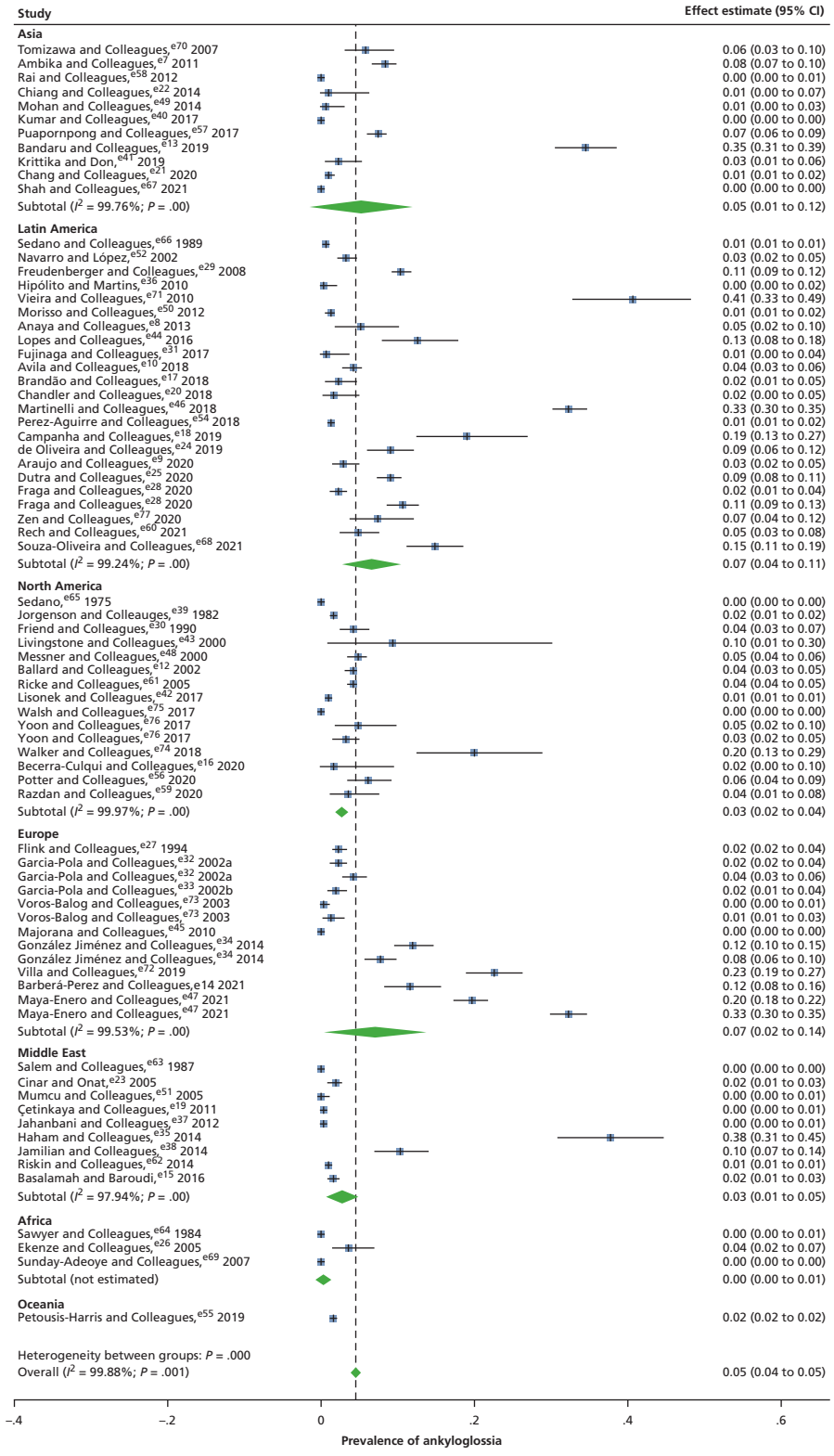
eFigure 1. (Continued)



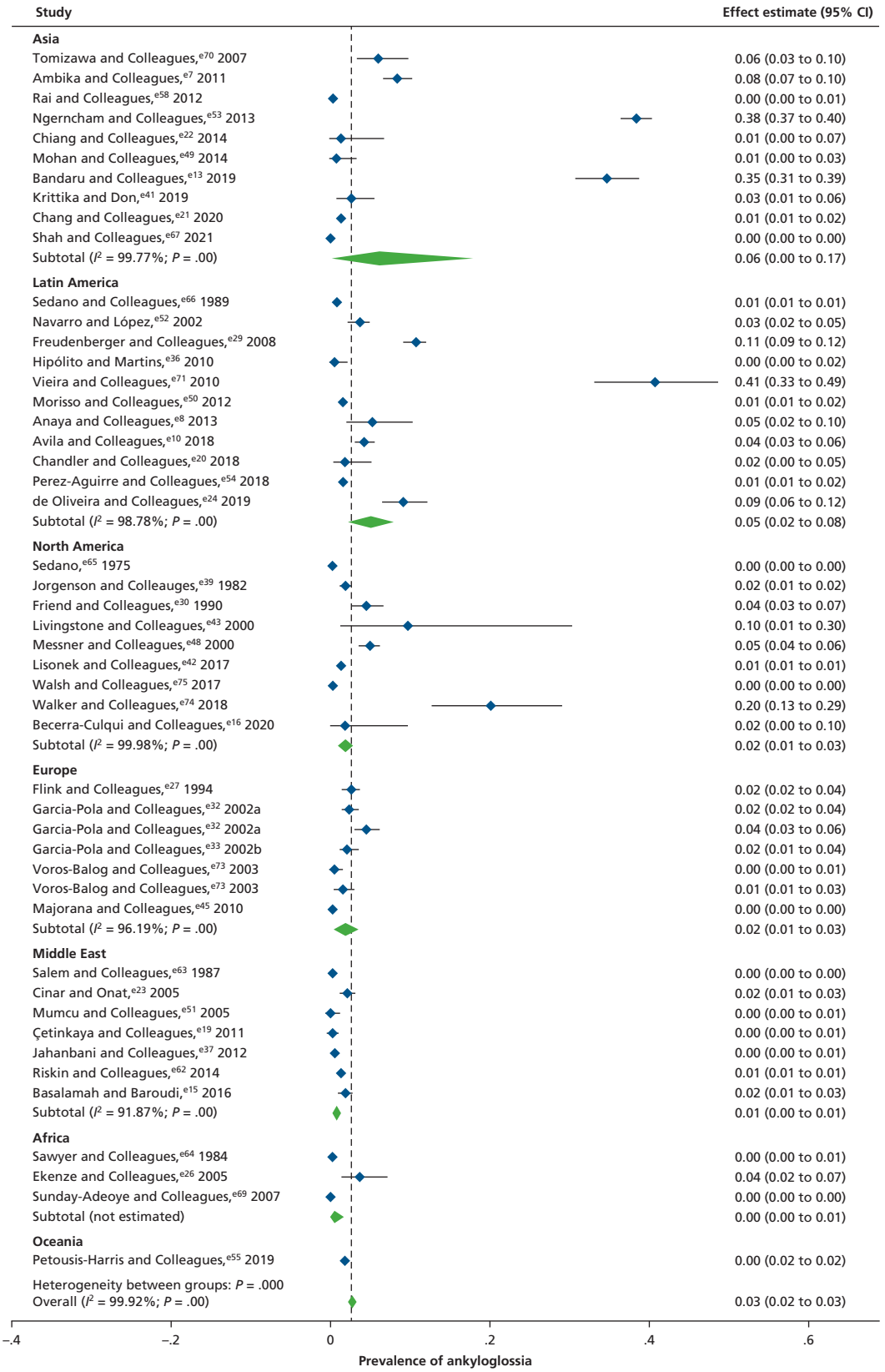
eFigure 2. Random-effect meta-analysis of overall prevalence of ankyloglossia. Prevalence: effect estimate. Overall: $\tau^2 = 0.01$; $\chi^2_{76} = 65{,}462.99$; $z = 31.73$; $P = .000$; $I^2 = 99.88\%$.



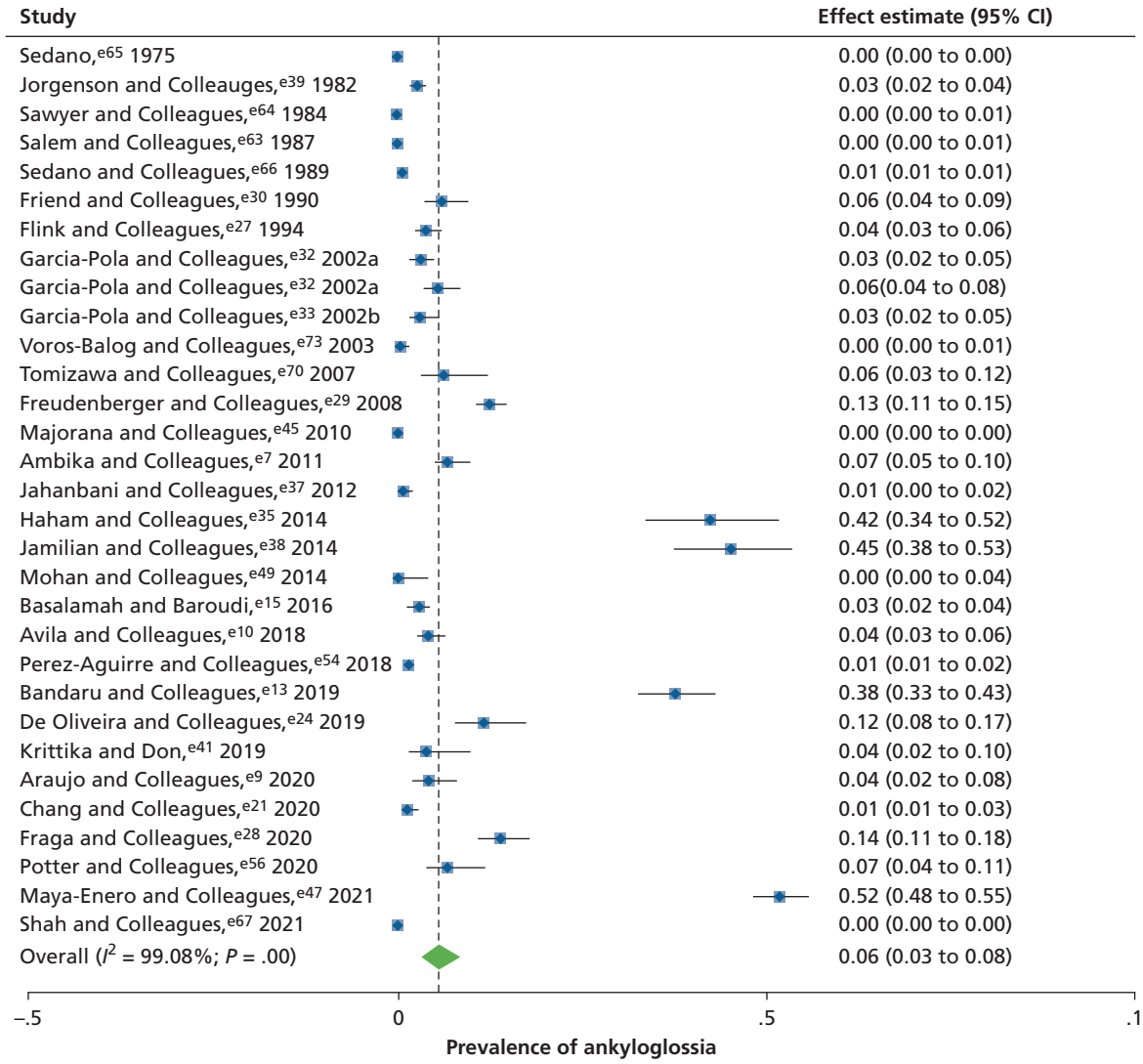
eFigure 3. Funnel plot of prevalence ratio and SE of log of prevalence ratio comparing boys and girls, with pseudo 95% CIs. Egger test for publication bias: $P = .062$.



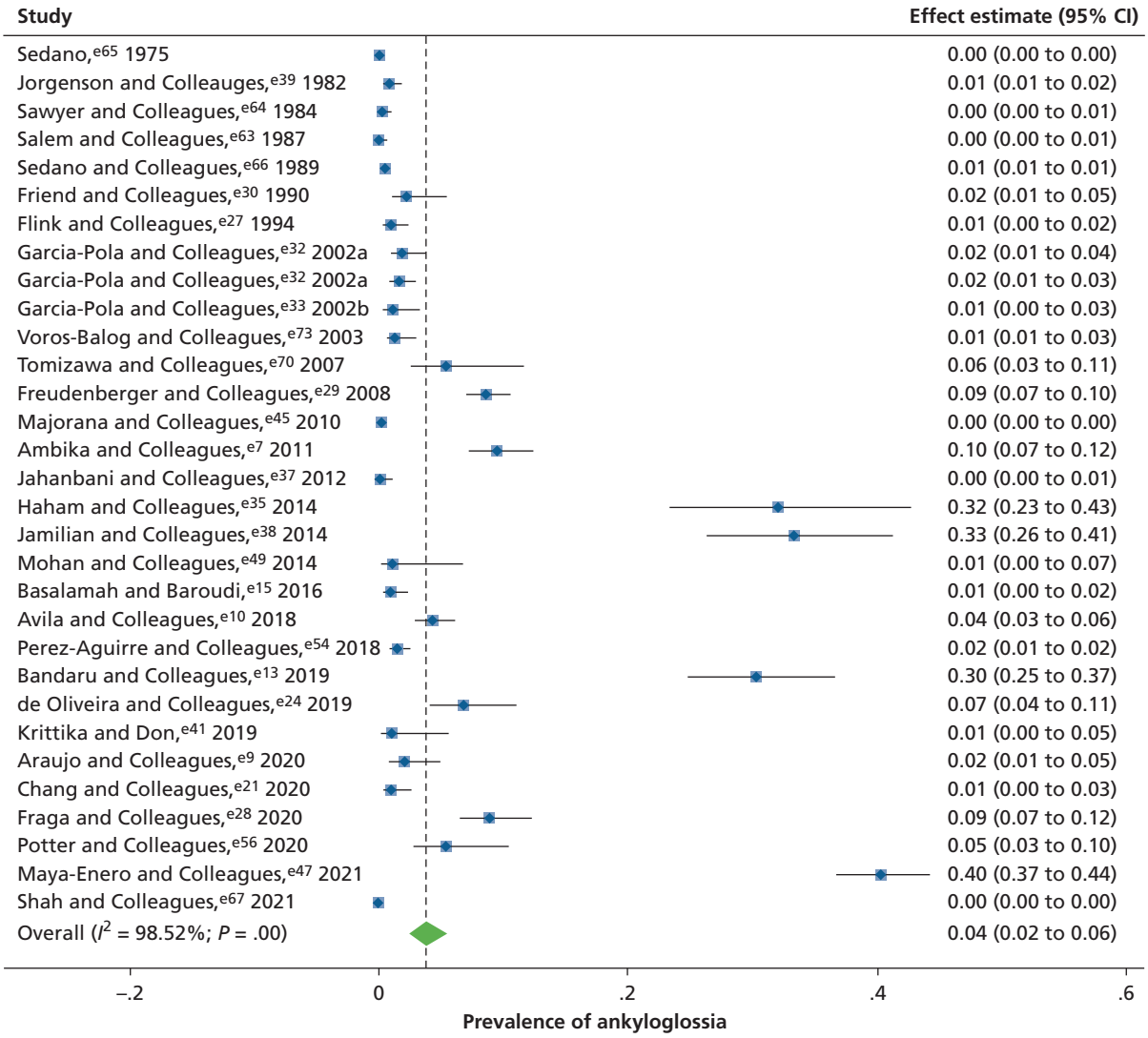
eFigure 4. Mixed-effects meta-analysis of ankyloglossia subgrouped by geographic region including all assessment tools. Prevalence: effect estimate. Asia: $\chi^2_{12} = 5{,}040.11$; $z = 3.42$, $P = .000$; $I^2 = 99.76\%$. Latin America: $\chi^2_{22} = 2{,}889.98$; $z = 6.74$, $P = .000$; $I^2 = 99.24\%$. North America: $\chi^2_{14} = 48{,}389.32$; $z = 10.45$, $P = .000$; $I^2 = 99.97\%$. Europe: $\chi^2_{12} = 2{,}562.76$; $z = 4.14$, $P = .000$; $I^2 = 99.53\%$. Middle East: $\chi^2_8 = 388.43$; $z = 5.01$, $P = .000$; $I^2 = 97.94\%$. Africa: χ^2_2 = not estimated; $z = 2.12$, $P = .000$; I^2 = not estimated. Oceania: χ^2_0 = not estimated; $z = 70.90$, $P = .000$; I^2 = not estimated. Overall: $\chi^2_{76} = 65{,}462.99$; $z = 31.73$, $P = .000$; $I^2 = 99.88\%$. Test for heterogeneity between subgroups: overall: $\tau^2 = 36.71$. $\chi^2_{(\text{degrees of freedom})} = \chi^2$ test for I^2 test.



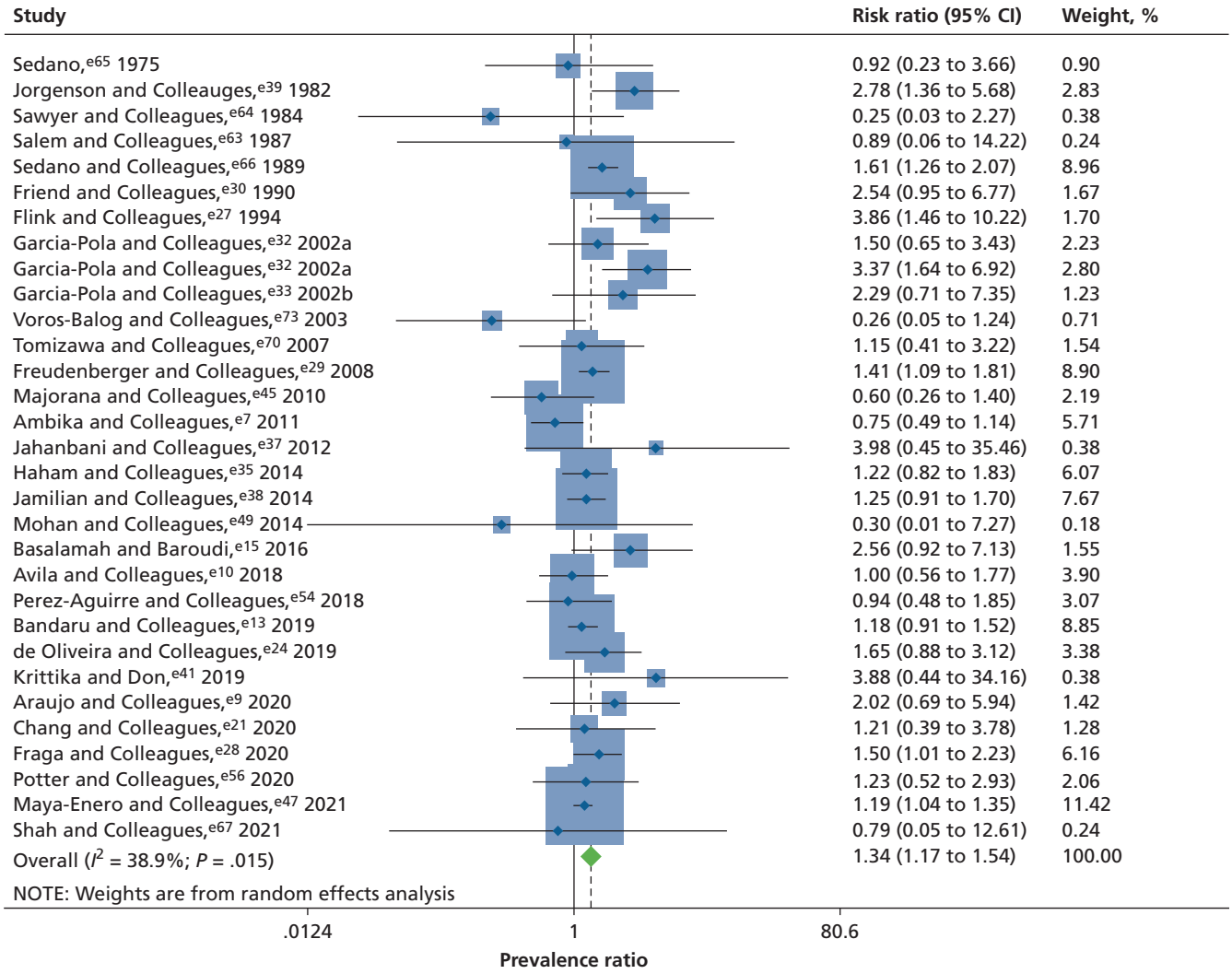
eFigure 5. Mixed-effects meta-analysis of ankyloglossia subgrouped by geographic region and sensitivity test excluding specific tools and maintaining only unspecific tools in the analysis. Prevalence: effect estimate. Asia: $\chi^2_9 = 3{,}999.30$; $z = 2.20$, $P = .000$; $I^2 = 99.77\%$. Latin America: $\chi^2_{10} = 819.59$; $z = 5.68$, $P = .000$; $I^2 = 98.78\%$. North America: $\chi^2_8 = 47{,}639.60$; $z = 6.18$, $P = .000$; $I^2 = 99.98\%$. Europe: $\chi^2_6 = 157.68$; $z = 4.01$, $P = .000$; $I^2 = 96.19\%$. Middle East: $\chi^2_6 = 73.83$; $z = 4.50$, $P = .000$; $I^2 = 91.87\%$. Africa: χ^2_2 = not estimated; $z = 2.12$, $P = .000$; I^2 = not estimated. Oceania: χ^2_0 = not estimated; $z = 70.90$, $P = .000$; I^2 = not estimated. Overall: $\chi^2_{47} = 57{,}003.96$; $z = 19.03$, $P = .000$; $I^2 = 99.92\%$. Test for heterogeneity between subgroups: overall: $\tau^2 = 25.45$. $\chi^2_{(\text{degrees of freedom})} = \chi^2$ for I^2 test.



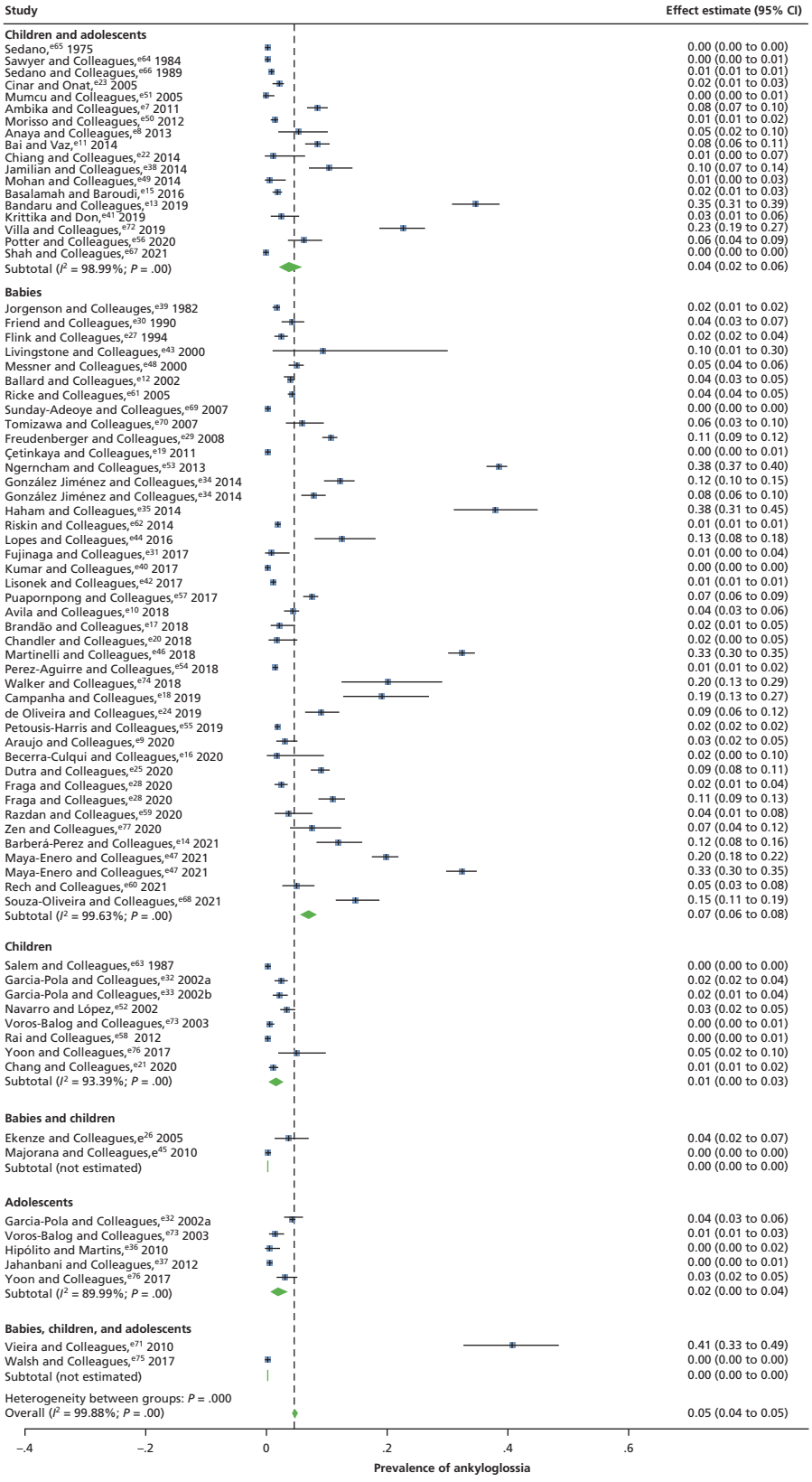
eFigure 6. Random-effect meta-analysis of overall crude prevalence of ankyloglossia among boys. Prevalence: effect estimate. Overall: $\tau^2 = 0.09$; $\chi^2_{30} = 3{,}244.42$; $z = 7.74$; $P = 0.00$; $I^2 = 99.08\%$.



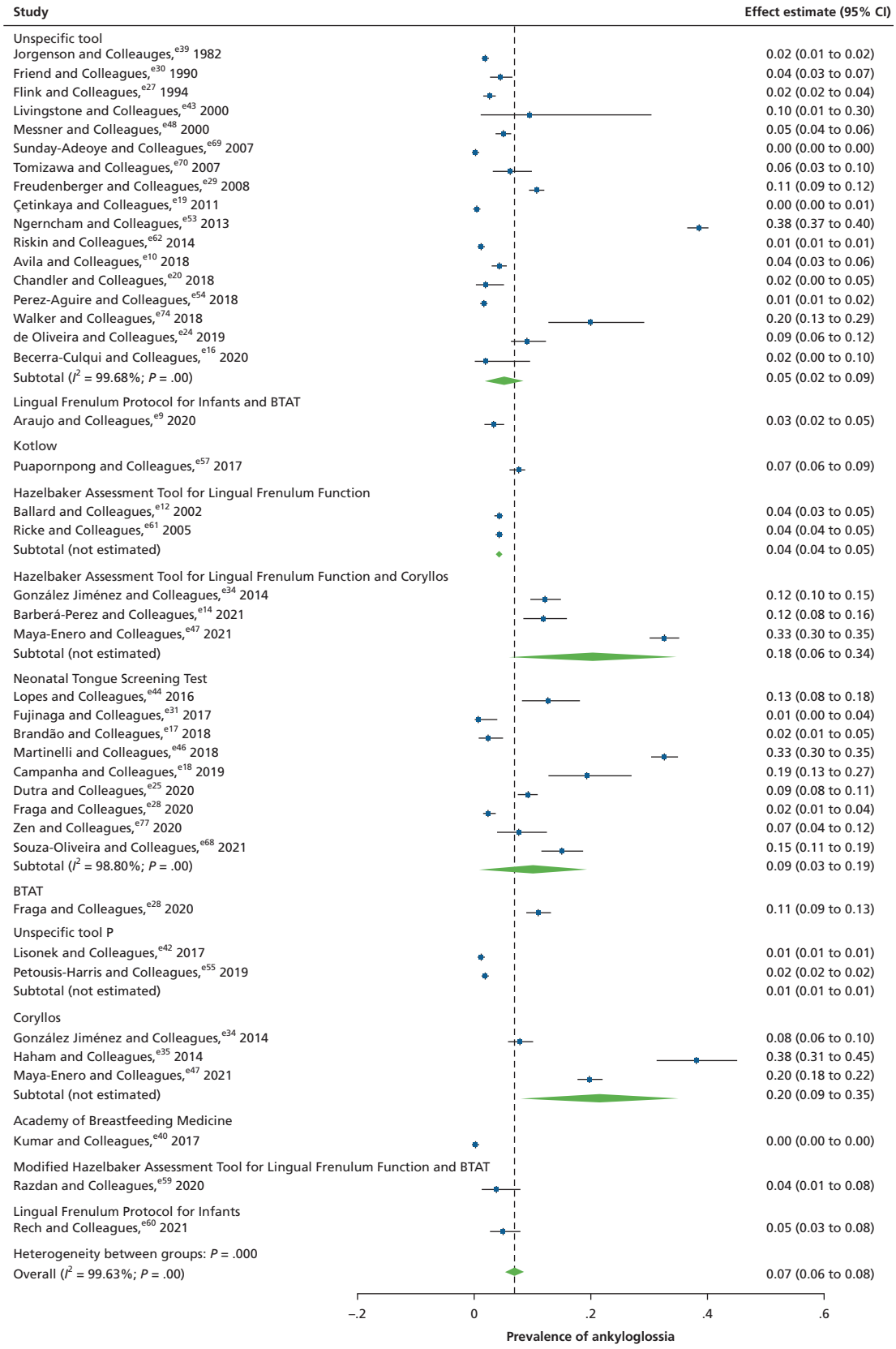
eFigure 7. Random-effect meta-analysis of overall crude prevalence of ankyloglossia among girls. Prevalence: effect estimate. Overall: $\tau^2 = 0.06$; $\chi^2_{30} = 2{,}030.73$; $z = 7.50$; $P = 0.00$; $I^2 = 98.52\%$.



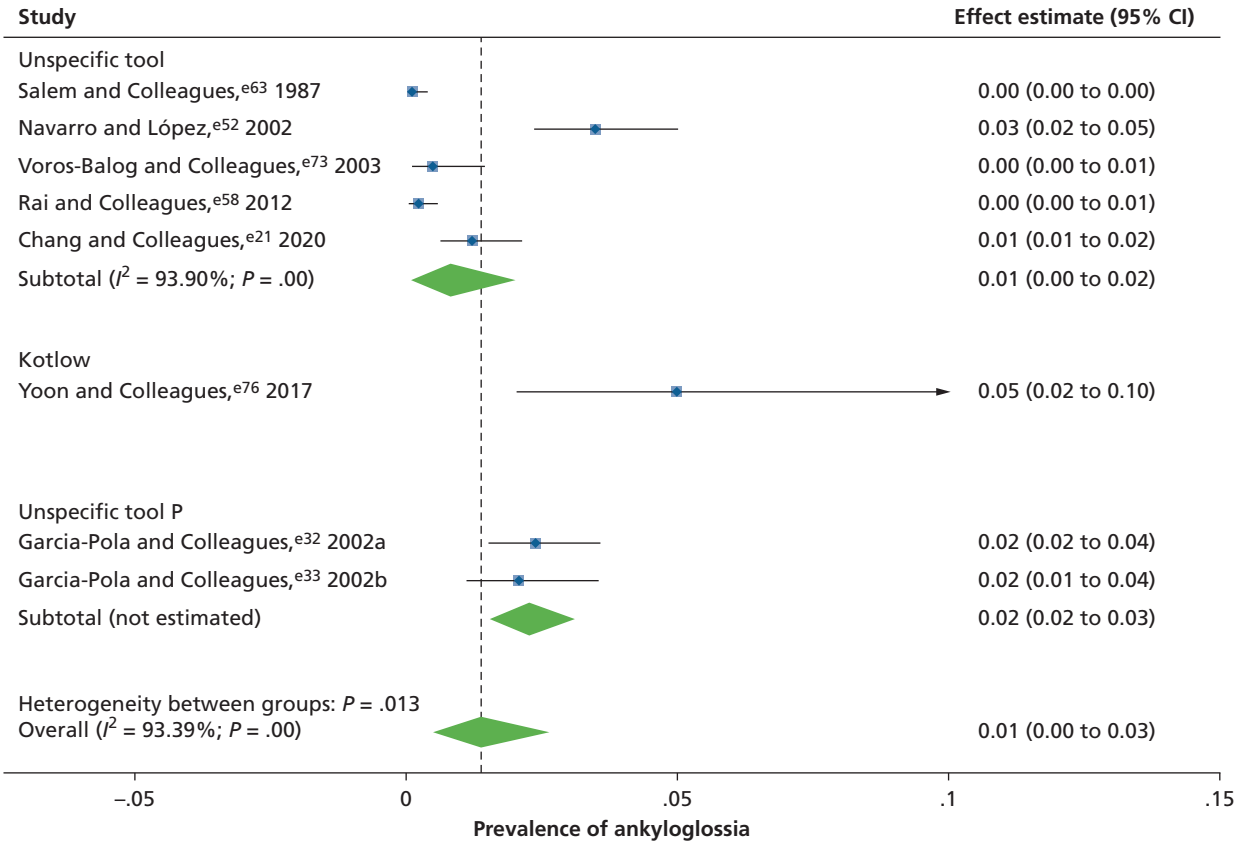
eFigure 8. Random-effect meta-analysis comparing the occurrence of ankyloglossia between boys and girls. Risk ratio = prevalence ratio. Overall: $\tau^2 = 0.0379$; $\chi^2_{30} = 49.11$; $z = 4.24$; $P = .000$; $I^2 = 38.9\%$.



eFigure 9. Mixed-effects meta-analysis of prevalence of ankyloglossia subgrouped by age. Prevalence: effect estimate. Infants: $\chi^2_{41} = 11{,}183.62$; $z = 16.85$, $P = .000$; $I^2 = 99.63\%$. Children: $\chi^2_7 = 105.91$; $z = 4.34$, $P = .000$; $I^2 = 93.39\%$. Adolescents: $\chi^2_4 = 39.97$; $z = 3.62$, $P = .000$; $I^2 = 89.99\%$. Infants and children: χ^2_1 = not estimated; $z = 5.32$, $P = .000$; I^2 = not estimated. Infants, children, and adolescents: $\chi^2_1 = 224.69$; $z = 224.69$, $P = .000$; $I^2 = 91.87\%$. Children and adolescents: $\chi^2_{17} = 1680.65$; $z = 6.56$, $P = .000$; $I^2 = 98.99\%$. Overall: $\chi^2_{76} = 65{,}462.99$; $z = 31.73$, $P = .000$; $I^2 = 99.88\%$. Test for heterogeneity between subgroups: overall: $\tau^2 = 301.54$. $\chi^2_{(\text{degrees of freedom})} = \chi^2$ test for I^2 test.

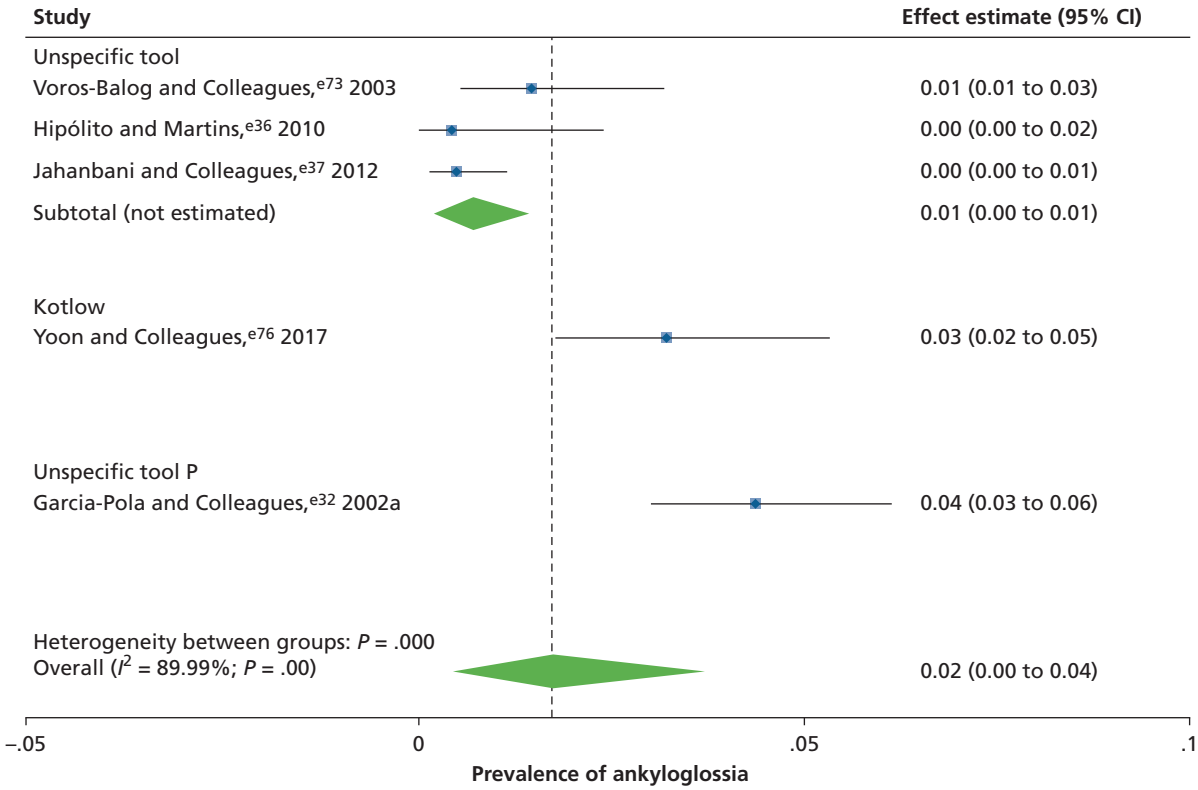


eFigure 10. Subgroup meta-analysis of prevalence of ankyloglossia among infants subgrouped by assessment tool. Prevalence: effect estimate. Unspecific tool: $\chi^2_{16} = 5{,}041.46$; $z = 4.76$, $P = .000$; $I^2 = 98.68\%$. Lingual Frenulum Protocol for Infants and BTAT (Bristol Tongue Assessment Tool): χ^2_0 = not estimated; $z = 6.65$, $P = .000$; $I^2 = 99.68\%$. Kotlow: χ^2_0 = not estimated; z =21.42; $P = 0.00$; I^2 = not estimated. HATLFF (Hazelbaker Assessment Tool for Lingual Frenulum Function): χ^2_1 = not estimated; $z = 32.04$, $P = .000$; I^2 = not estimated. HATLFF and Coryllos: χ^2_2 = not estimated; $z = 4.35$; $P = .000$; I^2 = not estimated. Neonatal Tongue Screening Test: $\chi^2_8 = 666.03$; $z = 4.22$, $P = .000$; $I^2 = 98.80\%$. BTAT: χ^2_0 = not estimated; $z = 19.02$, $P = .000$; I^2 = not estimated. Unspecific tool P: χ^2_1 = not estimated; $z = 403.89$, $P = .000$; I^2 = not estimated. Coryllos: χ^2_2 = not



eFigure 11. Subgroup meta-analysis of prevalence of ankyloglossia among children subgrouped by assessment tool. Prevalence: effect estimate. Unspecific tool: $\chi^2_4 = 65.62$; $z = 2.87$, $P = .000$; $I^2 = 93.90\%$. Kotlow: χ^2_0 = not estimated; $z = 4.51$; $P = 0.00$; I^2 = not estimated. Unspecific tool P: χ^2_1 = not estimated; $z = 10.76$, $P = .000$; I^2 = not estimated. Overall: $\chi^2_7 = 105.91$; $z = 4.34$, $P = 0.00$; $I^2 = 93.39\%$. Test for heterogeneity between subgroups: overall: $\tau^2 = 8.64$. $\chi^2_{(\text{degrees of freedom})} = \chi^2$ test for I^2 test.

eFigure 10 (continued). estimated; $z = 5.12$, $P = .000$; I^2 = not estimated. Academy of Breastfeeding Medicine: χ^2_0 = not estimated; $z = 15.07$, $P = .000$; I^2 = not estimated. Modified HATLFF and BTAT: χ^2_0 = not estimated; $z = 4.12$, $P = .000$; I^2 = not estimated. Lingual Frenulum Protocol for Infants: χ^2_0 = not estimated; $z = 6.94$, $P = .000$; I^2 = not estimated. Overall: $\chi^2_{41} = 11{,}183.6248$; $z = 16.85$, $P = .000$; $I^2 = 99.63\%$. Test for heterogeneity between subgroups: overall: $\tau^2 = 1{,}051.83$. $\chi^2_{(\text{degrees of freedom})} = \chi^2$ test for I^2 test.



eFigure 12. Subgroup meta-analysis of prevalence of ankyloglossia among infants subgrouped by assessment tool. Prevalence: effect estimate. Unspecific tool: χ^2_2 = not estimated; $z = 3.76$, $P = .000$; I^2 = not estimated. Kotlow: χ^2_0 = not estimated; $z = 6.65$; $P = 0.00$; I^2 = not estimated. Unspecific tool P: χ^2_0 = not estimated; $z = 10.48$, $P = .000$; I^2 = not estimated. Overall: $\chi^2_4 = 39.97$; $z = 3.62$, $P = 0.00$; $I^2 = 89.99\%$. Test for heterogeneity between subgroups: overall: $\tau^2 = 24.59$. $\chi^2_{(\text{degrees of freedom})} = \chi^2$ test for I^2 test.

eTable 1. Summary of finding table showing effect estimate (prevalence ratio) for ankyloglossia between boys and girls and certainty of evidence using Grading of Recommendations: Assessment, Development and Evaluation approach.*

		CERTAINTY ASSESSMENT					PATIENTS, n/N (%)		EFFECT	
STUDY, NO.	STUDY DESIGN	Risk of Bias	Inconsistency	Indirectness	Imprecision	Other Considerations	Boys	Girls	Relative (95% CI)	Certainty
31	Observational studies	Very serious†	Very serious‡	Not serious§	Not serious§	None§	1,317/ 45,233 (2.9)	886/ 40,746 (2.2)	Prevalence ratio, 1.34 (1.17 to 1.54)	Very low

* Relative measure presented as prevalence ratio. † The studies had very serious problems in several domains of Joanna Briggs Institute Critical Appraisal Tool,[14] such as representativeness of the target population, sample size, recruitment of the sample, coverage bias, reliability in the method used for diagnosis of ankyloglossia (unspecific tools), and lack of training and calibration of the examiners. ‡ The effect estimates of individual studies were not similar, lack of overlap of 95% CI and statistically significant I^2. § There were no problems due to indirectness, as the evidence generated from the included studies could be applied to the infants, children, and adolescents; boys; girls; ankyloglossia question. There were no problems due to imprecision, as the Optimal Information Size was fulfilled. The authors did not downgrade for publication bias, as the *P* value for the Egger test was 0.062, and all efforts were made to include all possible studies, including studies in Portuguese and Spanish. The authors did not find reasons to upgrade the certainty for dose response, magnitude of the effect, or effect of residual confounders.

eTable 2. Studies excluded from the systematic review and reasons for exclusion.

STUDY	REASON FOR EXCLUSION
Al-Maweri SA, Halboub ES, Al-Soneidar WA, Al-Sufyani GA. Oral lesions and dental status of autistic children in Yemen: a case–control study. *J Int Soc Prev Community Dent.* 2014;4(suppl 3):S199-S203.	The study did not report prevalence of ankyloglossia.
Amitai Y, Shental H, Atkins-Manelis L, Koren G, Zamir CS. Pre-conceptional folic acid supplementation: a possible cause for the increasing rates of ankyloglossia. *Med Hypotheses.* 2020;134:109508.	Was a case-control study.
Angulo-Núñez JJ, Rodriguez-Archilla A. Oral mucosal lesions in patients of Mérida, Venezuela. *Invest Clin.* 2015;56 (4):367-376.	The study did not report prevalence of ankyloglossia.
Aras MH, Göregen M, Güngörmüş M, Akgül HM. Comparison of diode laser and Er:YAG lasers in the treatment of ankyloglossia. *Photomed Laser Surg.* 2010;28(2):173-177.	Was not an observational study.
Ankur K, Bhasis JS, Baweja S. Tongue ties affecting breastfeeding in early term & full-term neonates. *Nutrition*.	Study was not found.
Ankur K, Joshi C, Prasad A, Bhasin JS, Baweja S. Prevalence of tongue-tie in infants weighing $\geq$1800 g at birth. *Indian J Pediatr.* 2021;88(10):1057.	The sample comprised babies with ankyloglossia followed prospectively for breast-feeding difficulties.
Ata N, Alataş N, Yılmaz E, Adam AB, Gezgin B. The relationship of ankyloglossia with gender in children and the ideal timing of surgery in ankyloglossia. *Ear Nose Throat J.* 2021;100(3):NP158-NP160.	The initial sample was suspected to have ankyloglossia.
Ataíde AP, Fonseca FP, Silva ARS, Júnior JJ, Lopes MA, Vargas PA. Distribution of oral and maxillofacial lesions in pediatric patients from a Brazilian southeastern population. *Int J Pediatr Otorhinolaryngol.* 2016;90:241-244.	The study did not report prevalence of ankyloglossia.
Awa HDM, Mvondo RMN, Nguefack S, Messanga CB, Ndombo POK. Les maladies rares et leurs manifestations cliniques orales dans deux formations hospitalières de Yaoundé. *Pan Afr Med J.* 2019; 32:195.	The sample had syndromes at the beginning of the study; the study did not report ankyloglossia.
Badawi N, Adelson P, Roberts C, Spence K, Laing S, Cass D. Neonatal surgery in New South Wales: what is performed where? *J Pediatr Surg.* 2003;38(7):1025-1031.	The study did not report prevalence of ankyloglossia.
Bajracharya D, Gupta S, Ojha B, Baral R. Prevalence of oral ucosal lesions in a tertiary care dental hospital of Kathmandu. *J Nepal Med Assoc.* 2017;56(207):362-366.	The study did not report prevalence of ankyloglossia. The initial sample was from biopsy cases.
Bamji MS, Sarma KR, Radhaiah G. Relationship between biochemical and clinical indices of B-vitamin deficiency: a study in rural school boys. *Br J Nutr.* 1979;41(3):431-441.	The study did not report prevalence of ankyloglossia.
Basra M, Patel N, Selbong UK. Tongue tie-do we need to treat? *Br J Oral Maxillofac Surg.* 2019;57(10):e25.	The initial sample had ankyloglossia at the beginning of the study.
Baxter R, Lashley A, Rendell NR. Tongue restriction questionnaire: a new screening tool to identify tongue-tied patients. *Compend Contin Educ Dent.* 2021;42(3):e1-e4.	The authors classified several degrees of ankyloglossia but did not determine the cutoff point for ankyloglossia.
Bellinger V, Solari D, Hogan M, Rodda K, Shadbolt B, Todd D. Tongue-tie division in the newborn: follow-up at 9 and 38 months. *Breastfeed Rev.* 2018;26(1):13-22.	The initial sample underwent frenectomy at the beginning of the study.
Boras VV, Rogulj AA, Alajbeg I, et al. The prevalence of oral mucosal lesions in Croatian children. *Paediatr Croat.* 2013;57(3):235-238.	The study did not report prevalence of ankyloglossia.
Bronoosh P, Kasraeian M, Ghazi Saeedi B. Oral abnormalities in an Iranian newborn population. *Pediatr. Dent J* 2014;24(1):8-11.	The study reported ankyloglossia together with bifid and geographic tongue, and it was not possible to extract data for ankyloglossia apart.
Brożek-Mądry E, Burska Z, Steć Z, Burghard M, Krzeski A. Short lingual frenulum and head-forward posture in children with the risk of obstructive sleep apnea. *Int J Pediatr. Otorhinolaryngol* 2021;144:110699.	Was a case-control study.
Buck LS, Frey H, Davis M, Robbins M, Spankovich C, Narisetty V. Characteristics and considerations for children with ankyloglossia undergoing frenulectomy for dysphagia and aspiration. *Am J Otolaryngol.* 2020;41(3):102393.	The initial sample underwent frenectomy at the beginning of the study.
Bundogji N, Zamora S, Brigger M, Jiang W. Modest benefit of frenotomy for infants with ankyloglossia and breastfeeding difficulties. *Int J Pediatr Otorhinolaryngol.* 2020;133:109985.	All the initial sample was suspected to have ankyloglossia or to have breast-feeding difficulties.
Bundy M, Rogerson N, Dalal A, et al. Lingual frenectomy in infants and its effect on breastfeeding. *Br J Oral Maxillofac Surg.* 2015;53(10):e75.	Was a review, editorial, commentary, or abstract.

eTable 2. Continued

STUDY	REASON FOR EXCLUSION
Caloway C, Hersh CJ, Baars R, Sally S, Diercks G, Hartnick CJ. Association of feeding evaluation with frenotomy rates in infants with breastfeeding difficulties. *JAMA Otolaryngol Head Neck Surg* 2019;145(9):817-822.	The initial sample was suspected to have ankyloglossia or to have breast-feeding difficulties.
Campanha SMA, Martinelli RLC, Palhares DB. Position of lips and tongue in rest in newborns with and without ankyloglossia. *CoDAS.* 2021;33(6):e20200069.	Same sample as Campanha SMA, Martinelli RLC, Palhares DB. Association between ankyloglossia and breastfeeding. *CoDAS.* 2019;31(1):e20170264.
Carvalho IF, Alencar PNB, Carvalho de Andrade MD, et al. Clinical and x-ray oral evaluation in patients with congenital Zika virus. *J Appl Oral Sci.* 2019;27:e20180276.	The sample had syndrome that affects craniofacial development.
Chandrasekaran PV, Palaniappan J, Rajendran A, Venugopal B, Gnanamoorthy P. Prevalence of ankyloglossia among children reporting with speech pathology to District Early Intervention Centre (DEIC): an observational study. *J Evolution Med Dent Sci.* 2020;9(11):860-863.	The sample had speech problems.
Chinnadurai S, Francis DO, Epstein RA, Morad A, Kohanim S, McPheeters M. Treatment of ankyloglossia for reasons other than breastfeeding: a systematic review. *Pediatrics.* 2015;135(6):e1467–e1474.	Was a systematic review.
Cawse-Lucas J, Waterman S, St Anna L. Clinical inquiry: does frenotomy help infants with tongue tie overcome breastfeeding difficulties? *J Fam Pract*. 2015;64(2):126-127.	Was an editorial, commentary, or abstract.
Chopra A, Lakhanpal M, Rao N, Gupta N, Vashisth S. Oral health in 4-6 years children with cleft lip/palate: a case control study. *N Am J Med Sci.* 2014;6(6):266-269.	Was a case-control study.
Cruz PV, Bendo CB, Bouzada MCF, Machado MGP, Martins CC. Oral mucosal lesions in newborns: relationship with prematurity, low birth weight, and associated factors. *J Clin Neonatol.* 2021;10:170-177.	Same sample as Souza-Oliveira AC, Cruz PV, Bendo CB, Batista WC, Bouzada MCF, Martins CC. Does ankyloglossia interfere with breastfeeding in newborns? A cross-sectional study. *J Clin Transl Res*. 2021;7(2):263-269.
Dabić DT, Kansky A, Boras VV. Prevalence of oral mucosal lesions in Slovenia. *Res J Pharm Biol Chem Sci.* 2015;6(5):1154-1157.	Was a study with adults and not related to ankyloglossia.
Daggumati S, Cohn JE, Brennan MJ, Evarts M, McKinnon BJ, Terk AR. Caregiver perception of speech quality in patients with ankyloglossia: comparison between surgery and non-treatment. *Int J Pediatr Otorhinolaryngol.* 2019;119:70-74.	The study included patients with ankyloglossia who underwent or did not undergo intervention.
da Silva Dalben G, Richieri-Costa A, de Assis Taveira L. Tooth abnormalities and soft tissue alterations in patients with G/BBB syndrome. *Oral Dis.* 2008;14(8):747-753.	Study had congenital syndrome that affects craniofacial development.
Dave J, Sinha V, Barot D, Modi N, Gurnani D, Patel T. Speech disorders encountered in routine ENT practice and the role of speech therapy in its effective management. *Indian J Otol.* 2013;19(4):169-172.	The initial sample had speech disorders.
DeGiovanni JC, Ma AC, Rosi-Schumacher MR, et al. Feeding issues in infants referred for frenotomy. *Otolaryngol Head Neck Surg*. 2021;165(1 suppl):P289-P317. https://doi.org/10.1177/01945998211030910g	The sample was referred to the hospital for frenotomy procedure.
de Oliveira LJC, Torriani DD, Correa MB, et al. Oral mucosal lesions' impact on oral health-related quality of life in preschool children. *Commun Dent Oral Epidemiol.* 2015;43(6):578-585.	The study did not report prevalence of ankyloglossia.
Díaz-Pizán ME, Lagravère MO, Villena R. Midline diastema and frenum morphology in the primary dentition. *J Dent Child.* 2006;73(1):11-14.	The study did not report prevalence of ankyloglossia.
Diercks GR, Hersh CJ, Baars R, Sally S, Caloway C, Hartnick CJ. Factors associated with frenotomy after a multidisciplinary assessment of infants with breastfeeding difficulties. *Int J Pediatr Otorhinolaryngol.* 2020;138:110212.	The sample was referred to the hospital for frenotomy procedures at the beginning of the study.
Dixon B, Gray J, Elliot N, Shand B, Lynn A. A multifaceted programme to reduce the rate of tongue-tie release surgery in newborn infants: observational study. *Int J Pediatr Otorhinolaryngol.* 2018;113:156-163.	The initial sample had ankyloglossia and underwent frenectomy.
Dollberg S, Manor Y, Makai E, Eyal Botzer. Evaluation of speech intelligibility in children with tongue-tie. *Acta Paediatr.* 2011;100(9):e125-e127.	The study did not report prevalence of ankyloglossia.
Dollberg S, Marom R, Botzer E. Lingual frenotomy for breastfeeding difficulties: a prospective follow-up study. *Breastfeed Med.* 2014;9(6):286-289.	The initial sample had ankyloglossia and underwent frenectomy.
Dogenski LC, Farina AP, Linden MSS, Trentin MS, Miyagaki DC, De Carli JP. Oral lesions found in a dental school in southern Brazil. *J Contemp Dent Pract.* 2018;19(9):1037-1041.	Study with adults not related to ankyloglossia

eTable 2. Continued

STUDY	REASON FOR EXCLUSION
Dos Santos PJB, Bessa CFN, De Aguiar MCF, Do Carmo MAV. Cross-sectional study of oral mucosal conditions among a central Amazonian Indian community, Brazil. *J Oral Pathol Med.* 2004;33(1):7-12.	The study did not report prevalence of ankyloglossia.
Douglas P. Preventing overdiagnosis in the first months of life. *BMJ Evid Based Med.* 2019;24(2):A1-A5.	Was a review, editorial, commentary, or abstract.
Du RY, Mcgrath C, Yiu CKY, King NM. Oral health in preschool children with cerebral palsy: a case-control community-based study. *Int J Paediatr Dent.* 2010;20(5):330-335.	The sample had cerebral palsy.
Du RY, Yiu CK, King NM, Wong VC, McGrath CP. Oral health among preschool children with autism spectrum disorders: a case-control study. *Autism.* 2014;19(6):746-751.	Was a case-control study.
Edmunds J, Hazelbaker A, Murphy JG, Philipp BL. Roundtable discussion: tongue-tie. *J Hum Lact.* 2012;28(1):14-17.	Was a review, editorial, commentary or abstract.
Edmunds JE, Fulbrook P, Miles S. Understanding the experiences of mothers who are breastfeeding an infant with tongue-tie. *J Hum Lact.* 2013;29(2):190-195.	The initial sample had ankyloglossia; it was not a prevalence study.
El-Bassyouni HT, Hassan N, Mahfouz I, Abd-Elnaby AE, Mostafa MI, Tosson AMS. Early detection and management of Prader-Willi syndrome in Egyptian patients. *J Pediatr Genet.* 2019;8(4):177-186.	The sample had Prader-Willi syndrome.
Ellehauge E, Jensen JS, Grønhøj C, Hjuler T. Trends of ankyloglossia and lingual frenotomy in hospital settings among children in Denmark. *Dan Med J.* 2020;67(5):A01200051.	The initial sample had ankyloglossia and need of frenectomy.
Erthal A, Lourenço SV, Nico MMS. Oral mucosal diseases in children: casuistics from the Department of Dermatology - University of São Paulo - Brazil. *An Bras Dermatol.* 2016;91(6):849-851.	The study did not report prevalence of ankyloglossia.
Espinosa-Zapata M, Loza-Hernández G, Mondragón-Ballesteros R. Prevalence of buccal mucosa lesions in pediatric patients. Preliminary report. *Cir Cir.* 2006;74(3):153-157.	The study did not report prevalence of ankyloglossia.
Feng J, Zhou Z, Shen X, Wang Y, Shi L, Wang Y. Prevalence and distribution of oral mucosal lesions: a cross-sectional study in Shanghai, China. *J Oral Pathol Med.* 2014;44(7):490-494.	The study reported oral lesion in adults.
Ferres-Amat E, Pastor-Vera T, Ferres-Amat E, Mareque-Bueno J, Prats-Armengol J, Ferrés-Padró E. Multidisciplinary management of ankyloglossia in childhood: treatment of 101 cases—a protocol. *Med Oral Patol Oral Cir Bucal.* 2016;21(1):e39-e47.	The initial sample had ankyloglossia.
Ferrés-Amat E, Pastor-Vera T, Rodriguez-Alessi P, Ferrés-Amat E, Mareque-Bueno J, Ferrés-Padró E. The prevalence of ankyloglossia in 302 newborns with breastfeeding problems and sucking difficulties in Barcelona: a descriptive study. *Eur J Paediatr Dent.* 2017;18(4):319-325.	The initial sample had ankyloglossia and breast-feeding difficulties.
Fletcher SG, Meldrum JR. Lingual function and relative length of the lingual frenulum. *J Speech Hear Res.* 1968;11(2):382-390.	Was not a prevalence study.
Fonteles CSR, Ribeiro EM, Santos MSA, Leite RFP, Assunção GS, Monteiro AJ. Lingual frenulum phenotypes in Brazilian infants with congenital Zika syndrome. *Cleft Palate Craniofac J.* 2018:55(10):1391-1398.	Had congenital syndromes that affect craniofacial development.
Fraga MDRBA, Barreto KA, Lira TCB, De Menezes VA. Diagnosis of ankyloglossia in newborns: is there any difference related to the screening method? *CoDAS.* 2021;33(1):e20190209.	Same sample as Fraga MDRBA, Barreto KA, Lira TCB, Menezes VA. Is the occurrence of ankyloglossia in newborns associated with breastfeeding difficulties? *Breastfeed Med.* 2020;15(2):96-102.
França ECL, Albuquerque LCA, Martinelli RLC, Gonçalves IMF, Souza CB, Barbosa MA. Surface electromyographic analysis of the suprahyoid muscles in infants based on lingual frenulum attachment during breastfeeding. *Int J Environ Res Public Health.* 2020;17(3):859.	Was not a prevalence study.
García AT. Prevalencia de lesiones bucales en tejido blando encontradas en la Clínica de Estomatología de la Facultad de Odontología de la Universidad de los Andes. Periodo 2015-2018. *ADM.* 2020; 77(1):11-16.	The sample was adults.
Ghaheri BA, Cole M, Fausel SC, Chuop M, Mace JC. Breastfeeding improvement following tongue-tie and lip-tie release: a prospective cohort study. *Laryngoscope.* 2016;127(5):1217-1223.	The initial sample had ankyloglossia.

eTable 2. Continued

STUDY	REASON FOR EXCLUSION
Ghaheri BA, Cole M, Mace JC. Revision lingual frenotomy improves patient-reported breastfeeding outcomes: a prospective cohort study. *J Hum Lact.* 2018;34(3):566-574.	The initial sample underwent frenectomy.
Gheno JN, Martins MAT, Munerato MC, Hugo FN, Filho MS, Weissheimer C. Oral mucosal lesions and their association with sociodemographic, behavioral, and health status factors. *Braz Oral Res.* 2015;29:1-6.	The study was about cancer screening.
Glynn RW, Colreavy M, Rowley H, Gendy S. Division of tongue tie: review of practice through a tertiary paediatric otorhinolaryngology service. *Int J Pediatr Otorhinolaryngol.* 2012;76(10):1434-1436.	The initial sample had frenectomy.
Gopal RM. An observational study of tongue tie release using bipolar diathermy at Vellore district in Tamilnadu. *J Evol Med Dent Sci.* 2018;7(52):5335-5339.	Was not a prevalence study.
Guzmán LR, Quiroz TC, Bailón NR, Felicesd LR, Costae CP, Guiuf CG. Herencia de la anquiloglosia: de tal palo, tal astilla. *Rev Pediatr Aten Primaria.* 2019;21(83): e129-35.	Was a case series.
Hale M, Mills N, Edmonds L, Dawes P, Dickson N, Barker D. Complications following frenotomy for ankyloglossia: a 24-month prospective New Zealand Paediatric Surveillance Unit study. *J Paediatr Child Health.* 2020;56(4):557-562.	The initial sample had frenectomy.
Hall DMB, Renfrew MJ. Tongue tie. *Arch Dis Child.* 2005;90(12):1211-1215.	Was a review, editorial, commentay, or abstract.
Han SH, Kim MC, Choi YS, Dawes P, Dickson N, Barker D. A study on the genetic inheritance of ankyloglossia based on pedigree analysis. *Arch Plast Surg.* 2012;39(4):329-332.	The initial sample underwent frenectomy.
Haug AC, Markestad T, Tiora E, Moster D. Tight tongue band in newborns. *Tidsskrift For Den Norske Laegeforening.* Published online August 27, 2021. doi:10.4045/tidsskr.21.0515	Was a commentary.
Hanna R, Parker S. The advantages of carbon dioxide laser applications in paediatric oral surgery: a prospective cohort study. *Lasers Med Sci.* 2016;31(8):1527-1536.	Was not an observational study.
Harris EF, Friend GW, Tolley EA. Enhanced prevalence of ankyloglossia with maternal cocaine use. *Cleft Palate-Craniofac J.* 1992;29(1):72-76.	Was a case-control study.
Harrison-Woolrych M, Paterson H, Tan M. Exposure to the smoking cessation medicine varenicline during pregnancy: a prospective nationwide cohort study. *Pharmacoepidemiol Drug Saf.* 2013;22(10):1086-1092.	The initial sample was pregnant women.
Hasan A, Cousin G. Ankyloglossia (tongue-tie). *Afr J Paediatr Surg.* 2015;12(1):101.	Was a case report.
Hazelbaker AK. Newborn tongue-tie and breast-feeding. *J Am Board Fam Pract.* 2005;18(4):326-326.	Was a review, editorial, commentary, or abstract.
Hurst N, Tucker K. Diagnosing ankyloglossia. *J Hum Lact.* 2013;29(3):423-423.	Was a review, editorial, commentary, or abstract.
Illing S, Minnee M, Wheeler J, Illing L. The value of frenotomy for ankyloglossia from a parental perspective. *NZ Med J.* 2019;132(1500):70-81.	The initial sample had ankyloglossia and frenectomy.
Janiszewska-Olszowska J, Gawrych E, Dydyk A, Studniak E, Biaduń-Popławska A, Zajączek S. Oro-palatal dysplasia Bettex–Graf: clinical findings, genetic background, treatment. *J Cranio-Maxillofac Surg.* 2013;41(1):e29-e32.	The sample had a syndrome.
Karabulut R, Sönmez K, Türkyilmaz Z, Demiroğullari B, Ozen IO, Bağbanci B. Ankyloglossia and effects on breast-feeding, speech problems and mechanical/social issues in children. *B-ENT.* 2008;4(2):81-85.	The initial sample had ankyloglossia.
Khoo AKK, Dabbas N, Sudhakaran N, Patel S. Nipple pain at presentation predicts success of tongue-tie division for breastfeeding problems. *Eur J Pediatr Surg.* 2009;19(6):370-373.	The initial sample had ankyloglossia and frenectomy.
Kleinman DV, Swango PA, Pindborg JJ. Epidemiology of oral mucosal lesions in United States schoolchildren: 1986-87. *Community Dent Oral Epidemiol.* 1994;22(4):243–253.	The study did not report prevalence of ankyloglossia.
Klockars T, Pitkäranta A. Pediatric tongue-tie division: indications, techniques and patient satisfaction. *Int J Pediatr Otorhinolaryngol.* 2009;73(10):1399-1401.	The initial sample had ankyloglossia and frenectomy.
Köse O, Güven G, Özmen İ, Altun C. The oral mucosal lesions in pre-school and school age Turkish children. *J Eur Academy Dermatol Venereol.* 2011;27(1):e136-e137.	The study did not report prevalence of ankyloglossia.

eTable 2. Continued

STUDY	REASON FOR EXCLUSION
Legbo JN, Opara WE. Day-care plastic surgery in Nigeria: coping with limited resources. *Ann Afri Med.* 2005;4(1):14-18.	It was not a prevalence study of ankyloglossia but a study of surgical procedures.
LeTran V, Osterbauer B, Buen F, Yalamanchili R, Gomez G. Ankyloglossia: last three-years of outpatient care at a tertiary referral center. *Int J Pediatr Otorhinolaryngol.* 2019;126:109599.	The sample had ankyloglossia at the beginning of the study.
Lima ALX, Dutra MRP. Influence of frenotomy on breastfeeding in newborns with ankyloglossia. *CoDAs.* 2021;33(1):e20190026.	The initial sample had ankyloglossia at the beginning of the study.
Linares-Vieyra C, del Carmen Meza-Sánchez J, González-Guevara MB, Murrieta-Pruneda JF, Salgado-Rodríguez SJ, Morales-Jaimes R. Lesiones de mucosa bucal: factores asociados en población infantil. *Rev Med Inst Mex Seguro Soc.* 2013;51(3):320-325.	The study did not report prevalence of ankyloglossia.
Lu HX, Tao DY, Lo ECM, Li R, Wang X, Tai BJ. The 4th National Oral Health Survey in the mainland of China: background and methodology. *Chin J Dent Res.* 2018;21(3):161-165.	The study did not report prevalence of ankyloglossia.
Machet L, Hüttenberger B, Georgesco G, Doré C, Jamet F, Bonnin-Goga B. Absence of inferior labial and lingual frenula in Ehlers-Danlos syndrome. *Am J Clinic Dermatol.* 2010;11(4):269-273.	The sample had a syndrome.
Marra PM, Itro A. Surgical management of frenula: laser therapy compared with Z-frenuloplasty technique. *Pesqui Bras Odontopediatria Clin Integr.* 2020;20:e0027.	Was not an observational study.
Martinelli RLDC, Marchesan IQ, Berretin-Felix G. Compensatory strategies for the alveolar flap production in the presence of ankyloglossia. *CEFAC.* 2019;21(3):e10419.	Was not a prevalence study.
Martinelli RLDC, Marchesan IQ, Berretin-Felix G. Tongue position for lingual frenulum assessment. *CEFAC.* 2020;22(1):e0120.	The sample had ankyloglossia at the beginning of the study.
McBride C. Tongue-tie. *J Paediatr Child Health.* 2005;41:242(5-6):242.	Was a review, editorial, commentary, or abstract.
Mercer NSG. Division of tongue tie: an assault on a baby. *BMJ.* 2021;372:n11.	Was an editorial.
Miller AS, Miller JE. Is tongue tie really the problem? Incidence of ankyloglossia in an infant population presented with suboptimal feeding: a cross-sectional survey. *J Clin Chiropr Pediatr.* 2017;16(1):1350-1354.	The initial sample had breast-feeding difficulties.
Molania T, Nahvi A, Delrobaee M, Salehi M. Frequency of oral mucosal lesions and awareness of these lesions in patients attending oral and maxillofacial clinic in Sari Dental School, Iran. *J Mazandaran Univ Med Sci.* 2017;26(146):80-87.	The language was Persian.
Muldoon K, Gallagher L, McGuinness D, Smith V. Effect of frenotomy on breastfeeding variables in infants with ankyloglossia (tongue-tie): a prospective before and after cohort study. *BMC Pregnancy Childbirth.* 2017;17(1):373.	The sample underwent frenectomy.
Naimer SA, Biton A, Vardy D, Zvulunov A. Office treatment of congenital ankyloglossia. *Med Sci Monit.* 2003;9(10):CR432-CR435.	Was an interventional study.
Neves M, do Amaral Giordani JM, Ferla AA, Hugo FN. Primary care dentistry in Brazil: from prevention to comprehensive care. *J Ambul Care Manage.* 2017;40(suppl 2):S35.	The study did not report prevalence of ankyloglossia.
Nolan C, Corry P, O'Rourke C, Fenton J. To examine what percentage of patients referred to centre for tongue tie release were referred for breastfeeding difficulties and how many of them stopped breastfeeding as a result. *Ir J Med Sci.* 2015;184:S165-S166.	Was a review, editorial, commentary, or abstract.
O'Callahan C, Macary S, Clemente S. The effects of office-based frenotomy for anterior and posterior ankyloglossia on breastfeeding. *Int J Pediatr Otorhinolaryngol.* 2013;77(5):827-832.	The sample had ankyloglossia and underwent frenectomy.
O'Leary CM, Slack-Smith LM. Dental hospital admissions in the children of mothers with an alcohol-related diagnosis: a population-based, data-linkage study. *J Pediatr.* 2013;163(2):515-520.	The study did not report prevalence of ankyloglossia.
O'Shea JE, Foster JP, O'Donnell CP, et al. Frenotomy for tongue-tie in newborn infants. Cochrane Database Syst Rev. 2017;3:CD011065.	Was a systematic review.
Padilla CD, Cutiongco EM, Sia JM. Birth defects ascertainment in the Philippines. *Southeast Asian J Trop Med Public Health.* 2003;34(suppl 3):239-243.	The study did not report prevalence of ankyloglossia.
Parlak A, Koybasi S, Yavuz T, et al. Prevalence of oral lesions in 13- to 16-year-old students in Duzce, Turkey. *Oral Dis.* 2006;12(6):553-558.	Ankyloglossia was reported together with other defects, and it was not possible to extract data.

eTable 2. Continued

STUDY	REASON FOR EXCLUSION
Pauws E, Moore GE, Stanier P. A functional haplotype variant in the *TBX22* promoter is associated with cleft palate and ankyloglossia. *J Med Genet.* 2009;46(8):555-561.	Was not a prevalence study.
Pereira NM, Maresh A. Trends in outpatient intervention for pediatric ankyloglossia. *Int J Pediatr Otorhinolaryngol.* 2020;138:110386.	The sample was referred for frenectomy.
Periyasamy Y, Ravindran V, Subhashini VC. Oral mucosal lesions in children with and without cleft lip and palate: a case control study. *Int J Res Pharm Sci.* 2020;11:1233-1238.	Was a case-control study.
Pinsak GF. A radiographic and model analysis of patients manifesting partial congenital ankyloglossia. *Am J Orthod.* 1977;72(3):331-332.	Was a review, editorial, commentary, or abstract.
Praborini A, Setiani A, Munandar A, Wulandari RA. A holistic supplementation regimen for tongue-tied babies with slow weight gain and failure to thrive. *Clin Lact.* 2018;9(2):78-87.	Was an interventional study with babies with ankyloglossia.
Pransky SM, Lago D, Hong P. Breastfeeding difficulties and oral cavity anomalies: the influence of posterior ankyloglossia and upper-lip ties. *Int J Pediatr Otorhinolaryngol.* 2015;79(10):1714-1717.	Hospital reference in treating patients with ankyloglossia and breast-feeding difficulties; the sample was suspected of having ankyloglossia at the beginning of the study.
Prasertsom P, Kaewkamnerdpong I, Krisdapong S. Condition-specific oral health impacts in Thai children and adolescents: findings from the National Oral Health–Related Quality of Life Survey. *Asia Pac J Public Health.* 2020;32(1):49-56.	The study did not report prevalence of ankyloglossia.
Pola M, Garcia MG, Martin JMG, Gallas M, Leston JS. A study of pathology associated with short lingual frenum. *J Dent Child.* 2002;69(1):59-62.	The sample had ankyloglossia and was suspected of having speech problems at the beginning of the study.
Puapornpong P, Raungrongmorakot K, Mahasitthiwat V, Ketsuwan S. Comparisons of the latching on between newborns with tongue-tie and normal newborns. *J Med Assoc Thai.* 2014;97(3):255-259.	Same sample as Puapornpong P, Paritakul P, Suksamarnwong M, Srisuwan S, Ketsuwan S. Nipple pain incidence, the predisposing factors, the recovery period after care management, and the exclusive breastfeeding outcome. *Breastfeed Med.* 2017;12(3),169-173.
Qiao Y, Shi H, Wang H, Wang M, Chen F. Oral health status of Chinese children with autism spectrum disorders. *Front Psychiatry.* 2020;11:398.	The study did not report prevalence of ankyloglossia.
Ruffoli R, Giambelluca M, Scavuzzo M, et al. Ankyloglossia: a morphofunctional investigation in children. *Oral Dis.* 2005;11(3):170-174.	Sample with ankyloglossia referred to treatment.
Salt H, Claessen M, Johnston T, Smart S. Speech production in young children with tongue-tie. *Int J Ped Otorhinolaryngol.* 2020; 134:110035.	Was not a prevalence study.
Sandberg-Wollheim M, Neudorfer O, Grinspan A, et al. Pregnancy outcomes from the branded glatiramer acetate pregnancy database. *Int J MS Care.* 2018;20(1):9-14.	The study did not report prevalence of ankyloglossia.
Sedano JR, Arroyo IC, Muñoz MD, Romero CA, Carrera ME, Fraile AG. Anquiloglosia neonatal¿ Existe un exceso de indicación intervencionista. *Acta Pediatr Esp.* 2016;74(2):45-49.	The sample was referred to frenectomy at the beginning of the study.
Serrano-Martinez M, Bagan J, Silvestre F, Viguer MT. Oral lesions in recessive dystrophic epidermolysis bullosa. *Oral Dis.* 2003;9(5):264-268.	The patients had dystrophic epidermolysis bullosa.
Sharma SD, Jayaraj S. Tongue-tie division to treat breastfeeding difficulties: our experience. *J Laryngol Otol.* 2015;129(10):986-989.	The sample had ankyloglossia or was suspected of having breast-feeding difficulties.
Shenoy RP, Agrawal R, Salam TA, Shenoy KP. Screening for temporomandibular disorders and other oral conditions among adolescents in Mangaluru Taluk. *World J Dent.* 2020;11(3):201-205.	The study did not report the prevalence of ankyloglossia.
Shulman JD. Prevalence of oral mucosal lesions in children and youths in the USA. *Int J Paediatr Dent.* 2005;15(2):89-97.	The study did not report prevalence of ankyloglossia.
Solis-Pazmino P, Kim GS, Lincango-Naranjo E, Prokop L, Ponce OJ, Truong MT. Major complications after tongue-tie release: a case report and systematic review. *Int J Pediatr Otorhinolaryngol.* 2020;110356.	Was a case report and systematic review.
Swain SK, Sahu MC, Choudhury J. Speech disorders in children: our experience in a tertiary care teaching hospital in eastern India. *Pediatr Pol.* 2018;93(3):217-220.	The patients had speech disorders.
Taani, DSMQ. Oral health in Jordan. *Int Dent J.* 2004; 54(6 suppl 1):395-400.	The study did not report prevalence of ankyloglossia.
Thapa P, Aryal KK, Dhimal M, et al. Oral health condition of school children in Nawalparasi District, Nepal. *J Nepal Health Res Counc.* 2015;13(29):7-13.	The study did not report prevalence of ankyloglossia.

eTable 2. Continued

STUDY	REASON FOR EXCLUSION
Tobey AH, Kozar AJ. Frequency of somatic dysfunction in infants with tongue-tie: a retrospective chart review. *AAO J.* 2017;28(4):10.	The initial sample had ankyloglossia.
Ünür M, Kayhan KB, Altop MS, Metin ZB, Keskin Y. The prevalence of oral mucosal lesions in children: a single center study. *J Istanb Univ Fac Dent.* 2015;49(3):29-38.	The study did not report prevalence of ankyloglossia.
Vaz AC, Bai PM. Lingual frenulum and malocclusion: an overlooked tissue or a minor issue. *Indian J Dent Res.* 2015;26(5):488-492.	Same sample as Bai PM, Vaz AC. Ankyloglossia among children of regular and special schools in Karnataka, India: a prevalence study. *J Clin Diag Res*. 2014;8(6),36-38.
Velten DB, Zandonade E, Miotto MHMB. Prevalence of oral manifestations in children and adolescents with cancer submitted to chemotherapy. *BMC Oral Health.* 2017;16(1):107.	The study did not report the prevalence of ankyloglossia.
Vieira EMM. *Estudo das Condições de Saúde Bucal e Avaliação da Microbiota Periodontopatogênica de uma População Indígena Brasileira.* Doctoral thesis. Universidade Estadual Paulista; 2009. Accessed September 9, 2022. https://repositorio.unesp.br/handle/11449/102343	Was a sample of adults.
Vieira EM, Ciesielski FI, Gaetti-Jardim EC, et al. Evaluation of oral health in a community of native Brazilians of the Umutina Reservation, Mato Grosso State. *Int J Odontostomat.* 2011;5(1):59-63.	Was a sample of adults.
Vieira-Andrade RG, Martins-Júnior PA, Corrêa-Faria P, et al. Oral mucosal conditions in preschool children of low socioeconomic status: prevalence and determinant factors. *Eur J Pediatr.* 2013;172(5):675-681.	The study did not report prevalence of ankyloglossia.
Wakhanrittee J, Khorana J, Kiatipunsodsai S. The outcomes of a frenulotomy on breastfeeding infants followed up for 3 months at Thammasat University Hospital. *Pediatr Surg Int.* 2016;32(10):945-952.	The initial sample had ankyloglossia and breast-feeding difficulties.
Walls A, Pierce M, Wang H, Steehler A, Steehler M, Harley EH Jr. Parental perception of speech and tongue mobility in three-year olds after neonatal frenotomy. *Int J Pediatr Otorhinolaryngol.* 2014;78(1):128-131.	Was a case-control study with patients with ankyloglossia who underwent frenectomy and controls.
Wei EX, Tunkel D, Boss E, Walsh J. Ankyloglossia: update on trends in diagnosis and management in the United States, 2012-2016. *Otolaryngol Head Neck Surg.* 2020;163(5):1029-1031.	Same database as Walsh J, Links A, Boss E, Tunkel D. Ankyloglossia and lingual frenotomy: national trends in diagnosis and management in the United States, 1997-2012. *Otolaryngol Head Neck Surg.* 2017;156(4):735-740.
Xu L, Han P, Liu Y, et al. Study on the effect of kidney transplantation on the health of the patients' offspring: a report on 252 Chinese children. *Cell Biochem Biophys.* 2014;68(1):173-179.	The sample had kidney transplantation with several health problems.
Yin W, Yang YM, Chen H, et al. Oral health status in Sichuan Province: findings from the oral health survey of Sichuan, 2015–2016. *Int J Oral Sci.* 2017;9(1):10-15.	The study did not report prevalence of ankyloglossia.
Yin Y, Yu Z, Zhao M, Wang Y, Guan X. Comprehensive evaluation of the risk of lactational mastitis in Chinese women: combined logistic regression analysis with receiver operating characteristic curve. *Biosci Rep.* 2020;40(3):BSR20190919.	Was a case-control study of mastitis in lactating women.
Yilmaz AE, Gorpelioglu C, Sarifakioglu E, Dogan DG, Bilici M, Celik N. Prevalence of oral mucosal lesions from birth to two years. *Niger J Clin Pract.* 2011;14(3):349-353.	The study did not report prevalence of ankyloglossia.
Yoon AJ, Zaghi S, Ha S, Law CS, Guilleminault C, Liu SY. Ankyloglossia as a risk factor for maxillary hypoplasia and soft palate elongation: a functional-morphological study. *Orthod Craniofac Res.* 2017;20(4):237-244.	Was a sample of adults.
Zeng H, Cai H, Wang Y, Shen Y. Growth and development of children prenatally exposed to telbivudine administered for the treatment of chronic hepatitis B in their mothers. *Int J Infect Dis.* 2015;33:97-103.	The mothers were treated for chronic hepatitis B during pregnancy, and the study assessed the growth and development of their children.

eTable 3. Study characteristics and data extraction.

AUTHORS, YEAR	COUNTRY, SETTING	SAMPLE, NO.	SAMPLE WITH ANKYLOGLOSSIA, NO.	POPULATION	DIAGNOSTIC CRITERIA
Sedano,[e65] 1975	United States, 4 schools	6,810	8	Children and adolescents	Own criteria
Jorgenson and Colleauges,[e39] 1982	United States, hospital	2,164	39	Infants	Own criteria
Sawyer and Colleagues,[e64] 1984	Nigeria, 3 schools	2,203	5	Children and adolescents	Own criteria
Salem and Colleagues,[e63] 1987	Saudi Arabia, schools of Gizan region	1,932	2	Children	Own criteria
Sedano and Colleagues,[e66] 1989	Mexico, a nonprobabilistic sample of 73 schools in 6 cities	32,022	266	Children and adolescents	Own criteria
Friend and Colleagues,[e30] 1990	United States, 1 hospital	500	22	Infants	Own criteria
Flink and Colleagues,[e27] 1994	Sweden, hospital	1,021	25	Infants	Own criteria
Livingstone and Colleagues,[e43] 2000	Canada, hospital	21	2	Infants	Not reported
Messner and Colleagues,[e48] 2000	United States, 1 hospital	1,041	50	Infants	Not reported
Ballard and Colleagues,[e12] 2002	United States, hospital	3,036	127	Infants	HATLFF*
Garcia-Pola and Colleagues,[e32] 2002a	Spain, dental health center population in 1999 (Asturias)	962 children and 732 adolescents	23 children; 32 adolescents	Children and adolescents	Own criteria
Garcia-Pola and Colleagues,[e33] 2002b	Spain, national database in 1991 (Asturias)	624	13	Children	Own criteria
Navarro and López,[e52] 2002	Cuba, 1 primary school	829	29	Children	Not reported
Voros-Balog and Colleagues,[e73] 2003	Hungary, hospital	610 children; 407 adolescents	3 children; 6 adolescents	Children and adolescents	Own criteria
Cinar and Onat,[e23] 2005	Turkey, screening program in schools from 1999 to 2000 in Istanbul	940	19	Children and adolescents	Own criteria
Ekenze and Colleagues,[e26] 2005	Nigeria, hospital	218	8	Infants, children, and adolescents	Not reported
Mumcu and Colleagues,[e51] 2005	Turkey, cluster sampling with home visits in Istanbul	269	0	Children and adolescents	Nonvalidated diagnostic criteria (WHO†)
Ricke and Colleagues,[e61] 2005	United States, hospitals	3,490	148	Infants	HATLFF
Sunday-Adeoye and Colleagues,[e69] 2007	Nigeria, 1 hospital	33,659	19	Infants	Not reported
Tomizawa and Colleagues,[e70] 2007	Japan, dental hospital	234	14	Infants	Not reported
Freudenberger and Colleagues,[e29] 2008	Mexico, low-income hospital	2,182	231	Infants	Own criteria
Hipólito and Martins,[e36] 2010	Brazil, 2 re-education centers	231	1	Adolescents	Not reported
Majorana and Colleagues,[e45] 2010	Italy, clinical charts from a dental school	10,128	22	Infants and children	Nonvalidated diagnostic criteria
Vieira and Colleagues,[e71] 2010	Brazil, a census of 1 indigenous reserve	160	65	Infants, children, and adolescents	Own criteria (Formolo)
Ambika and Colleagues,[e7] 2011	India, school	1,003	84	Children and adolescents	Not reported

* HATLFF: Hazelbaker Assessment Tool for Lingual Frenulum Function. † WHO: World Health Organization protocol for screening infants, without a description of the criteria. ‡ NTST: Neonatal Tongue Screening Test. § BTAT: Bristol Tongue Assessment Tool.

eTable 3. Continued

AUTHORS, YEAR	COUNTRY, SETTING	SAMPLE, NO.	SAMPLE WITH ANKYLOGLOSSIA, NO.	POPULATION	DIAGNOSTIC CRITERIA
Çetinkaya and Colleagues,[e19] 2011	Turkey, 1 hospital (from 2006 to 2007)	2,021	6	Infants	Own criteria
Jahanbani and Colleagues,[e37] 2012	Iran, a probabilistic sample from schools	1,020	5	Adolescents	Not reported
Morisso and Colleagues,[e50] 2012	Brazil, public schools	1,526	21	Children and adolescents	Not reported
Rai and Colleagues,[e58] 2012	India, not reported	1,800	4	Children	Not reported
Anaya and Colleagues,[e8] 2013	Colombia, dental school	134	7	Children	Own criteria
Ngerncham and Colleagues,[e53] 2013	Thailand, 1 dental hospital	2,679	1,028	Infants	Own criteria
Bai and Vaz,[e11] 2014	India, school	700	59	Children and adolescents	Kotlow classification
Chiang and Colleagues,[e22] 2014	Taiwan, hospital	81	1	Children and adolescents	Nonvalidated diagnostic criteria (WHO)
González Jiménez and Colleagues,[e34] 2014	Spain, 6 public hospitals	677	82 HATLFF + Corrylos; 53 Corryllos	Infants	HATLFF and Coryllos classification
Haham and Colleagues,[e35] 2014	Israel, 6 public hospitals	200	76	Infants	Coryllos classification
Jamilian and Colleagues,[e38] 2014	Iran, stratified sample from schools in a city	300	31	Children and adolescents	Kotlow classification
Mohan and Colleagues,[e49] 2014	India, 3 orphanages and 3 schools	160	1	Children and adolescents	Nonvalidated diagnostic criteria (WHO)
Riskin and Colleagues,[e62] 2014	Israel, 1 hospital	21,424	250	Infants	Own criteria
Basalamah and Baroudi,[e15] 2016	Saudi Arabia, school	1,000	18	Children and adolescents	Not reported
Lopes and Colleagues,[e44] 2016	Brazil, hospital	190	24	Infants	NTST[‡]
Fujinaga and Colleagues,[e31] 2017	Brazil, 1 hospital	139	1	Infants	NTST
Kumar and Colleagues,[e40] 2017	India, 3 private hospitals	25,786	64	Infants	Academy of Breastfeeding Medicine
Lisonek and Colleagues,[e42] 2017	Canada, national database (all hospital-based live births in Canada from 2002 to 2015 (except Quebec)	3,611,986	40,457	Infants	Own criteria
Puapornpong and Colleagues,[e57] 2017	Thailand, hospital	1,649	122	Infants	Kotlow classification
Walsh and Colleagues,[e75] 2017	United States, Kids Inpatient Database (KID)	37,990,863	80,842	Infants, children, and adolescents	Not reported
Yoon and Colleagues,[e76] 2017	United States, 1 private orthodontic practice	140 children; 436 adolescents	7 children; 14 adolescents	Children and adolescents	Kotlow classification
Avila and Colleagues,[e10] 2018	Cuba, clinic	1,095	46	Infants	Own criteria
Brandão and Colleagues,[e17] 2018	Brazil, hospital	268	6	Infants	NTST
Chandler and Colleagues,[e20] 2018	Brazil, hospital	168	3	Infants	Own criteria

eTable 3. Continued

AUTHORS, YEAR	COUNTRY, SETTING	SAMPLE, NO.	SAMPLE WITH ANKYLOGLOSSIA, NO.	POPULATION	DIAGNOSTIC CRITERIA
Martinelli and Colleagues,[e46] 2018	Brazil, not reported	1,715	558	Infants	NTST
Perez-Aguirre and Colleagues,[e54] 2018	Mexico, 2 hospitals	2,216	33	Infants	Not reported
Walker and Colleagues,[e74] 2018	United States, hospital	100	20	Infants	Own criteria
Bandaru and Colleagues,[e13] 2019	India, school	568	197	Children and adolescents	Not reported
Campanha and Colleagues,[e18] 2019	Brazil, hospital	130	25	Infants	NTST
De Oliveira and Colleagues,[e24] 2019	Brazil, hospital	400	36	Infants	Not reported
Krittika and Don,[e41] 2019	India, dental clinic	200	5	Children and adolescents	Own criteria
Petousis-Harris and Colleagues,[e55] 2019	New Zealand, Health National Index Database of New Zeland	69,389	1,284	Infants	Not reported
Villa and Colleagues,[e72] 2019	Italy, 1 school	504	114	Children and adolescents	Kotlow classification
Araujo and Colleagues,[e9] 2020	Brazil, hospital	449	14	Infants	Lingual Frenulum Protocol for Infants and BTAT[§]
Becerra-Culqui and Colleagues,[e16] 2020	United States, pregnancy registry (Kaiser Permanente Southern California)	55	1	Infants	Not reported
Chang and Colleagues,[e21] 2020	Taiwan, probabilistic residential sample	981	12	Children	Nonvalidated diagnostic criteria (WHO)
Dutra and Colleagues,[e25] 2020	Brazil, hospital	1,350	123	Infants	NTST
Fraga and Colleagues,[e28] 2020	Brazil, 7 public maternities of Recife	882	21 NTST; 96 BTAT	Infants	NTST and BTAT
Potter and Colleagues,[e56] 2020	United States, research institute part of another study	324	20	Children	HATLFF (modified)
Razdan and Colleagues,[e59] 2020	United States, hospital	161	6	Infants	Modification of HATLFF and BTAT
Zen and Colleagues,[e77] 2020	Brazil, 1 hospital	174	13	Infants	NTST
Barberá-Perez and Colleagues,[e14] 2021	Spain, hospital (cluster sampling of 1 city)	305	36	Infants	HATLFF and Coryllos classification
Maya-Enero and Colleagues,[e47] 2021	Spain, 1 hospital	1,392	275 Coryllos types 1, 2, 3, 4; 453 Corryllos types 1 and 2 with symptomatic HATLFF	Infants	Coryllos classification and HATLFF
Rech and Colleagues,[e60] 2021	Peru, 1 maternity	304	15	Infants	Lingual Frenulum Protocol for Infants
Shah and Colleagues,[e67] 2021	India, school	7,533	2	Children and adolescents	Not reported
Souza-Oliveira and Colleagues,[e68] 2021	Brazil, 1 hospital	391	58	Infants	NTST

e1. Amir LH, James JP, Donath SM. Reliability of the Hazelbaker assessment tool for lingual frenulum function. *Int Breastfeed J*. 2006;1(1):3.
e2. Ingram J, Johnson D, Copeland M, Churchill C, Taylor H, Emond A. The development of a tongue assessment tool to assist with tongue-tie identification. *Arch Dis Child Fetal Neonatal Ed*. 2015;100(4):F344-F348.
e3. Coryllos EW, Genna CW, Salloum AC. Congenital tonguetie and its impact on breastfeeding. In: *Breastfeeding: Best for Baby and Mother*. American Academy of Pediatrics; 2004.
e4. Kotlow LA. Ankyloglossia (tongue-tie): a diagnostic and treatment quandary. *Quintessence Int*. 1999;30(4):259-262.
e5. Martinelli RLC, Marchesan IQ, Lauris JR, Honório HM, Gusmão RJ, Berretin-Felix G. Validity and reliability of the neonatal tongue screening test. *Rev CEFAC*. 2016;18(6):1323-1331.
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