

Treatment of Ankyloglossia for Reasons Other Than Breastfeeding: A Systematic Review

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abstract

BACKGROUND AND OBJECTIVE: Children with ankyloglossia, an abnormally short, thickened, or tight lingual frenulum, may have restricted tongue mobility and sequelae, such as speech and feeding difficulties and social concerns. We systematically reviewed literature on feeding, speech, and social outcomes of treatments for infants and children with ankyloglossia.

METHODS: Medline, PsycINFO, Cumulative Index of Nursing and Allied Health Literature, and Embase were searched. Two reviewers independently assessed studies against predetermined inclusion/exclusion criteria. Two investigators independently extracted data on study populations, interventions, and outcomes and assessed study quality.

RESULTS: Two randomized controlled trials, 2 cohort studies, and 11 case series assessed the effects of frenotomy on feeding, speech, and social outcomes. Bottle feeding and social concerns, such as ability to use the tongue to eat ice cream and clean the mouth, improved more in treatment groups in comparative studies. Supplementary bottle feedings decreased over time in case series. Two cohort studies reported improvement in articulation and intelligibility with treatment. Other benefits were unclear. One randomized controlled trial reported improved articulation after Z-frenuloplasty compared with horizontal-to-vertical frenuloplasty. Numerous noncomparative studies reported speech benefits posttreatment; however, studies primarily discussed modalities, with outcomes including safety or feasibility, rather than speech. We included English-language studies, and few studies addressed longer-term speech, social, or feeding outcomes; nonsurgical approaches, such as complementary and alternative medicine; and outcomes beyond infancy, when speech or social concerns may arise.

CONCLUSIONS: Data are currently insufficient for assessing the effects of frenotomy on nonbreastfeeding outcomes that may be associated with ankyloglossia.

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Dr McPheeters conceptualized and designed the study, and drafted the initial manuscript; Drs Francis, Epstein, Morad, Chinnadurai, and Kohanim carried out the initial analyses, and reviewed and revised the manuscript; and all authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

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Ankyloglossia is a congenital condition characterized by an abnormally short, thickened, or tight lingual frenulum that restricts mobility of the tongue. Ankyloglossia may be associated with other craniofacial abnormalities, but is also often an isolated anomaly.¹ It can reduce tongue mobility and has been associated with functional limitations in breastfeeding, swallowing, and articulation; orthodontic problems, including malocclusion, open bite, and separation of lower incisors; mechanical problems related to oral clearance; and psychological stress in affected individuals.

Although most ankyloglossia research is focused on infant breastfeeding issues, concerns beyond infancy also have been recognized, including speech-related issues, such as difficulty with articulation, and social concerns related to limited tongue mobility. There may be long-term feeding sequelae, unrelated to breastfeeding, such as difficulty with bottle-feeding and deglutition. Individuals with untreated ankyloglossia may experience trouble with licking foods, such as ice cream, kissing, drooling, playing wind instruments, oral hygiene, and licking the lips. Self-esteem or psychological issues also may be a concern for affected older patients. However, the absence of data on the natural history of untreated ankyloglossia creates uncertainty. Some propose that a short frenulum will elongate spontaneously with progressive stretching and thinning of the frenulum with age and use, and thus no treatment is necessary.¹ However, there are no prospective longitudinal data to support this assertion. Absence of evidence makes it difficult to objectively inform parents about the long-term implications of ankyloglossia, which complicates the decision-making process, and guidance to date has focused exclusively on breastfeeding issues.

This review describes outcomes reported in studies identified for a broader Agency for Healthcare Research and Quality (AHRQ)-commissioned systematic review of interventions for infants and children with congenital ankyloglossia. The aim of this article was to investigate the benefits and harms of treatment of infants and children who are born with ankyloglossia and who present some time in childhood for treatment because of nonbreastfeeding difficulties. The full review and its protocol are available at <http://www.effectivehealthcare.ahrq.gov/search-for-guides-reviews-and-reports/?pageaction=displayproduct&productID=1991>.

METHODS

Search Strategy

We searched Medline through the PubMed interface, the Cumulative Index of Nursing and Allied Health Literature, Embase (Excerpta Medica Database), and PsycINFO (psychology and psychiatry literature), with no publication date restrictions by using vocabulary terms and key terms related to ankyloglossia and its therapies. Reference lists of all included articles and recent reviews related to ankyloglossia therapies were hand-searched to identify any additional relevant articles.

Study Selection

We sought studies to answer the following key questions:

- What are the benefits of various treatments in newborns, infants, and children with ankyloglossia intended to prevent, mitigate, or remedy attributable medium- and long-term feeding sequelae, including trouble with bottle-feeding, spilling and dribbling, difficulty moving food boluses in the mouth, and deglutition?
- What are the benefits of various treatments in infants and children with ankyloglossia intended to

prevent, mitigate, or remedy attributable medium- and long-term other sequelae, including articulation disorders, poor oral hygiene, oral and oropharyngeal dysphagia, sleep-disordered breathing, orthodontic issues including malocclusion, open bite due to reverse swallowing, lingual tipping of the lower central incisors, separation of upper central incisors, crowding, narrow palatal arch, and dental caries?

- What are the benefits of various treatments for ankyloglossia in children up to 18 years of age intended to prevent or address social concerns related to tongue mobility (ie, speech, oral hygiene, excessive salivation, kissing, spitting while talking, and self-esteem)?

Inclusion and exclusion criteria were developed in consultation with technical expert panel of clinicians and researchers who treat and study ankyloglossia. Treatment effectiveness data were extracted from comparative study designs (ie, randomized controlled trials [RCTs], nonrandomized trials, prospective or retrospective cohort studies). Harms data were collected comprehensively from all study types, including case series and case reports. Each study was reviewed independently by 2 investigators against inclusion criteria (Table 1) with adjudication by a senior investigator as needed.

Data Extraction

Two investigators independently extracted data about study design; descriptions of the study populations, interventions, and comparison groups; and baseline and outcomes data (including harms/adverse events) by using standardized forms. Principal outcomes of interest for this analysis were feeding issues, including dribbling and choking; speech outcomes, including articulation; and social satisfaction. Outcomes related to breastfeeding

TABLE 1 Inclusion and Exclusion Criteria

Category	Criteria
Study population	Inclusion: Children ages 0–18 with ankyloglossia or ankyloglossia with concomitant tight labial frenulum (lip-tie) Exclusion: Studies with participants with Van der Woude syndrome, Pierre Robin syndrome, Down syndrome, or craniofacial abnormalities were excluded, as were studies of premature infants (<37 wk of gestation ¹⁹)
Publication languages	Inclusion: English Exclusion: Non-English
Admissible evidence (study design and other criteria)	Included study designs RCTs, prospective and retrospective cohort studies, non-RCTs, prospective and retrospective case series, and crossover studies Case reports to assess harms Other criteria Original research studies providing sufficient detail regarding methods and results to enable use and aggregation of the data and results Studies must address 1 or more of the following: <ul style="list-style-type: none"> • Surgical interventions (simple anterior frenotomy, laser frenulectomy, posterior frenulectomy, Z-plasty repair) • Nonsurgical treatments include complementary and alternative medicine (CAM) therapies (eg, craniosacral therapy, myofascial release, and other chiropractic therapies), lactation intervention, speech therapy, physical therapy, oral motor therapy, and stretching exercises/therapy • Baseline and outcome data (including harms) related to interventions for ankyloglossia Relevant outcomes must be able to be extracted from data in the papers Data must be presented in the aggregate (versus individual participant data)

are presented in the full report and in a separate article.

Study Quality Assessment

Two team members independently conducted risk-of-bias assessment of each study by using forms developed by the review team based on the Evidence-based Practice Center (EPC) methods guidance with input from content experts. Discrepancies were adjudicated through discussion between the assessors to reach consensus or via a senior reviewer. Results of assessments were then translated to the AHRQ quality designation standards of “good,” “fair,” and “poor.”

Strength of evidence of current research was assessed using methods established in the AHRQ Effective Health Care Program’s *Methods Guide for Effectiveness and Comparative Effectiveness Reviews*.² Assessments

were based on 5 domains: study limitations, consistency in direction of effect, directness in measuring intended outcomes, precision of effect, and reporting bias. (Table 2). Strength of evidence was determined for major intervention-outcome pairs using a prespecified approach described in the full review.³

Data Synthesis

The paucity of research and heterogeneity obviated the ability to perform any meta-analyses. Therefore, characteristics of the study populations and interventions were summarized and descriptive statistics used to report study outcomes.

RESULTS

In all, 15 studies addressed the benefits of treatment intended to improve nonbreastfeeding outcomes (Fig 1), including feeding, speech, and

social outcomes; there were no studies of nonsurgical treatments.^{3–17}

Speech Outcomes

After breastfeeding, speech concerns were the second most prevalent outcome described in the ankyloglossia literature. The specific outcomes measured varied among these studies, but were generally related to assessments of articulation and intelligibility. A speech-language pathologist measured speech outcomes in 2 studies^{7,12} with the third using parent report.⁴ Although we looked for them, no studies included data related to sleep-disordered breathing, dental issues, or dysphagia in the nonbreastfeeding child. Two cohort studies attempted to assess the effectiveness of frenotomy,^{4,7} and 1 RCT compared 2 surgical approaches to frenotomy.¹²

The 1 poor-quality RCT reported improved articulation in patients treated with 4-flap Z-frenuloplasty compared with horizontal-to-vertical frenuloplasty.¹² This study did not compare results to no treatment. Both cohort studies were of poor quality, and both^{4,7} reported an improvement in articulation and intelligibility with ankyloglossia treatment, but benefits in word, sentence, and fluent speech were not demonstrated. Numerous noncomparative studies reported a speech benefit after treating ankyloglossia; however, these studies primarily discussed modalities, with safety and feasibility as the main outcome, rather than measures of speech itself.^{5,6,8,10,13,14,16,18} Given the lack of good-quality studies and limitations in the measurement of outcomes, the strength of the evidence for the effect of surgical interventions to improve speech and articulation is insufficient.

Feeding Outcomes

Three studies provided data specifically on feeding processes other than breastfeeding. One was an RCT¹¹ (fair quality), 1 was a

TABLE 2 Strength of Evidence Grades and Definitions

Grade	Definition
High	We are very confident that the estimate of effect lies close to the true effect for this outcome. The body of evidence has few or no deficiencies. We believe that the findings are stable (ie, another study would not change the conclusions).
Moderate	We are moderately confident that the estimate of effect lies close to the true effect for this outcome. The body of evidence has some deficiencies. We believe that the findings are likely to be stable, but some doubt remains.
Low	We have limited confidence that the estimate of effect lies close to the true effect for this outcome. The body of evidence has major or numerous deficiencies (or both). We believe that additional evidence is needed before concluding either that the findings are stable or that the estimate of effect is close to the true effect.
Insufficient	We have no evidence, we are unable to estimate an effect, or we have no confidence in the estimate of effect for this outcome. No evidence is available or the body of evidence has unacceptable deficiencies, precluding reaching a conclusion.

Excerpted from Berkman et al 2014.²⁰

poor-quality retrospective cohort study,⁴ and the remaining study was a case series.⁹ All studies were single-center or single-surgeon studies. Two studies were conducted in the United Kingdom^{9,11} and 1 study in the United States.⁴ Comparative data

were included in 2 studies.^{4,11} In summary, the RCT¹¹ randomized infants born with ankyloglossia and diagnosed within the first 5 months with breastfeeding or bottle-feeding problems to either frenotomy ($n = 28$) or intensive advice and support

from lactation consultants ($n = 29$; control group). Outcomes were based solely on maternal report within 48 hours of randomization. However, the control group was offered, and most elected to receive, frenotomy within that time frame, thus eliminating the ability to assess “medium- to long-term” comparative feeding outcomes. Nonetheless, among pretreatment bottle-fed infants, 76% had major problems with dribbling, and 71% had “excess wind” (gas). Mothers reported significant improvement in feeding in all 8 who received the frenotomy and in none who did not. The interval to ascertainment of outcomes was not specifically reported, but outcomes were obtained within the first 4 weeks of life. The retrospective cohort study compared parent-reported outcomes

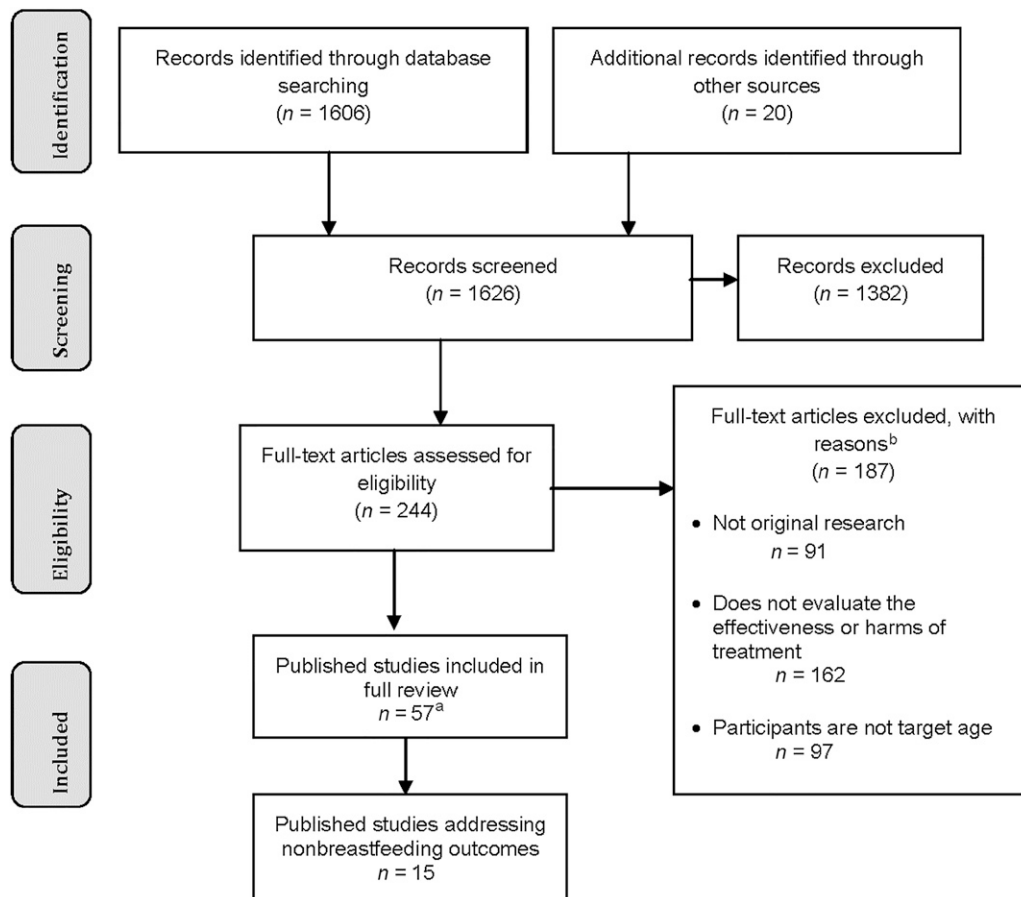


FIGURE 1

Disposition of articles identified by the search strategy. ^aIncludes data from 1 unpublished thesis. ^bNumbers do not tally, as studies could be excluded for multiple reasons.

at age 3 years for children born in 2010 who (1) received frenotomy for tongue-tie ($n = 71$; frenotomy group), (2) were offered but declined frenotomy for tongue-tie ($n = 15$; no-frenotomy group), and (3) children without ankyloglossia ($n = 18$; control group).⁴ Three questions rated on a 5-point Likert scale were used to assess a child's difficulty (1) cleaning his or her teeth with the tongue, (2) licking the outside of his or her lips, and (3) eating ice cream. For all questions, the frenotomy group performed better than the no-frenotomy group at age 3 years and did not differ significantly from the comparison group without ankyloglossia. *P* values were presented without reporting the central tendency (eg, median, mean) or variance (interquartile range, SD) from which they were calculated. Therefore, further comparative description or analysis was not possible.

Bottle-feeding and ability to use the tongue to eat ice cream and clean the mouth (ie, oral hygiene) improved more in treatment groups in the 2 comparative studies that considered these end points,^{4,11} none of which was of good quality. Supplementary bottle-feedings decreased over time in the case series.⁹ With only 2 comparative studies, both with significant methodological limitations, including heterogeneous populations and measured outcomes, existing data are insufficient to draw conclusions about the benefits of surgical interventions on medium- and long-term or nonbreastfeeding feeding (ie, bottle) outcomes for infants and children with ankyloglossia.

Social Outcomes

Only 1 poor-quality comparative, retrospective cohort study assessed outcomes related to social concerns other than speech.⁴ It reported significantly improved ability to clean teeth with tongue, licking outside of lips, and eating ice cream in the treatment group compared with

untreated participants. The intermediate outcome of improved tongue movement or mobility after ankyloglossia repair was assessed in 2 comparative studies: 1 poor-quality RCT^{7,12} and 1 poor-quality cohort study.⁷ The RCT assessed tongue mobility by using 2 different surgical techniques for treating ankyloglossia and found that both approaches significantly improved tongue mobility, but that Z-frenuloplasty was superior in terms of a measure of articulation problems.^{7,12} In the cohort study, individuals with untreated ankyloglossia had the worst tongue mobility followed in order by children with treated ankyloglossia, and those with no history of ankyloglossia at approximately age 6.⁷

With only 1 poor-quality comparative study, strength of the evidence related to the ability of treatment of ankyloglossia to alleviate social concerns is currently insufficient. Also, with only 3 comparative studies with small sizes and limitations in the measurement of outcomes related to tongue mobility, we considered the strength of the evidence for the effect of surgical interventions to improve the short-term outcome of mobility to be insufficient.

Harms

Our assessment of harms is described in the full report and the other article, but harms were generally mild to nonexistent, and included bleeding that resolved quickly, as would be expected. No harms of treatment associated with older children's care were reported.

DISCUSSION

Ankyloglossia studies have burgeoned largely secondary to the observed benefit of breastfeeding on infant health. However, some practitioners and family also think that frenotomy treatment of ankyloglossia with frenotomy has medium- to long-term speech,

feeding, and social benefits to the child. These perpetuated beliefs are based on a small body of literature, few of which use a comparison group. The scientific evidence is therefore insufficient to inform patients, their families, and clinicians about the potential benefits or harms of ankyloglossia treatment when considered for reasons other than breastfeeding difficulties.

Specifically, our review identified a total of 4 comparative studies that evaluated outcomes other than breastfeeding (ie, feeding, speech, social concerns) and none was of good quality. Anecdotally, clinicians on our research team, as well as key informants, report that patients present with speech difficulty and with social issues, such as discomfort with kissing, but those clinical scenarios are currently unrepresented in the effectiveness literature. As such, there is a lack of evidence base to counsel patients and their families on what medium- and long-term outcomes to expect from surgical or other forms of intervention.

Only 1 study with comparative poor-quality retrospective cohort data addressed feeding issues other than breastfeeding.⁴ The study's intervention group received frenotomy for ankyloglossia, which was identified within the first month of life, and was compared with mother-child dyads that were also offered, but declined, frenotomy for the same indication in the same time period. Although this is a common decisional dilemma for parents of infants with congenital ankyloglossia, in usual clinical care, surgical intervention is typically not considered unless congenital ankyloglossia co-occurs with breastfeeding concerns. Thus, treatment of ankyloglossia for other types of other feeding difficulties is poorly studied and understood, as the preponderance of related literature focuses on mitigating those issues related to breastfeeding problems.

Studies providing data on speech outcomes were all rated as poor quality and included an RCT^{7,12} and 2 retrospective cohort studies.^{4,7} The RCT compared 2 different frenuloplasty approaches for treatment of children of a mean age of ~6 years with a tight frenulum effecting articulation or intelligibility¹² and found that children treated with either 4-flap Z-frenuloplasty or horizontal-to-vertical frenuloplasty had significant improvement in articulation as judged by trained speech-language pathologists. This benefit, however, did not translate into improved fluent speech scores. Applicability and generalizability of these findings is limited because of the small sample size, the inadequate characterization of candidate children, and because specialist pediatric craniofacial surgeons performed these surgeries at an urban tertiary care center.

Similarly, cohort studies were performed solely in urban tertiary care centers. One assessed speech outcomes on 3-year-old children treated for ankyloglossia as neonates compared with those who had untreated ankyloglossia, and a control group without a history of ankyloglossia.⁴ Pediatric otolaryngologists made the diagnosis using prespecified diagnostic criteria. However, the reason that infants presented for treatment of ankyloglossia was not described. Further limiting its applicability is that outcomes were assessed by using a nonvalidated parent-reported telephone survey. Thus, there was no objective speech evaluation. There is a high risk for ascertainment bias; that is, parents of children with ankyloglossia would have a higher index of concern for speech issues than those whose children never had been diagnosed with tongue mobility restriction. The second poor-quality retrospective cohort had a relatively small sample size ($n = 23$) of children ~6 years of age who were divided into those with treated ankyloglossia, untreated ankyloglossia, and a control

group.⁷ It was performed at a tertiary care facility in an Israeli urban center. Unfortunately, its applicability is limited for reasons similar to those previously described except that speech-language pathologists objectively assessed speech by using a standardized assessment tool. Both retrospective studies lacked explanations about the rationale for initial surgical intervention or reason the parent chose not to intervene (eg, tongue-tie severity, breastfeeding difficulties).

The population studied to evaluate the benefit of ankyloglossia repair for social concerns included children and adults with wide variation in ages. Studies were rated as poor quality, were retrospective, and few in number. Outcomes in 1 study were assessed by parental report and subject to recall bias⁴ and social outcomes assessed were limited to licking lips, cleaning teeth with tongue, and eating ice cream. The social concerns or implications of these issues are unclear. No other comparative study considered social concerns. In addition, at least 2 case series did consider the impact of ankyloglossia on kissing and playing a wind instrument¹³ and drooling and oral hygiene.⁸

Limiting these findings was the absence of preprocedure status of these patients in these domains and how each was assessed. In addition to not including a comparison group of any type, case series are strongly affected by selection bias and are, by nature, not comparative studies. Moreover, patients were selected either by retrospective chart review or as they presented to otolaryngology clinics. Only surgical interventions were studied and no 2 studies measured the same outcomes. In most, social concerns were measured as a secondary outcome.

Limitations of Evidence Base

There are glaring deficiencies in this evidence base. The most salient is

lack of comparative data, against a backdrop of inadequate natural history data related to ankyloglossia. Clinicians and policymakers lack the requisite information to properly counsel parents and children about the benefits and harms of ankyloglossia treatment as it pertains to outcomes other than breastfeeding. In practice, uncertainty in evidence can be associated with parochialism and practice variation. With a growing emphasis in evidence-based practice across medical specialties, future research should characterize which, if any, tongue-tied infants or children need surgical intervention. It is also notable that despite use of alternative nonsurgical treatments in practice, including craniosacral therapy and lactation consultation, no comparative studies in the literature assessed their effectiveness.

Study Limitations

This review included only studies published in English. Our scan and review of non-English references revealed a high percentage of noneligible items. Given the high percentage of noneligible items in this scan (97%), we feel that excluding non-English studies did not introduce significant bias into the review. The review focused on comparative studies (studies including an intervention and a comparison group). However, we did identify case series data to determine whether it could provide support for the comparative findings, and to identify potential harms of intervention.

Research Gaps

Because there are so few available data on other feeding outcomes, the entire research question related to feeding outcomes other than breastfeeding represents a gap and a potential area for future research. Similarly, substantially more research is needed to consider whether treatment of ankyloglossia in infancy

prevents future production difficulties, as well as whether treatment later in life with frenotomy leads to improvement when speech problems exist. To conduct this research effectively, methods for evaluating risk and presence of speech production difficulties will need to be standardized, and outcomes agreed on. Furthermore, an understanding of the natural history of speech concerns in children with ankyloglossia is of paramount importance and is currently lacking, as are comparative studies that use standardized measurement tools for speech outcomes.

Finally, no standard definitions of tongue mobility or established norms for mobility exist, and further research is needed to determine such

parameters. Standardization would allow for comparability between studies and could allow potential study data aggregation and meta-analysis in future systematic reviews. Social concerns are difficult to measure objectively, so there will likely always be a subjective component to measuring these types of outcomes. Larger studies that assess both treated and untreated individuals could provide useful data to minimize the potential bias found in the existing literature. Similarly, future research in objective measurement tools, or validated self-report tools, is needed.

CONCLUSIONS

Although individuals and clinicians report anecdotally that challenges

and concerns persist into childhood related to feeding, speech, and social outcomes among children with ankyloglossia, evidence is sparse on management of the condition. Very little is known about whether ankyloglossia treatment, particularly frenotomy, is associated with positive changes in these nonbreastfeeding outcomes.

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COMPANION PAPER: A companion to this article can be found on page XXX, and online at www.pediatrics.org/cgi/doi/10.1542/peds.2015-0658.

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