



Effective Health Care Program

Treatments for Ankyloglossia and Ankyloglossia With Concomitant Lip-Tie

Executive Summary

Introduction

Ankyloglossia is a congenital condition characterized by an abnormally short, thickened, or tight lingual frenulum, or an anterior attachment of the lingual frenulum, that restricts mobility of the tongue.¹ It variably causes reduced anterior tongue mobility and has been associated with functional limitations in breastfeeding; swallowing; articulation; orthodontic problems, including malocclusion, open bite, and separation of lower incisors; mechanical problems related to oral clearance; and psychological stress. One review including studies of infants, children, and adults reported rates of ankyloglossia ranging from 0.1 to 10.7 percent,² but definitive incidence and prevalence statistics are elusive due to an absence of a criterion standard or clinically practical diagnostic criteria.

Recognition of potential benefits of breastfeeding in recent years has resulted in a renewed interest in the functional sequelae of ankyloglossia. In infants with anterior or posterior ankyloglossia, there is a reported 25- to 80-percent incidence of breastfeeding difficulties, including failure to thrive, maternal nipple damage, maternal breast pain, poor milk supply, maternal breast engorgement, and refusing the breast.² Ineffective latch is hypothesized to underlie these problems.

Effective Health Care Program

The Effective Health Care Program was initiated in 2005 to provide valid evidence about the comparative effectiveness of different medical interventions. The object is to help consumers, health care providers, and others in making informed choices among treatment alternatives. Through its Comparative Effectiveness Reviews, the program supports systematic appraisals of existing scientific evidence regarding treatments for high-priority health conditions. It also promotes and generates new scientific evidence by identifying gaps in existing scientific evidence and supporting new research. The program puts special emphasis on translating findings into a variety of useful formats for different stakeholders, including consumers.

The full report and this summary are available at www.effectivehealthcare.ahrq.gov/reports/final.cfm.

Mechanistically, infants with restrictive ankyloglossia cannot extend their tongues over the lower gumline to form a proper seal and therefore use their jaws to keep the breast in the mouth for breastfeeding.



Adequate tongue mobility is required for breastfeeding, and infants with ankyloglossia often cannot overcome their deficiency with conservative measures such as positioning and latching techniques, thereby requiring surgical correction.²

Nonetheless, consensus on ankyloglossia's role in breastfeeding difficulties is lacking. A minority of surveyed pediatricians (10%) and otolaryngologists (30%) believe it commonly affects feeding, while 69 percent of lactation consultants feel that it frequently causes breastfeeding problems.³ Therefore, depending on the audience, enthusiasm for its treatment varies. Currently, the U.K. National Health Service and the Canadian Paediatric Society recommend treatment only if it interferes with breastfeeding.^{4,5} A standard definition of "interference" with breastfeeding is not provided, leaving room for interpretation and variation in treatment thresholds. The absence of data on the natural history of untreated ankyloglossia further promulgates uncertainty. Some propose that a short frenulum elongates spontaneously due to progressive stretching and thinning of the frenulum with age and use.¹ However, there are no prospective longitudinal data on the congenitally short lingual frenulum. Without this information it is difficult to inform parents fully about the long-term implications of ankyloglossia, thereby complicating the decision-making process.

Although most ankyloglossia research is focused on the infant and breastfeeding issues, concerns beyond infancy include speech-related issues, such as difficulty with articulation, and social concerns related to limited tongue mobility. Individuals with untreated ankyloglossia may experience difficulty with oral mechanism, particularly in relation to licking ice cream, kissing, drooling, playing wind instruments, and licking the lips. Self-esteem or psychological issues may also be a concern for affected older patients.

Treatment Strategies

Ankyloglossia may be treated with surgical or nonsurgical approaches. Surgical modalities include frenotomy, frenulectomy, and frenuloplasty. These interventions involve clipping or cutting of the lingual frenulum, generally without sedation. Laser frenotomy or frenulotomy has also been described, and proponents argue that its use is more exact and provides better hemostasis than standard frenotomy or frenulotomy. Frenuloplasty, more technically involved than frenotomy or frenulotomy, generally refers to rearranging tissue or adding grafts

after making incisions and closing the resultant wound in a specific pattern to lengthen the anterior tongue. Frenuloplasty is most commonly performed under a general anesthetic and used in older infants and children or in more complex frenulum repairs.

Nonsurgical approaches include speech therapy, lactation interventions, and observation to determine if intervention is warranted.

Scope and Key Questions

Scope of the Review

This systematic review provides a review of potential benefits of treatments (surgical and nonsurgical) as well as harms associated with those therapies in individuals with ankyloglossia and tight labial frenulum (lip-tie) concomitant with ankyloglossia. We sought information on outcomes related to breast- and bottle-feeding and related to tongue-tie in later life (e.g., orthodontic and dental issues, speech, self-esteem).

Key Questions

We synthesized evidence in the published literature to address the following Key Questions (KQs):

KQ 1. What are the benefits of various treatments in breastfeeding newborns and infants with ankyloglossia intended to improve breastfeeding outcomes? Surgical treatments include frenotomy (anterior and/or posterior), frenuloplasty (transverse to vertical frenuloplasty), laser frenulectomy/frenulotomy, and Z-plasty repair. Nonsurgical treatments include complementary and alternative medicine therapies (e.g., craniosacral therapy), lactation intervention, physical/occupational therapy, oral motor therapy, and stretching exercises/therapy.

KQ 2a. 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 bottle-feeding, spilling and dribbling, difficulty moving food boluses in the mouth, and deglutition?

KQ 2b. What are the benefits of various treatments in infants and children with ankyloglossia intended to prevent, mitigate, or remedy attributable other medium- and long-term 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?

KQ 3. What are the benefits of various treatments for ankyloglossia in children through 18 years of age intended to prevent or address social concerns related to tongue mobility (i.e., speech, oral hygiene, excessive salivation, kissing, spitting while talking, and self-esteem)?

KQ 4. What are the benefits of simultaneously treating ankyloglossia and concomitant tight labial frenulum (lip-tie) in infants and children through age 18 intended to improve or remedy breastfeeding, articulation, orthodontic and dental, and other feeding outcomes? What are the relative benefits of treating only ankyloglossia when tight labial frenulum (lip-tie) is also diagnosed?

KQ 5. What are the harms of treatments for ankyloglossia or ankyloglossia with concomitant lip-tie in neonates, infants, and children through age 18?

Analytic Framework

Figure A depicts KQs 1, 4, and 5 within the context of the PICOTS (population, intervention, comparator, outcomes, timing, setting). The figure examines surgical and nonsurgical treatments in neonates and infants to improve breastfeeding outcomes. Intermediate outcomes include maternal nipple pain, ability to latch and maintain latch, tongue mobility, and aerophagia. Final outcomes include duration of breastfeeding, failure to thrive, infant weight gain, and oral and oropharyngeal dysphagia. Harms (KQ 5) may occur at any point after the intervention is received.

Figure A. Analytic framework for ankyloglossia in neonates and infants

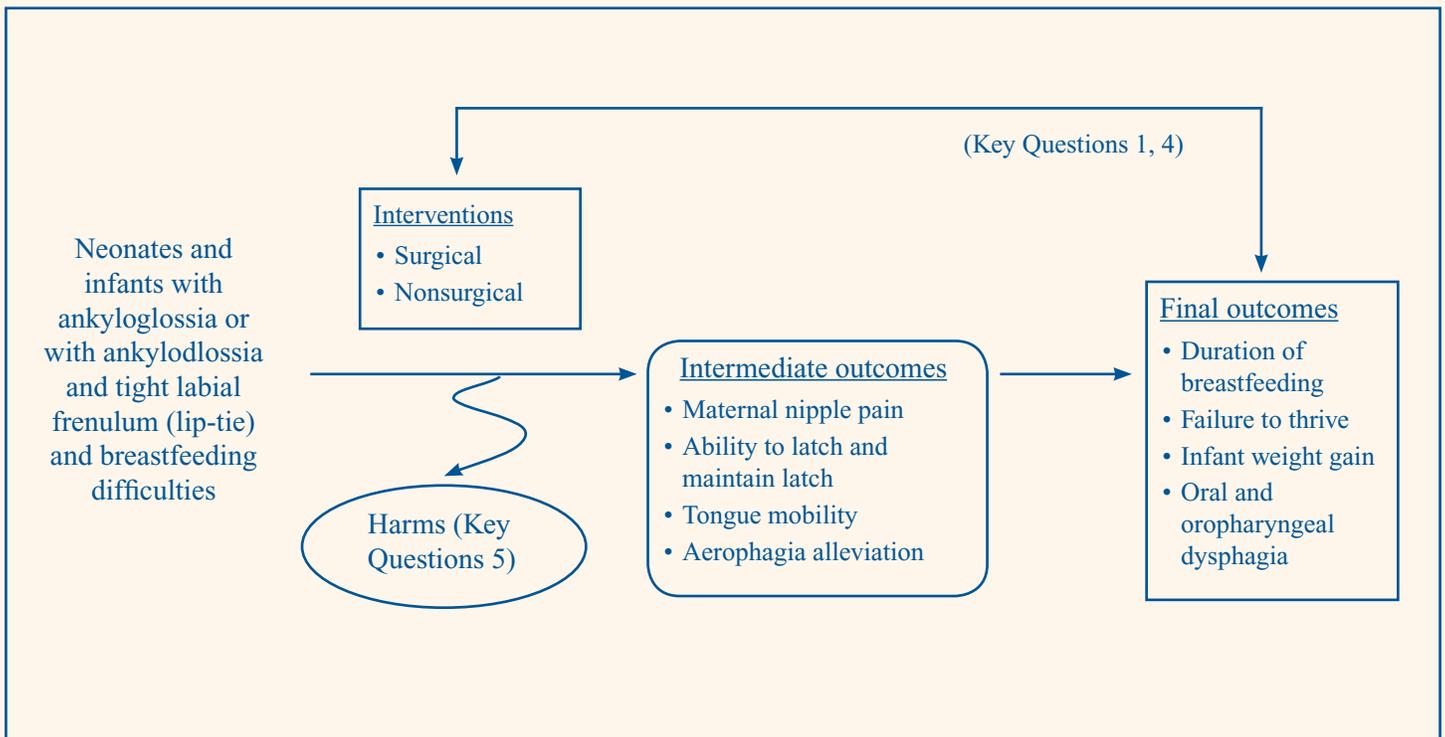
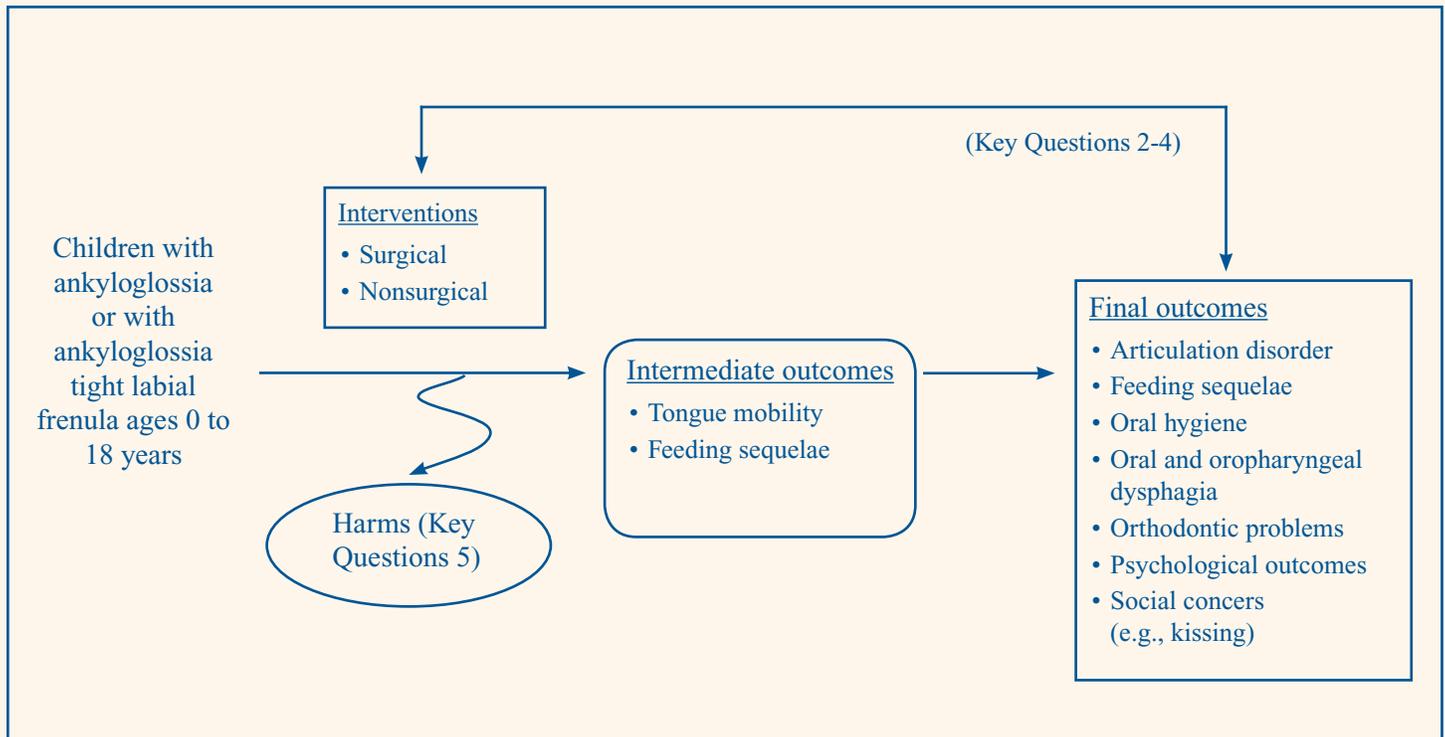


Figure B depicts KQs 2, 3, 4, and 5 within the context of the PICOTS. The figure examines surgical and nonsurgical treatments in infants and children with ankyloglossia (KQ 2, KQ 3) or ankyloglossia with concomitant lip-tie (KQ 4). The intermediate outcomes include maternal nipple

pain and tongue mobility, and final health outcomes are articulation disorder, oral hygiene, oral and oropharyngeal dysphagia, orthodontic problems, psychological outcomes, and social concerns, including kissing. Harms (KQ 5) may occur at any point after the intervention is received.

Figure B. Analytic framework for ankyloglossia in infants and children through 18 years of age



Methods

Literature Search Strategy

A librarian employed search strategies provided in Appendix A of the full report to retrieve research on interventions for children with ankyloglossia. We searched MEDLINE® via the PubMed® interface, PsycINFO® (psychology and psychiatry literature), the Cumulative Index of Nursing and Allied Health Literature (CINAHL®) and Embase (Excerpta Medica Database). We limited

searches to the English language and imposed no publication date restrictions. Our last search was conducted in August 2014. We manually searched reference lists of included studies and of recent narrative and systematic reviews and meta-analyses.

Inclusion and Exclusion Criteria

We developed criteria for inclusion and exclusion in consultation with a Technical Expert Panel (Table A).

Table A. Inclusion criteria

Category	Criteria
Study population	Children ages 0–18 with ankyloglossia or ankyloglossia with concomitant tight labial frenulum (lip-tie); studies with participants with Van der Woude syndrome, Pierre Robin syndrome or sequence, Down syndrome, or craniofacial abnormalities were excluded ,as were studies of premature babies (<37 weeks of gestation ⁵)
Publication languages	English only
Admissible evidence (study design and other criteria)	<p><u>Admissible designs</u></p> <p>Randomized controlled trials, prospective and retrospective cohort studies, nonrandomized controlled trials, prospective and retrospective case series, and crossover studies</p> <p>Case reports to assess harms</p> <p><u>Other criteria</u></p> <p>Original research studies providing sufficient detail regarding methods and results to enable use and aggregation of the data and results</p> <p>Studies must address one or more of the following:</p> <ul style="list-style-type: none"> • Surgical interventions (simple anterior frenotomy, frenulotomy, or frenectomy; laser frenotomy, , or frenulectomy; posterior frenulectomy; Z-plasty repair) • Nonsurgical treatments, including complementary and alternative medicine therapies (e.g., 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 <p>Relevant outcomes must be able to be extracted from data in the papers</p> <p>Data must be presented in the aggregate (vs. individual-participant data)</p>

Study Selection

Two reviewers independently assessed each abstract. If one reviewer concluded that the article could be eligible based on the abstract, we retained it for full-text assessment. Two reviewers independently assessed the full text of each included study. Disagreements were resolved by a senior reviewer.

Data Extraction and Synthesis

We extracted data from included studies into an evidence table that reports study design, descriptions of the study populations (for applicability), description of the intervention, and baseline and outcome data on constructs of interest. Data were initially extracted by one team member and reviewed for accuracy by a second. The final evidence table is presented in Appendix D of the full report.

We extracted outcomes for all included studies, and data are presented in summary tables and analyzed qualitatively in the text.

Quality (Risk-of-Bias) Assessment of Individual Studies

We used four tools to assess the quality of individual studies: the Cochrane Risk of Bias Tool for Randomized Controlled Trials;⁶ a cohort study assessment instrument based on questions and a tool for case series, both adapted from RTI Item Bank questions;⁷ and a four-item harms assessment instrument for cohort studies derived from the McMaster Quality Assessment Scale of Harms (McHarm) for Harms Outcomes⁸ and the RTI Item Bank.⁷ The tools are presented in Appendix E of the full report.

Quality assessment of each study was conducted by two team members independently. Discrepancies were adjudicated through discussion between the assessors to reach consensus or via a senior reviewer. The results of these tools were then translated to the Agency for Healthcare Research and Quality standard of “good,” “fair,” and “poor” quality designations, as described in the full report. Quality ratings for each study are in Appendix F of the full report.

Strength of the Body of Evidence

Two senior investigators graded the entire body of evidence using methods based on the “Methods Guide for Effectiveness and Comparative Effectiveness Reviews.”⁹ The team reviewed the final strength-of-evidence designation. Strength of evidence is assessed for a limited set of critical outcomes, typically those related to effectiveness of an intervention, and reported in comparative studies.

The possible grades were—

- **High:** High confidence that the evidence reflects the true effect. Further research is unlikely to change estimates.
- **Moderate:** Moderate confidence that the evidence reflects the true effect. Further research may change our confidence in the estimate of effect and may change the estimate.
- **Low:** Low confidence that the evidence reflects the true effect. Further research is likely to change confidence in the estimate of effect and is also likely to change the estimate.
- **Insufficient:** Evidence is either unavailable or does not permit a conclusion.

Applicability

Applicability describes issues related to how applicable (generalizable) the included studies are likely to be in practice. We assessed applicability by identifying potential population, intervention, comparator, outcome, and setting (PICOS) factors likely to affect the generalizability of results (i.e., applicability to the general population of children with ankyloglossia). For this particular review, the most likely factors that could affect applicability are the severity/degree of ankyloglossia, age range of participants, setting of intervention (e.g., newborn nursery, outpatient office), and provider (e.g., otolaryngologist, lactation consultant, dentist, pediatrician).

Results

Article Selection

We identified 1,626 nonduplicative titles or abstracts with potential relevance, with 244 proceeding to full-text review (Figure 3 of the full report). We excluded 187 studies at full-text review, which yielded 57 published studies included in the review. We also included one unpublished thesis in our results; thus, the report summarizes data from 58 unique publications.

KQ 1. Benefits of Interventions To Improve Breastfeeding Outcomes

Twenty-nine studies addressed the benefits of surgical treatments intended to improve breastfeeding outcomes; there were no studies of nonsurgical treatments. These studies included five randomized controlled trials (RCTs) conducted in the United Kingdom (n = 3),¹⁰⁻¹² United States (n = 1),¹³ or Israel (n = 1)¹⁴ and one poor-quality retrospective cohort study conducted in the United States.¹⁵ We rated the RCTs as good,^{10,11,13} fair,¹² and poor¹⁴ quality for outcomes related to breastfeeding effectiveness and maternal pain related to breastfeeding. One poor-quality retrospective cohort study and 23 case series also addressed outcomes of surgical treatment. We focused on RCTs of higher quality in this summary but noted that the lower quality studies typically reported improvements in breastfeeding effectiveness.

Two RCTs compared frenotomy to sham surgery,^{11, 13} one to usual care,¹⁰ and one to intensive lactation consultation,¹² and one used a crossover design to compare frenotomy followed by sham surgery to sham surgery followed by frenotomy, with assessment of breastfeeding after each order of intervention (i.e., frenotomy and sham).¹⁴ Similarly, the retrospective cohort study compared frenotomy to usual care.¹⁵ For all studies, sham comparison involved taking infants to an intervention room for the same amount of time as the infants receiving the procedure and then returning them to the mothers.

The earliest reported RCT used nonblinded maternally assessed breastfeeding effectiveness and reported that 96 percent of frenotomized infants had improved feeding within 48 hours, compared with 3 percent in the control group, but this study had significant limitations.¹² In a later RCT, mothers again self-reported improved breastfeeding among infants immediately after frenotomy (78% in the treated group vs. 47% in the comparison group; $p < 0.02$).¹¹

Three RCTs used an observer to assess breastfeeding effectiveness. In all three, the observer was blinded to the treatment. Among these,^{10,11,13} one reported improvement in breastfeeding effectiveness based on the Infant Breastfeeding Assessment Tool (IBFAT; score range, 0 [poor feeding] to 12 [vigorous and effective feeding]) score immediately postfrenotomy compared with sham treatment (mean, 11.6 ± 0.81 vs. 8.07 ± 0.86 ; $p = 0.026$).¹³ In contrast, in two of the three RCTs, the independent blinded observers did not detect a difference in breastfeeding improvement. Outcomes that failed to show a difference in these two RCTs included percent improvement (50% vs. 40%) immediately after intervention¹¹ and Latch, Audible

swallowing, Type of nipple, Comfort, Hold (LATCH) and IBFAT change 5 days postintervention: LATCH change score median 1 (interquartile range [IQR], 0 to 2) versus median 1 (IQR, 0 to 2); $p = 0.52$ and IBFAT change score 0 (IQR, -1.8 to 1.0) versus 0 (IQR 0 to 1); $p = 0.36$.¹⁰

One RCT reported significant and immediate improvement in maternally reported nipple pain among frenotomized infants compared with sham treatment.¹³ Both remaining RCTs found nonsignificant reductions in maternally reported nipple pain between the frenotomy and sham groups at immediate¹¹ and 5-day¹⁰ postprocedure assessments. However, in the one study that assessed pain at 5 days (the longest followup), a large number of infants in the control group had crossed over to receive frenotomy before outcomes were assessed.¹⁰

Harms were rare and nonsignificant, and are discussed in more detail in KQ 5.

KQ 2a. Benefits of Treatments To Mitigate Feeding Sequelae

Three studies examined medium- and long-term benefits related to feeding outcomes and sequelae of various interventions for infants and children with ankyloglossia.^{12,16,17} One was an RCT¹² (fair quality for feeding outcomes) and one was a poor-quality retrospective cohort study;¹⁶ the remaining study was a case series, so it provided no data for comparison.¹⁷

In one RCT that included bottle-fed infants, 76 percent had major problems with dribbling and 71 percent had “excess wind” (gas). Mothers reported significant improvement in bottle-feeding in all eight infants who received the frenotomy and in none of the nine who did not. The interval to ascertainment of the outcomes was not specifically reported, but outcomes were obtained within the first 4 weeks of life.¹²

The retrospective cohort study compared parent-reported (typically maternal) outcomes at age 3 years for three groups of children born in 2010: children who received frenotomy for tongue-tie ($n = 71$; frenotomy group); those whose parents were offered frenotomy for tongue-tie for their children but declined it ($n = 15$; no-frenotomy group); and children without ankyloglossia ($n = 18$; control group).¹⁶ The frenotomy group performed better than the no-frenotomy group at age 3 years on cleaning the teeth with the tongue, licking the outside of the lips, and eating ice cream, and did not differ significantly from the comparison group without ankyloglossia.

KQ 2b. Benefits of Treatments To Prevent Other Sequelae

Two cohort studies attempted to assess the effectiveness of frenotomy for preventing other sequelae,^{16,18} and one RCT compared two surgical approaches to frenotomy.¹⁹ A speech-language pathologist measured speech outcomes in two studies,^{18,19} with the third study using parental assessment.¹⁶ No studies included data related to sleep-disordered breathing, occlusal issues, and dysphagia in nonbreastfeeding children.

Two poor-quality cohort studies^{16,18} reported an improvement in articulation and intelligibility with ankyloglossia treatment, but benefits in word and sentence accuracy and intelligibility and fluent speech were unclear. The one poor-quality RCT comparing surgical methods reported improved articulation in patients treated with four-flap Z-frenuloplasty compared with horizontal-to-vertical frenuloplasty.¹⁹ Numerous noncomparative studies²⁰⁻²⁶ reported a speech benefit after treating ankyloglossia; however, these studies primarily discussed modalities, with safety, feasibility, or utility as the main outcome rather than speech itself, and they provided no comparative data.

KQ 3. Benefits of Treatments To Prevent Social Concerns Related to Tongue Mobility

Only one poor-quality retrospective cohort study assessed outcomes related to social concerns other than speech in 3-year-old children who had received frenotomy as infants.¹⁶ The group that had received frenotomy had better parent-reported ability to clean teeth with tongue, lick outside of lips, and eat ice cream compared with untreated participants.

KQ 4. Benefits of Simultaneously Treating Ankyloglossia and Lip-Tie

We did not identify any studies addressing this question.

KQ 5. Harms of Treatments for Ankyloglossia or Ankyloglossia With Concomitant Lip-Tie in Neonates, Infants, and Children Through Age 18

In order to identify all possible harms, we sought harms from all comparative studies and case series that we identified as potentially providing effectiveness data, and we sought case reports of harms. With this approach, we examined harms information from 46 studies that reported that they had looked for harms, either reporting actual harms or specifically indicating that they found none. These included 6 RCTs, 1 cohort study, 25 case series,

and 15 case reports. Most studies that reported harms information explicitly noted that no significant harms were observed ($n = 17$) or reported minimal harms. Among studies reporting harms, bleeding was most frequently reported. Bleeding was typically described as minor and limited. Reoperation was noted in seven studies. Few studies described the specific methods they used to collect harms data.

Discussion

Key Findings

Most of the studies included in this review addressed outcomes related to breastfeeding. Overall, three good-quality^{10,11,13} and one fair-quality¹² RCT assessed whether surgical treatment of ankyloglossia improved breastfeeding effectiveness. Maternally reported breastfeeding effectiveness was significantly improved in the treated group compared with the untreated group in both RCTs that evaluated it either as a primary¹² or secondary¹¹ outcome. Only one of three RCTs that used blinded independent observers found significantly improved breastfeeding effectiveness among frenotomized infants immediately postprocedure.¹³ A third RCT evaluated the mother's breastfeeding self-efficacy and found a significant improvement from baseline in the frenotomy group 5 days postprocedure.¹⁰ In all, some evidence suggests that maternally reported breastfeeding outcomes improved, but data are unavailable to assess the durability of effects.

These same studies had disparate findings about whether frenotomy decreased maternal nipple pain during breastfeeding. Only the RCT performed on infants at 6 days of age showed a significant reduction in maternal pain.¹³ Those performed on infants a few weeks older did not report either an immediate¹¹ or 5-day¹⁰ reduction in pain. The difference between earlier frenotomy and later frenotomy on nipple pain may relate to cumulative trauma on the breast from several additional weeks with inefficient latch from tongue-tied infants.

We identified three studies examining feeding outcomes other than breastfeeding: one RCT,¹² one-poor quality retrospective cohort study,¹⁶ and one case series.¹⁷ Bottle-feeding and ability to use the tongue to eat ice cream and clean the mouth improved more in treatment groups in comparative studies. Bottle feedings to supplement breast feeding decreased over time in the case series.

Following breastfeeding outcomes, outcomes related to speech were most often reported in the ankyloglossia literature. Two poor-quality cohort studies^{16,18} reported

an improvement in articulation and intelligibility with ankyloglossia treatment, but benefits in word and sentence accuracy and intelligibility and fluent speech were unclear. One poor-quality RCT reported improved articulation in patients treated with Z-frenuloplasty compared with horizontal-to-vertical frenuloplasty.¹⁹ Numerous noncomparative studies reported a speech benefit after treating ankyloglossia; however, these studies primarily discussed modalities, with safety, feasibility, or utility as the main outcome, rather than speech itself.^{23,26-28}

Few studies addressed social concerns. One retrospective cohort study noted improvements in using the tongue to clean the teeth and for licking in the treatment group compared with untreated participants.¹⁶ In two comparative studies reporting on tongue mobility, mobility improved in treated patients.^{18,19}

Harms of surgical interventions included minor bleeding, which was typically self-limiting, and need for reoperation, which was rare. Minor bleeding is not an unexpected occurrence in this type of surgical intervention. Eighteen studies reported that no significant harms were observed.

Strength of Evidence

Breastfeeding Outcomes

Very few higher quality comparative studies have addressed the effectiveness of surgical interventions to improve breastfeeding outcomes. In those few studies, mothers consistently reported improved breastfeeding effectiveness, but outcome measures were heterogeneous and very short term. Future studies could provide additional data to confirm or change the measure of effectiveness; thus, we consider the strength of evidence to be low at this time. We considered the strength of evidence (confidence in the estimate of effect) to be low for an immediate reduction in nipple pain. Improvements were reported in the current studies, but additional studies are needed to confirm and support these results. Only one poor-quality cohort study addressed effects on the length of breastfeeding; thus, we considered the strength of evidence to be insufficient.

Other Feeding Outcomes

With only two comparative studies, both with significant study limitations, existing data are insufficient to draw conclusions about the benefits and harms of surgical interventions for infants and children with ankyloglossia on medium- and long-term feeding outcomes. The studies used different populations and measured different outcomes.

Speech Outcomes

Given the lack of good-quality studies and limitations in the measurement of outcomes, we considered the strength of the evidence for the effect of surgical interventions to improve speech and articulation to be insufficient.

Social Concerns Related to Tongue Mobility

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

Harms

We considered the strength of evidence for minimal and short-lived bleeding as a minor harm of surgical interventions as moderate based on an expanded search for harms reports in addition to the comparative data. We considered the strength of evidence for reoperation and pain as harms to be insufficient, given the small number of outcomes available for analysis. We acknowledge that harms are not systematically reported, and thus there may be substantial underreporting.

Table B. Strength of evidence for studies addressing surgical approaches for ankyloglossia

Outcome; Number of Studies and Quality (Total Participants)	Study Limitations	Consistency	Directness	Precision	Reporting Bias	Finding/Strength of Evidence
Breastfeeding Outcomes						
<i>Nipple pain</i> RCT: 3 good, ^{10,11,13} 1 poor ¹⁴ (251) Retrospective cohort: 1 poor ¹⁵ (367)	Low	Inconsistent	Direct	Imprecise	Undetected	Low SOE for an immediate reduction in nipple pain postprocedure due to inconsistent results across small studies.
<i>Breastfeeding effectiveness</i> RCTs: LATCH—2 good, ^{10,11} 1 poor ¹⁴ (193) IBFAT—1 good ¹³ (58) BSES-SF—1 fair ¹⁰ (107) Retrospective cohort: 1 poor ¹⁵ (367)	Low	Inconsistent	Direct	Imprecise	Undetected	Low SOE for improved breastfeeding. Mothers consistently reported improved breastfeeding effectiveness, but outcome measures were heterogeneous and very short term. Observer-rated measures did not show significant improvements. Future studies could provide additional data to confirm or change the measure of effectiveness.
<i>Length of breastfeeding</i> Retrospective cohort: 1 poor ¹⁵ (367)	High	NA	Direct	Imprecise	Undetected	Insufficient SOE due to the high risk of bias of the 1 retrospective study

Table B. Strength of evidence for studies addressing surgical approaches for ankyloglossia (continued)

Outcome; Number of Studies and Quality (Total Participants)	Study Limitations	Consistency	Directness	Precision	Reporting Bias	Finding/Strength of Evidence
Other Feeding Outcomes						
<i>Feeding outcomes</i> RCT: 1 poor ¹² (57) Retrospective cohort: 1 poor ¹⁶ (104)	High	Consistent	Indirect	Imprecise	Undetected	Insufficient SOE for all feeding outcomes, given small number of participants, lack of standard outcome measures, and poor quality of studies.
Speech Outcome						
<i>Speech and articulation</i> Retrospective cohort: 1 poor ¹⁶ (104) Prospective cohort: 1 poor ¹⁸ (23)	High	Inconsistent	Indirect	Imprecise	Undetected	Insufficient SOE based on 2 poor-quality cohort studies.
<i>Oral motor skills</i> Retrospective cohort: 1 poor ¹⁶ (104) Prospective cohort: 1 poor ¹⁸ (23)	High	Consistent	Indirect	Imprecise	Undetected	Insufficient SOE based on 2 poor-quality cohort studies.
Social Outcomes						
<i>Social concerns</i> Retrospective cohort: 1 poor ¹⁶ (104)	High	NA	Indirect	Imprecise	Undetected	Insufficient SOE based on 1 poor-quality cohort study.
<i>Tongue mobility</i> RCT: 1 poor ¹⁹ (16) Retrospective cohort: 1 poor ¹⁸ (15)	High	Consistent	Direct	Imprecise	Undetected	Insufficient SOE based on 2 small poor-quality studies.
Harms						
<i>Bleeding</i> RCT: 1 poor ¹¹ (60) Case series: 14 poor ^{17,22,25,28-38} , 2 good ^{27,39} (963)	High	Consistent	Direct	Imprecise	Suspected	Moderate SOE for minimal and short-lived bleeding based on an extensive search for harms reports in addition to the comparative data. Studies consistently reported minimal to no bleeding.

Table B. Strength of evidence for studies addressing surgical approaches for ankyloglossia (continued)

Outcome; Number of Studies and Quality (Total Participants)	Study Limitations	Consistency	Directness	Precision	Reporting Bias	Finding/Strength of Evidence
Reoperation RCT: 1 poor ¹⁰ (107) Retrospective cohort: 1 poor ¹⁵ (367) Case series: 1 good, ³⁹ 4 poor ^{23,24,40,41} (4,080)	High	Consistent	Direct	Imprecise	Suspected	Insufficient SOE due to very small numbers for the outcome.
Pain Case series: 2 good ^{27,42} (84)	High	Consistent	Indirect	Imprecise	Suspected	Insufficient SOE for minimal short-lived pain in infants. No studies reported excessive crying or an inability to feed soon after the intervention, but pain is arguably difficult to assess in infants, so outcomes were indirect and from poor-quality or noncomparative studies.

BSES –SF = Breastfeeding Self-Efficacy Scale-Short Form; IBFAT = Infant Breastfeeding Assessment Tool; LATCH = Latch, Audible swallowing, Type of nipple, Comfort, Hold; NA = not applicable; RCT = randomized controlled trial; SOE = strength of evidence.

Applicability

Newborns referred for treatment of ankyloglossia were born primarily at tertiary-care centers and recognized as having difficulty with breastfeeding concomitant with ankyloglossia. The frenotomy procedure itself is not technically difficult and is likely performed similarly across birthing sites; however, the criteria by which the decision is made to perform frenotomy are less clear. Moreover, newborns of mothers not choosing to breastfeed may not be recognized as having and/or diagnosed with ankyloglossia, as breastfeeding difficulties were used as an indicator to evaluate for ankyloglossia. At minimum, the studies in this report apply only to infants with both ankyloglossia and feeding difficulties; data on ankyloglossia absent feeding difficulties were unavailable.

In these studies, various clinicians were involved in making the ankyloglossia diagnoses. However, assessment of breastfeeding difficulty and diagnostic criteria for ankyloglossia were not universally described. Lack of

a consistent objective measure to define and classify this condition may limit the reproducibility of findings. Furthermore, the age of patients in these studies varied from a median of 6 days of age in one study¹³ up to a mean of 33 days of age (range, 6 to 115) in another study.¹¹ Applicability of findings to older infants cannot be gleaned from these data, nor can durability of results.

Frenotomy was the only intervention employed in the good-quality RCTs.^{10,11,13} However, the specifics of the procedure were variably reported. The degree of posterior extension of the frenulum incision was not clearly defined and appears to be at the discretion and clinical expertise of the clinician. Also, the severity of the ankyloglossia was inconsistently reported, making interstudy generalizations difficult and, more importantly, limiting the broader applicability of findings.

The comparators used were sham surgery^{11,13} and no intervention.¹⁰ Both “no intervention” and “sham surgery” are perhaps misnomers, however, since these infant-mother

dyads underwent usual care, which could include, but is not limited to, lactation consultation, supportive care, and bottle-feeding advice.

The population studied in the question of benefit of ankyloglossia repair for social concerns included children and adults with wide variation in ages.

Research Gaps

A critical unknown at this point is a good description of the natural history of ankyloglossia by severity, including long-term risk of feeding, social, and speech production difficulties. Future studies should consider direct comparisons of alternative treatments, as currently available literature addressed only the comparison of frenotomy with sham. In order to conduct these studies, it would be helpful if the field could agree on a standardized approach to identifying and classifying ankyloglossia; this would also improve our ability to synthesize the data across studies.

Given variation in outcomes that may be associated with earlier versus later frenotomy, future studies should assess timing of frenotomy to determine whether more significant reduction in maternal pain is achievable by earlier treatment and whether mothers are more apt to breastfeed longer if the frenotomy is done earlier.

A significant gap in research is in understanding the durability of outcomes. Good-quality comparative studies evaluated breastfeeding effectiveness immediately^{11,13} or within 5 days of frenotomy;¹⁰ however, none adequately assessed whether effectiveness and other outcomes (e.g., changes in maternal nipple pain) were maintained months or, if appropriate, years later. Longer term followup of both treated infants and controls is needed. Because of the paucity of available data on other feeding outcomes, this entire research question 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 speech production difficulties, as well as whether treatment later in life with frenotomy leads to improvement when speech problems arise. To conduct this research effectively, methods for evaluating risk and presence of speech production difficulties will need to be standardized, and outcomes agreed on. Understanding of the natural history of speech concerns in children with ankyloglossia is lacking, as are comparative studies that use standardized measurement tools for speech outcomes.

No standard definitions of tongue mobility or established norms for mobility exist, and further research is needed to determine such parameters. Social concerns are difficult to measure objectively, so there will likely always be a subjective component to social 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

A small body of evidence suggests that frenotomy may be associated with improvements in breastfeeding as reported by mothers, and potentially in nipple pain. However, with small, inconsistently conducted studies, strength of evidence is low to insufficient, preventing us from drawing firm conclusions at this time. Research is lacking on nonsurgical interventions, as well as on outcomes other than breastfeeding, particularly speech and dental outcomes. In particular, there is a lack of evidence on significant long-term outcomes, such as exclusive breastfeeding at 6 months of age or at 1 year of age, growth, and other measures of health outcomes. Harms are minimal and rare; the most commonly reported harm is self-limited bleeding. Future research is needed on a range of issues, including prevalence and incidence of ankyloglossia and problems with the condition. The field is currently challenged by a lack of standardized approaches to assessing and studying the problems of infants with ankyloglossia.

References

1. Lalakea ML, Messner AH. Ankyloglossia: does it matter? *Pediatr Clin North Am.* 2003 Apr;50(2):381-97. PMID: 12809329.
2. Suter VG, Bornstein MM. Ankyloglossia: facts and myths in diagnosis and treatment. *J Periodontol.* 2009 Aug;80(8):1204-19. PMID: 19656020.
3. Messner AH, Lalakea ML. Ankyloglossia: controversies in management. *Int J Pediatr Otorhinolaryngol.* 2000 Aug 31;54(2-3):123-31. PMID: 10967382.
4. UK National Health Service. National Institute for Health and Clinical Excellence. Division of ankyloglossia (tongue-tie) for breastfeeding. NICE Interventional Procedure Guidance 149. December 2005. www.nice.org.uk/guidance/ipg149/resources/guidance-division-of-ankyloglossia-tonguetie-for-breastfeeding-pdf. Accessed March 23, 2015.
5. Rowan-Legg A. Canadian Paediatric Society Position Statement: Ankyloglossia and Breastfeeding. Canadian Paediatric Society, 2014. www.cps.ca/en/documents/position/ankyloglossia-breastfeeding. Accessed March 23, 2015.

6. Higgins JPT, Green S, eds. *Cochrane Handbook for Systematic Reviews of Interventions*. The Cochrane Collaboration; 2011. www.cochrane-handbook.org. Accessed December 17, 2014.
7. Viswanathan M, Berkman ND, Dryden DM. *Assessing Risk of Bias and Confounding in Observational Studies of Interventions or Exposures: Further Development of the RTI Item Bank*. Methods Research Report. AHRQ Publication No. 13-EHC106-EF. Rockville, MD: Agency for Healthcare Research and Quality; 2013. www.ncbi.nlm.nih.gov/books/NBK154461/. Accessed December 17, 2014.
8. Santaguidda P, Raina P. *McMaster Quality Assessment Scale of Harms (McHarm) for Primary Studies: Manual for Use of the MCHarm*. Hamilton, ON: McMaster University.
9. *Methods Guide for Effectiveness and Comparative Effectiveness Reviews*. AHRQ Publication No. 10(14)-EHC063-EF. Rockville, MD: Agency for Healthcare Research and Quality; January 2014. Chapters available at www.effectivehealthcare.ahrq.gov. Accessed December 17, 2014
10. Emond A, Ingram J, Johnson D, et al. Randomised controlled trial of early frenotomy in breastfed infants with mild-moderate tongue-tie. *Arch Dis Child Fetal Neonatal Ed*. 2013 Nov 18;99(3):F189-95. PMID: 24249695.
11. Berry J, Griffiths M, Westcott C. A double-blind, randomized, controlled trial of tongue-tie division and its immediate effect on breastfeeding. *Breastfeed Med*. 2012 Jun;7(3):189-93. PMID: 21999476.
12. Hogan M, Westcott C, Griffiths M. Randomized, controlled trial of division of tongue-tie in infants with feeding problems. *J Paediatr Child Health*. 2005 May-Jun;41(5-6):246-50. PMID: 15953322.
13. Buryk M, Bloom D, Shope T. Efficacy of neonatal release of ankyloglossia: a randomized trial. *Pediatrics*. 2011 Aug;128(2):280-8. PMID: 21768318.
14. Dollberg S, Botzer E, Grunis E, et al. Immediate nipple pain relief after frenotomy in breast-fed infants with ankyloglossia: a randomized, prospective study. *J Pediatr Surg*. 2006 Sep;41(9):1598-600. PMID: 16952598.
15. Steehler MW, Steehler MK, Harley EH. A retrospective review of frenotomy in neonates and infants with feeding difficulties. *Int J Pediatr Otorhinolaryngol*. 2012 Sep;76(9):1236-40. PMID: 22704670.
16. Walls A, Pierce M, Wang H, et al. Parental perception of speech and tongue mobility in three-year olds after neonatal frenotomy. *Int J Pediatr Otorhinolaryngol*. 2014 Jan;78(1):128-31. PMID: 24315215.
17. Miranda BH, Milroy CJ. A quick snip - a study of the impact of outpatient tongue tie release on neonatal growth and breastfeeding. *J Plast Reconstr Aesthet Surg*. 2010 Sep;63(9):e683-5. PMID: 20493791.
18. Dollberg S, Manor Y, Makai E, et al. Evaluation of speech intelligibility in children with tongue-tie. *Acta Paediatr*. 2011 Sep;100(9):e125-7. PMID: 21401716.
19. Heller J, Gabbay J, O'Hara C, et al. Improved ankyloglossia correction with four-flap Z-frenuloplasty. *Ann Plast Surg*. 2005 Jun;54(6):623-8. PMID: 15900148.
20. Marchesan IQ, Martinelli RL, Gusmao RJ. Lingual frenulum: changes after frenectomy. *J Soc Bras Fonoaudiol*. 2012;24(4):409-12. PMID: 23306695.
21. Glynn RW, Colreavy M, Rowley H, et al. Division of tongue tie: review of practice through a tertiary paediatric otorhinolaryngology service. *Int J Pediatr Otorhinolaryngol*. 2012 Oct;76(10):1434-6. PMID: 22810118.
22. Choi YS, Lim JS, Han KT, et al. Ankyloglossia correction: Z-plasty combined with genioglossus myotomy. *J Craniofac Surg*. 2011 Nov;22(6):2238-40. PMID: 22134257.
23. Klockars T, Pitkaranta A. Pediatric tongue-tie division: indications, techniques and patient satisfaction. *Int J Pediatr Otorhinolaryngol*. 2009 Oct;73(10):1399-401. PMID: 19660817.
24. Yeh ML. Outpatient division of tongue-tie without anesthesia in infants and children. *World J Pediatr*. 2008 May;4(2):106-8. PMID: 18661764.
25. Lalakea ML, Messner AH. Ankyloglossia: the adolescent and adult perspective. *Otolaryngol Head Neck Surg*. 2003 May;128(5):746-52. PMID: 12748571.
26. Dave J, Sinha V, Barot D, et al. Speech disorders encountered in routine ENT practice and the role of speech therapy in its effective management. *Indian J Otolaryngol*. 2013 Oct-Dec;19(4):169-72.
27. Puthussery FJ, Shekar K, Gulati A, et al. Use of carbon dioxide laser in lingual frenectomy. *Br J Oral Maxillofac Surg*. 2011 Oct;49(7):580-1. PMID: 20728254.
28. Messner AH, Lalakea ML. The effect of ankyloglossia on speech in children. *Otolaryngol Head Neck Surg*. 2002 Dec;127(6):539-45. PMID: 12501105.
29. Srinivasan A, Dobrich C, Mitnick H, et al. Ankyloglossia in breastfeeding infants: the effect of frenotomy on maternal nipple pain and latch. *Breastfeed Med*. 2006 Winter;1(4):216-24. PMID: 17661602.
30. Blenkinsop A. A measure of success: audit of frenulotomy for infant feeding: problems associated with tongue-tie. *MIDIRS Midwifery Digest*. 2003;13(3):389-92.
31. Ridgers I, McCombe K, McCombe A. A tongue-tie clinic and service. *Br J Midwifery*. 2009;17(4):230-3.
32. Sethi N, Smith D, Kortequee S, et al. Benefits of frenulotomy in infants with ankyloglossia. *Int J Pediatr Otorhinolaryngol*. 2013 May;77(5):762-5. PMID: 23453795.
33. Geddes DT, Langton DB, Gollow I, et al. Frenulotomy for breastfeeding infants with ankyloglossia: effect on milk removal and sucking mechanism as imaged by ultrasound. *Pediatrics*. 2008 Jul;122(1):e188-94. PMID: 18573859.
34. Wallace H, Clarke S. Tongue tie division in infants with breast feeding difficulties. *Int J Pediatr Otorhinolaryngol*. 2006 Jul;70(7):1257-61. PMID: 16527363.

35. Amir LH, James JP, Beatty J. Review of tongue-tie release at a tertiary maternity hospital. *J Paediatr Child Health*. 2005 May-Jun;41(5-6):243-5. PMID: 15953321.
36. Fiorotti RC, Bertolini MM, Nicola JH, et al. Early lingual frenectomy assisted by CO2 laser helps prevention and treatment of functional alterations caused by ankyloglossia. *Int J Orofacial Myology*. 2004 Nov;30:64-71. PMID: 15832863.
37. Ballard JL, Auer CE, Khoury JC. Ankyloglossia: assessment, incidence, and effect of frenuloplasty on the breastfeeding dyad. *Pediatrics*. 2002 Nov;110(5):e63. PMID: 12415069.
38. Masaitis NS, Kaempf JW. Developing a frenotomy policy at one medical center: a case study approach. *J Hum Lact*. 1996 Sep;12(3):229-32. PMID: 9025430.
39. Argiris K, Vasani S, Wong G, et al. Audit of tongue-tie division in neonates with breastfeeding difficulties: how we do it. *Clin Otolaryngol*. 2011 Jun;36(3):256-60. PMID: 21752209.
40. 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 May;77(5):827-32. PMID: 23523198.
41. Hong P, Lago D, Seargeant J, et al. Defining ankyloglossia: a case series of anterior and posterior tongue ties. *Int J Pediatr Otorhinolaryngol*. 2010 Sep;74(9):1003-6. PMID: 20557951.
42. Mettias B, O'Brien R, Abo Khatwa MM, et al. Division of tongue tie as an outpatient procedure. Technique, efficacy and safety. *Int J Pediatr Otorhinolaryngol*. 2013 Apr;77(4):550-2. PMID: 23411135.

Full Report

This executive summary is part of the following document: Francis DO, Chinnadurai S, Morad A, Epstein RA, Kohanim S, Krishnaswami S, Sathe NA, McPheeters ML. Treatments for Ankyloglossia and Ankyloglossia With Concomitant Lip-Tie. Comparative Effectiveness Review No. 149. (Prepared by the Vanderbilt Evidence-based Practice Center under Contract No. 290-2012-00009-I.) AHRQ Publication No. 15-EHC011-EF. Rockville, MD: Agency for Healthcare Research and Quality; May 2015. www.effectivehealthcare.ahrq.gov/reports/final.cfm.

