Scoping literature review: gastrointestinal signs in infants with ankyloglossia

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Background: Improvement of reported gastrointestinal (GI) signs are used to prove efficacy of ankyloglossia correction. The aim of this scoping review was to summarize GI signs observed in infants with restricted tongue mobility known as ankyloglossia, or tongue tie, through the discussion of relevant quantitative data of associated GI signs. Although common in infancy, reflux is one of the relevant signs discussed in this review.

Methods: PubMed and CINAHL were used for this scoping review that resulted in a total of 132 articles with 17 repeated articles between the two searches. Inclusion criteria were infants from birth to 23 months, written in the English language, and relevant peer-reviewed research articles. Articles were excluded if not original research, research that did not address GI signs, qualitative research, if children were older than 23 months, or if written in another language. All articles from inception to the date of the search on March 31, 2023, were considered. Out of the 115 reviewed articles, seven were included and manually referenced in the final quantitative comparison of infants with ankyloglossia experiencing GI signs.

Results: The search identified four research articles that showed statistically significant decreases in Infant Gastroesophageal Questionnaire Revised version (I-GERQ-R) total scores in infants with ankyloglossia who had undergone surgical release via frenotomy. Vomiting and hematemesis were also found in some infants with ankyloglossia.

Conclusions: The most prevalent GI sign related to ankyloglossia in infants was gastroesophageal reflux. Though common in early life, it is believed that aerophagia due to ankyloglossia may be a possible etiology for reflux in this population. Clinical implication is important in investigating possible differential diagnoses associated with reflux, including ankyloglossia. Further research is needed to better understand the physiologic rationale for GI signs in infants with ankyloglossia, and how these may improve over time following treatment via frenotomy.

Keywords: Ankyloglossia; infant; gastrointestinal signs (GI signs); frenotomy; tongue tie

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Introduction

Ankyloglossia, more commonly known as tongue tie, is a minor congenital anomaly where the lingual frenulum limits the mobility of the tongue by being unusually thick, tight, or short (1,2). It has been debated over multiple research articles on how to classify, define, and assess ankyloglossia. According to the American Academy of Pediatrics (AAP), anterior and posterior ankyloglossia are not well-defined and highly debated amongst experts (2). The AAP encourages providers to focus on the general definition of ankyloglossia such that it creates restrictive tongue mobility (2). In 2019, Mills et al. (3) summarized a comprehensive study of the mechanism of ankyloglossia using the dissection of the lingual frenulum of cadavers. This work better explained ankyloglossia as the dysfunction of the lingual frenulum by restricting the connective tissues that suspend the tongue to the floor of the mouth, causing immobility and instability (3). In 2020, a panel of pediatric otolaryngologists published a definition of ankyloglossia in the Clinical Consensus Statement as “a condition of limited tongue mobility caused by a restrictive lingual frenulum” (4). In this review, ankyloglossia is represented as restricted mobility and stability of the tongue, in line with the definitions above. Frenotomy is a common surgical intervention used to correct restricted tongue mobility in infants that has raised controversy in both safety and effectiveness (5,6). There is a paucity of high-quality research, a lack of evidence-based practice guidelines, and limited longitudinal data on symptom improvement following frenotomy (5). Clinical consensus of the impact of ankyloglossia for infants has not been established beyond agreement that ankyloglossia is one potential cause of breastfeeding challenges (2).

The overall prevalence of ankyloglossia is approximately 8% in infants (7). During infancy, breastfeeding difficulty is one of the most prominent effects of ankyloglossia due to poor latch, poor sucking, or maternal nipple pain (8). Poor latch and impaired sucking have been associated with aerophagia, contributing to the gastrointestinal (GI) signs of reflux in infants with ankyloglossia (9-11). Aerophagia is the increased ingestion of air, postulated to be caused by impaired latch onto the breast and dysregulated tongue movement. The increased ingestion of air can lead to symptoms of reflux, pain, and a distended abdomen (12). However, gastroesophageal reflux (GER) is considered a normal finding in infancy that peaks between the age of 1 to 4 months with the condition typically resolving after 6 months of age (13). It has been reported that breastfed infants are less likely to have reflux, with most cases being benign and not requiring treatment (14). However, infantile reflux can cause significant anxiety for parents, negatively impact quality of life for both parents and the child, and has been linked to development of gastroesophageal reflux disease (GERD) later in life (15).

Beyond infancy, untreated ankyloglossia has been associated with mechanical issues such as feeding and speaking difficulties, decreased ability to clear food from teeth and challenges playing wind instruments (2,6). Identifying signs associated with ankyloglossia in infancy that may necessitate frenotomy may help to prevent these sequelae in later childhood and adulthood.

There has been a significant increase in frenotomy rates over the last three decades (16) with GI signs frequently referenced to support frenotomy (17). Thus, the aim of this scoping review was to summarize GI signs observed in infants with restricted tongue mobility secondary to ankyloglossia through the discussion of relevant quantitative data of associated GI signs. The authors focus on GI signs, defined as objective evidence of the disease observed by others (i.e., healthcare providers and caregivers), since GI symptoms would require subjective report from the infant, which is not possible (18). We present this article in accordance with the PRISMA-ScR reporting checklist (available at https://pm.amegroups.com/article/view/10.21037/pm-23-35/rc).
Methods

Search method

Using PubMed and CINAHL, an advanced search on GI signs and symptoms in infants with ankyloglossia was done using MeSH terms, relevant keywords, and filters for infant age (Figure 1). The search on PubMed, (“Ankyloglossia”[Mesh] OR “tongue tie AND (“Gastroesophageal Reflux”[Mesh] OR “Signs and Symptoms, Digestive”[Mesh] OR “reflux” OR “constipation” OR “gagging” OR “flatulence” OR “aspiration” OR “latch” OR “breastfeed”) with filter applied: “Infant: birth–23 months, Infant: 1–23 months”, resulted in 64 articles. The search on CINAHL, “MH ankyloglossia or tongue tie or tongue-tie” resulted in 68 articles. After 17 duplicates were removed, a total of 115 articles were found.

Search criteria

Inclusion criteria were: written in the English language, peer-reviewed research article, children less than 23 months of age, relevant GI signs, and quantitative data. A preset filter of infant per PubMed and CINAHL determined infancy age to range from birth to 23 months. In addition, due to the limited published work regarding ankyloglossia, time parameter of the research articles was not restricted to increase access to all information on the topic. All articles from inception to the date of the search on March 31, 2023, were considered. Articles that were not original research or were qualitative in nature, research in children over two years of age, and/or articles that did not include evaluation of GI signs were excluded from this review. A total of 115 abstracts were reviewed by both authors. Articles were reviewed in full by the first author, followed by discussion between both authors to resolve discrepancies. The seven articles selected full inclusion were reviewed independently by both authors to retrieve relevant data reported in this scoping review. Both authors worked independently to retrieve data from the reports. The first author used a systematic method for retrieval data from the seven included studies. First, the author extracted the sample size, infant age range, the measurement tool(s) used to evaluate GI signs, and the time frame for assessment using the measurement tool(s). Next, the author retrieved the GI signs study investigators proposed to be associated with ankyloglossia and the study outcome(s). Finally, both authors discussed the data extraction to address and resolve any discrepancies. It was not necessary to obtain additional data from study investigators.
Measurement tools used in research

A summary of each measurement tool used in the included research articles for this scoping review is provided.

Infant Gastroesophageal Questionnaire Revised version (I-GERQ-R)
The I-GERQ-R is a 12-item caregiver-reported questionnaire that was derived from a 138-item I-GERQ that measures signs associated with reflux (20). The purpose of the revised version is for use in clinical trials to determine the effectiveness of an intervention on GER (20). The I-GERQ-R has a score range of 0 to 40, with a score greater or equal to 16 suggesting a diagnosis of GER, provided a thorough history and physical exam supports the diagnosis (20). One study suggested that a change in score of 6 points between time points can be considered clinically significant; however, a difference of three or four has been considered a minimally important difference (MID) (20). Using Cochrane’s Q and I² as a distribution-based method to assess a clinically significant difference or MID in the I-GERQ-R scoring, the authors determined consistency in the change of scores in multiple study designs, suggesting validity and reliability of this tool (20).

Kleinman et al. (21) conducted a study with 278 infant caregivers to assess the validity and reliability of the I-GERQ-R. The authors assessed a comparison and a control group, with report of daily signs and symptoms, correlation to physician-rated severity, and correlation to caregiver-rated severity (21). All comparisons were statistically significant (P<0.05) suggesting validity of the questionnaire (21).

Infant Gastrointestinal Symptoms Questionnaire (GSQ-I)
The GSQ-I is a 13-item caregiver-reported GI-related feeding behaviors questionnaire that assesses feeding tolerance in infants, focusing on digestive and elimination patterns over the last week. Likert scales are used, with scores ranging from 1 (no symptoms) to 5 (severe symptoms). A minimum score of 13 indicates no GI distress with a score of 65 indicating extreme GI distress (22). Riley et al. (22) tested the validity and reliability of GSQ-I through four studies assessing interrater reliability, retest reliability, and validity. To test interrater reliability, parents were interviewed twice on the same day to evaluate their ability to repeat the same answers. They found that there was little to no change in their answers (22). To assess retest reliability, parents took the questionnaire on day 1 and day 9. One limitation in this measure is the GI maturity in infants over time. Parents were asked to follow daily GI symptoms such as using the GSQ-I on Day 0 and Day 10 post-frenotomy. The authors found a statistically significant improvement between Day 0 and Day 10 on GSQ-I mean scores. It is unclear if these scores correlated with clinical significance. Two of the above studies suggest aerophagia as a potential cause of reflux (12,26). Imaging or auscultation

Results

GI signs associated with ankyloglossia

Reflux
Six out of the seven included research articles mention reflux as a GI sign in infants with ankyloglossia. Ghaheri et al. (23) and Hill et al. (27) asked mothers about symptom complaints for their infants diagnosed with ankyloglossia. Hill et al. (27) found approximately 12 percent (n=13) of 113 mothers had concerns about infantile reflux (seeking diagnosis). Ghaheri et al. (23) demonstrated that 45% of 237 (n=106) mothers reported signs associated with reflux in their infants at the start of the study (i.e., arching of the back, unable to lay flat after eating). Neither study reported a formal diagnosis of reflux by a healthcare provider, a limitation of both studies. Ghaheri et al. (23,26) also measured GI signs of reflux in two studies using I-GERQ-R scores preoperatively, 1 week and 1 month after frenotomy, with a similar approach by Slagter et al. (24) and Hand et al. (25). Slagter et al. (24) is the first study to follow-up longitudinally at 6 months post-frenotomy demonstrating a continued downward trend of I-GERQ-R mean total scores in infants (Figure 2). There was a significant decrease in I-GERQ-R scores at both 1- and 6-month time points (Table 1). A statistically significant, all values P<0.05, decline in total I-GERQ-R scores were present in all four studies (Figure 2). In 2022, Ghaheri et al. (10) measured reflux signs such as using the GSQ-I on Day 0 and Day 10 post-frenotomy. The authors found a statistically significant improvement between Day 0 and Day 10 on GSQ-I mean scores. It is unclear if these scores correlated with clinical significance. Two of the above studies suggest aerophagia as a potential cause of reflux (12,26). Imaging or auscultation
to confirm excess air in the abdomen would help support this postulation in future research.

Other GI signs
Ramoser et al. (11) explored the short-term (1 to 18 weeks after frenotomy) and long-term (15 months after frenotomy) outcomes of signs associated with ankyloglossia in 295 infants. Twenty-three infants (8% of the total sample) experienced signs of aerophagia, vomiting, and hematemesis. The short-term outcomes after frenotomy showed improvement signs in 84% of infants (n=247). Specifically, there were improvements in latch onto the breast, decreased nipple pain, less aerophagia, reduction in episodes of vomiting, and resolution of hematemesis. Long term outcomes demonstrated continued improvement in these signs for 77% of participants (n=227) (11).

Quality analysis on included articles
The research articles included in this review were rated based on the John Hopkins Evidenced-Based Quality Scale (29,30). This scale rates both evidence and quality of research, with level 1 representing the highest evidence (e.g., randomized control trial) and A quality providing

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<td><strong>Author/year</strong></td>
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**, P value is considered statistically significant P≤0.05; demonstrating that there is a relationship between the two variables in the study or that the relationship between reflux and ankyloglossia correction are not by chance (28). I-GERQ-R, Infant Gastroesophageal Questionnaire Revised version; GSQ-I, Infant Gastrointestinal Symptoms Questionnaire; GI, gastrointestinal.
generalizable and consistent results (29). Research articles by Ghaheri et al. (23,26), Slagter et al. (24), Ramoser et al. (11), and Hand et al. (25) were considered Level II of good quality, B, due to their quasi-experimental design, sufficient sample size, consistency, and definitive conclusions. The quasi-experimental design is non-randomized, evaluating an intervention, to demonstrate its relationship with an outcome. In these studies, frenotomy to treat ankyloglossia was the intervention of interest. The relationship between frenotomy and improvement in GI signs using the previously described screening tools was the desired outcome in these studies (31). These studies did not include a control group due to ethical considerations (16,32). The ethical considerations need to benefit the parties involved in human studies (32). By having a control group, these studies could raise ethical issues in the lack of intervention for those showing signs that may be due to ankyloglossia, and the potential effects on feeding that could impair growth and development. In all studies included, it was unclear what other interventions may have been used concurrently alongside frenotomy, such as lactation support or thickened feedings. It is also critical to recognize the maturity of the GI tract that occurs over time that may have resulted in improved reflux signs. Lastly, there are several different classification systems to diagnose ankyloglossia, none of which have been thoroughly tested for validity and reliability, limiting our ability to compare severity of ankyloglossia between studies and the relationship between severity and signs of reflux.

Ghaheri et al. (10) was assessed as Level II and lower quality, C, due to its limited sample size and lower quality conclusions. The research article was able to quantitatively identify a relationship between reflux and ankyloglossia with statistically significant differences between Day 0 and Day 10 GSQ-I scores on those with surgical treatment compared to an observational group (10). However, the similarities in the answers and scores on the GSQ-I of Day 0 between the observational group and surgical treatment group were not statistically significant (10). This does not mean that there is a lack of similarities in the comparisons but that it did not meet the standard cut off to achieve statistical significance (33). Lastly, the research by Hill et al. (27) is classified as Level V, with B-level quality, due to lack of experimentation. The authors were able to draw fair conclusions with clear aims and objectives in evaluating the relationship between ankyloglossia and GI signs (9,27).

**Discussion**

**Key findings and strengths**

This scoping review critiqued seven articles on GI signs
in infants with ankyloglossia. As defined, ankyloglossia is the restriction of tongue mobility and stability due to anatomical differences in the lingual frenulum (2,3). Six out of the seven articles discussed the presence of GER in infants with ankyloglossia, with improvement post-frenotomy in both short and long-term follow-up. GER is a common sign amongst infants under the age of 6 months, considered to be a normal GI variation early in life (13,20). The etiology of reflux is unclear, but presumed mechanisms include transient lower esophageal sphincter relaxations unassociated with swallowing, delayed gastric emptying, and other respiratory tract mechanisms (13). However, within review, there were both statistically and clinically significant improvements in reflux following treatment of ankyloglossia via frenotomy. This may suggest that reflux in infants with ankyloglossia may extend beyond the normal spectrum of signs expected early in life.

This review encourages us to consider ankyloglossia as a possible etiology for reflux, given its improvement after treatment of the oral restriction. Maternal concerns of reflux for infants with ankyloglossia signs were discussed in two studies (23,27). Four of the articles demonstrated mean I-GERQ-R scores that indicated “severe” reflux in infants with untreated ankyloglossia (Figure 2), with decreased in scores post-surgical intervention. Improving tongue mobility with frenotomy may positively impact both latch and swallowing. While most studies are limited to short-term follow-up, signs of reflux were reduced at 1-week, 1-month, and 6-month post-frenotomy (Figure 2). The difference in the mean total I-GERQ-R scores preoperative to postoperative ranged from 5 to 8 (Figure 2), with differences in scores above 6 considered statistically significant (20). Aerophagia was also one of the caregiver-reported signs present in infants with ankyloglossia (11), though this cannot be confirmed without physical examination or radiographic studies.

Other findings were Ghaheri et al. (10) used the GSQ-I to score specific symptoms that can lead to reflux and Ramoser et al. (11) detailed caregiver reported signs of vomiting and hematemesis. It is important to note, however, that hematemesis can be caused by other underlying issues beyond ankyloglossia such as esophagitis from GERD, ulcers, or swallowed maternal blood (34). A case study by Brooks et al. (9) showed aspiration and significant dysphagia during feeding for one infant with ankyloglossia. This was postulated to cause reflux and needs to be evaluated in future research with a larger sample.

**Limitations**

Beyond those discussed throughout this scoping review, additional limitations include the limited research on this subject, differing study methods (i.e., sample size, questionnaires, follow-up), the highly disputed definition of ankyloglossia, and the presumptive association between ankyloglossia and reported GI signs without other differential diagnoses discussed or considered. While the overall quality of the included research was fair, more research is necessary. Future research could be strengthened by the inclusion of control groups, larger sample sizes, use of one consistent definition and classification of ankyloglossia, and longitudinal data on GI signs.

**Clinical implications and future research**

GER is commonly mis- and over-diagnosed without proper treatment or consideration for other diagnoses (12). GER represents the backward movement of gastric contents into the esophagus while GERD affects daily life and contributes to complications such as erosion of the esophagus, often requiring pharmacologic treatment. The recommended treatments for GER are thickened liquids, hydrolyzed protein-based formula, or eventually, invasive pharmacological treatment (13,34). Recent research has demonstrated negative sequelae of pharmacologic treatment options (35), with providers opting for non-pharmacologic management (e.g., thickened liquids, remaining upright after feeding). Many factors can affect the conservative treatment of thickeners such as nutritional properties, precipitation from thickeners, and bowel morbidities (36). By recognizing the long-term implications of conservative and invasive treatments that may cause side and adverse effects (2,12), it is important to first investigate possible differential diagnoses associated with reflux, including ankyloglossia. To screen for ankyloglossia, the healthcare provider should use screening tools such as the Hazelbaker Assessment Tool for Lingual Frenulum Function, the Coryllos system, and/or The Martinelli Lingual Frenulum Protocol and evaluate appearance, function, and impact on feeding (2). It is important to note that the appearance of the tongue alone does not provide sufficient data to support frenotomy.

Further research is necessary to quantify the relationship between ankyloglossia and reflux; this scoping review serves as an important summary for clinicians to assess for ankyloglossia and consider specialty referral and
possible surgical intervention as one way to improve reflux. However, it is important to recognize that in all seven studies included in this scoping review, mean infant age was less than 6 months. With reflux considered a normal occurrence in this age group, more research is necessary to evaluate longitudinal improvement in reflux for infants with ankyloglossia without surgical intervention. Maternal breastfeeding symptoms, parental distress, and quality of life should also be considered when making the decision to treat.

Beyond reflux, other pharyngeal and GI signs such as vomiting, hematemesis, and regurgitation should be explored in future research. Although frenotomy is deemed a minor surgical intervention, there have been rare complications reported including bleeding, infection, scarring, and need for additional intervention (2,12). It is unclear which children benefit most from the procedure and who should undergo treatment. Future research is needed to explore GI signs to determine when surgical intervention is necessary to relieve reflux and reduce the use of potentially harmful treatments.

Conclusions

This scoping review has summarized GI signs in infants with ankyloglossia, comparing these before and after treatment via frenotomy. Most notably, signs of reflux improved with ankyloglossia treatment, suggesting a relationship between GI signs and ankyloglossia. Future research is required to further investigate the association between GI signs/symptoms and ankyloglossia; as well as the physiologic rationale for these findings.

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Footnote

Reporting Checklist: The authors have completed the PRISMA-ScR reporting checklist. Available at https://pm.amegroups.com/article/view/10.21037/pm-23-35/rc

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Conflicts of Interest: Both authors have completed the ICMJE uniform disclosure form (available at https://pm.amegroups.com/article/view/10.21037/pm-23-35/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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