



Effectiveness of Frenuloplasty in Ankyloglossia on Speech Quality: A Systematic Review

Yossy Yoanita Ariestiana¹, Mohammad Gazali¹, Husnul Basyar^{2*}

¹Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Hasanuddin University - Hasanuddin University Hospital - Dental Hospital Hasanuddin University, Makassar, Indonesia; ²Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Hasanuddin University, Makassar, Indonesia

Abstract

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***Correspondence:** Husnul Basyar, Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Hasanuddin University, Makassar, Indonesia. E-mail: husnul2204@gmail.com
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BACKGROUND: Ankyloglossia, known as tongue-tie, is an inherited anomaly and is caused by a short nonelastic frenulum that causes limited tongue movement. Because of the limitations of tongue movement, it can affect the quality of speech. Ankyloglossia treatment is to divide or separate fibrous bands or frenuloplasty. The purpose of making this systematic review is to systematically review the results of frenuloplasty governance in cases of ankyloglossia related to speech quality.

AIM: The aim of this systematic review is to explain the original study in patients with Ankyloglossia who underwent a frenectomy procedure on the tongue tie. This study included research evaluating results related to speech quality.

METHODS: PubMed, EMBASE, and Cochrane Databases, No restrictions on published studies until September 10, 2021. Studies are included if subjects of all ages have ankyloglossia and performed frenuloplasty procedures. The results assessed were the level of speech quality in the subjects of preoperative ankyloglossia and postoperative frenuloplasty.

RESULTS: Overall, 473 abstracts resulted from literature searches; 13 studies met the criteria for data extraction and analysis. Of the 13 studies, eight studies were Randomize control trial studies and 5 case-control studies. Three studies evaluated speech outcomes using Likert scores, 3 studies using questionnaires, and 7 studies using different assessments pre-operative and post-operative speech quality.

CONCLUSION: Frenuloplasty in subjects with ankyloglossia mostly gives good results in terms of speech quality. Improvements in articulation and mention of consonant phonation improve after frenuloplasty. Frenuloplasty with the 4 flap Z-Plasty Technique provides better results compared to conventional and horizontal vertical frenuloplasty. A long-term study of the correlation of ankyloglossia and speech difficulties and the effects of frenuloplasty is needed.

Introduction

The lingual frenulum is a small fold of soft tissue that extends from the base of the mouth to the middle line of the bottom of the tongue. Ankyloglossia, known as tongue-tie, is an inherited anomaly and is caused by a short non-elastic frenulum that causes limited tongue movement. The prevalence of Ankyloglossia is 4–10%, more common in men. It is an entity and not a risk factor. Ankyloglossia can be accompanied by other craniofacial anomalies such as aortic slits and cleft lip [1], [2].

Ankyloglossia treatment is to divide or separate fibrous bands or frenectomy to get better tongue mobility [3], [4]. Although this procedure is a mild and safe procedure, when and how to intervene is still a matter of debate. In some cases of frenectomy, there are serious and potentially life-threatening complications. In addition to the anterior and posterior lingual of the tongue, the procedure on the upper lip-tie is also a concern for the participants Clinical [5].

Due to the limitations of tongue movement, some of the symptoms that can occur are problems in breastfeeding, speech, and oral hygiene. The most important problem in preschool-aged children is the problem of articulation due to the limited mobility of the tip of the tongue. Due to the wide range of symptoms, a multidisciplinary approach involving several disciplines must be coordinated on the governance of Ankyloglossia. Although Ankyloglossia has been known for centuries, the clinical implications of its diagnosis and management are still controversial [6], [7].

The aim of this systematic review is to explain the original study in patients with Ankyloglossia who underwent a frenectomy procedure on the tongue tie. This study included research evaluating results related to speech quality. This study intends to assist medical personnel in considering the procedure of frenectomy in patients with Ankyloglossia by providing an up-to-date literature assessment, as well as for further research.

Methods

Procedures and registration

The statements and reporting selected in this systematic review follow the latest current guidelines [8]. A detailed research protocol is designed with the aim of explaining the scope of study, objectives, hypotheses, and methodologies. The data obtained are created in the form of a data collection form that is converted in the form of a table.

Eligibility criteria

The research questions for this study are formulated as follows: Population: each individual with Ankyloglossia. Intervention: Frenectomy in Ankyloglossia. Comparison: Before and after the release of Ankyloglossia. Outcome: Subjective and objective assessment of speech quality is assessed through several assessment methods [9].

Studies are included if the subject is of any age and has Ankyloglossia and Ankyloglossia release is carried out to overcome speech problems (articulation). The Ankyloglossia release procedure in question is a frenotomy or frenulotomy (division of lingual frenulum), frenuloplasty (phrenotomy with tailoring), and frenectomy or phrenulectomy (lingual frenulum excision), 4 z-plasty flaps, 4 z-frenuloplasty flaps, and horizontal vertical frenectomy.

Studies included in the criteria must be in English, the types of studies included are case series, case control studies, cohort studies, and randomized control studies. Duplicate studies and case series with < 5 people were excluded from the study. Must be full text accessible. Unpublished studies were excluded. Studies with patients who have other congenital histories are excluded (example of cleft lip and palate).

Data sources

A comprehensive search strategy is used to search for data on the following journal search sites: MEDLINE, EMBASE, and Cochrane Database of Systematic Review (Figure 1). Titles and abstracts are taken for all studies identified by the search strategy. Bibliography of the studies obtained in the full text is hand-searched in addition to relevant studies that were not identified or obtained on the original database search.

Types of research

Two authors (YY and HB) independently reviewed the list of titles and abstracts generated by

<p>PubMed: Run Until Sept 10th 2021 Search Strategy: 1. (((((((((((frenuloplasty) OR (tongue tie release)) OR (tongue-tie release) OR (ankyloglossia release)) OR (lingual frenulum release)) OR (lingual frenum release)) OR (lingual frenum surgery)) OR (lingual frenulum surgery)) OR (frenulectomy) OR (frenotomy) OR (frenectomy)) OR (frenulotomy) OR (frenuloplasty) OR (frenulectomy) 2. (((((((((((ankyloglossia) OR (tongue tie) OR (tongue-tie)) OR (short lingual frenum)) OR (short lingual frenulum)) OR (lingual frenum[MeSH Terms]) OR (lingual frenulum[MeSH Terms]) 3. (((((((((((speech[MeSH Terms]) OR (speech disorders[MeSH Terms]) OR (speech limitations[MeSH Terms]) OR (speech difficulty[MeSH Terms]) OR (speech intelligibility[MeSH Terms]) OR (articulation[MeSH Terms]) OR (articulation disorder[MeSH Terms]) OR (talk[MeSH Terms]) 4. 1 AND 2 5. 1 AND 2 AND 3</p> <p>COCHRANE: Run Until Sept 10th 2021 Search Strategy: 1. Frenectomy OR frenuloplasty OR frenulectomy OR frenotomy OR tongue tie release 2. Ankyloglossia OR tongue tie OR short lingual frenum OR short lingual frenulum OR lingual frenulum 3. Speech OR speech disorders OR speech difficulty OR articulation disorder OR articulation 4. 1 AND 2 AND 3</p> <p>EMBASE Run Until Sept 10th 2021 Search Strategy: 1. Frenuloplasty OR Surgery OR Surgical OR Tongue tie release OR tongue-tie-release OR lingual frenum surgery OR lingual-frenul-surgery OR Frenotomy OR Frenulectomy OR Frenectomy 2. Ankyloglossia OR tongue tie OR short lingual frenum OR short lingual frenulum OR lingual frenulum 3. Speech OR speech disorders OR speech difficulty OR articulation disorder OR articulation OR talk 4. 1 AND 2 AND 3</p>
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Figure 1: Literature search strategy

the literature search for each study that met the criteria mentioned above. The third author (GA) is involved when there is a conflict or problem. The study of full text is evaluated and adjusted to the inclusion criteria by two authors independently.

The types of data extracted are as follows: Author, year of publication, study design, number of cases or patients, description and characteristics of patients or cases, description of interventions performed, evaluation and outcome of intervention results. For quantitative results, the average/median value, confidence interval or p-value is recorded, if possible; data is synthesized into tables in the form of excel.

Bias risk analysis

Bias risk analysis in individual studies was assessed using the Joanna Briggs Institute (JBI) Global Appraisal Tools published by the Faculty of Health and Medical Sciences at the University of Adelaide, South Australia in 2020. In this systematic review study, there are 2 types of research methods (Case Control and Randomize Control Trial) each conducted assessment using checklist for case-control studies and checklist for randomized controlled trials.

Assessment conclusion

The assessment aspect that is evaluated before and after the frenectomy is performed is an assessment on the quality aspect of speech.

Synthesis of results

The data are synthesized into tables, according to the nature of the size of the result. No meta-analysis

is performed due to heterogeneity of data and lack of access to raw data.

Results

Types of research

A total of 473 abstracts were generated from databases and bibliographic searches. Figure 2 shows the flow diagram of the data search and exclusion process [10]. A total of 49 studies met the criteria for a literature review, 424 abstracts were eliminated because they did not discuss frenectomy in Ankioglosia, or the article did not include the subject of the study or only discussed about Ankioglosia without including the procedure, or only included procedures without information on the results of the research. After further review, 13 studies met the criteria for data extraction and analysis [11], [12], [13], [14], [15], [16], [17], [18], [19], [20], [21], [22], [23]. Four articles were issued because the sample number of the serial case was < 5 subjects and there were articles that could not be accessed by the full article.

Risk of bias in and between studies

In the case-control study, all studies qualified from the JBI criteria, where there were 3 studies that met 100% of the criteria (Salt *et al.*; Daggumati *et al.*; Dollberg *et al.*), 2 studies that met the 80% criteria (Triptodi *et al.* and Walls *et al.*). In the Randomized

Control Trial Study, 6 studies met the 70% criteria (Baxter *et al.*; Messner *et al.*; Ito *et al.*; Mascosan *et al.*; Camargo *et al.*; Puthussery *et al.*), 2 studies that meet the criteria above 70% (Kim *et al.* and Heller *et al.*). Details of quality assessment according to JBI criteria are displayed in Table 1a and b.

Characteristics of the study

Of the 13 studies conducted by data extraction and data analysis, 5 were *Case Control* studies and 8 were *Randomize Control Trial* studies. From 13 studies, 3 studies evaluated how to speak using the *Likert Score*, 3 studies using questionnaire assessments, and 7 studies using different assessments. The total subjects in the 13 studies were 442 with age ranges ranging from 1 to 33 years. Of the 442 subjects, 158 patients underwent a frenectomy using a laser, 35 underwent frenuloplasty procedures, and 214 underwent a frenotomy without a laser. The most commonly reported indication in 13 studies was speech problems and this case the most frequently complained about was consonant articulation and cloaking. The characteristics of the study are summarized in Table 2.

All studies included in the inclusion criteria use well-explainable samples. There is only 1 study included in the inclusion criteria using 5 research subjects, the average number of research subjects from the 13 studies is 34.4 studies with a range of 10–20 subjects, 2 studies with a subject range of 20–30, 7 studies with subjects over 30.

Likert score after Ankioglosia handling (Table 3)

Four studies reported evaluations of pre-operative speech and postoperative frenectomy using a Likert score. Baxter *et al.*, by using the Likert Score evaluating several speech anomalies in terms of speech in 37 patients who underwent a frenectomy using a laser, the change did not occur shown in the failure aspect with a value of $p = 0.500$. After a frenectomy reported no obstacles in communication, easy to understand by parents and others, children began to easily speak quickly and easily put out words, speech delays were experienced in some vocabulary just now, children who experienced speech delays produced new words, muttered less with a p -value on the indicator below 0.05.

Walls *et al.* reported that standard articulation tests conducted by speech pathologists who conducted a survey of parents by telephone showed an improvement in speech quality in 104 subjects conducted frenotomy at the time of the baby with an average age of 9 days. Salt *et al.*, who conducted case control studies between the treatment group ($n = 21$), the non-treatment group ($n = 17$) and the non-tongue-tie group ($n = 21$) showed significant

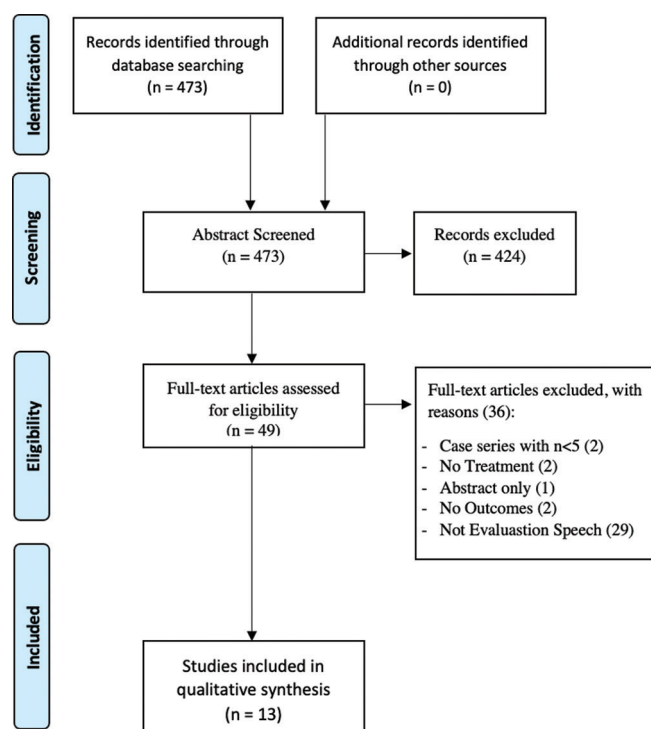


Figure 2: Flowchart of the article selection process

Table 1a: Quality assessment of individual studies (case control)

Study/criteria	Were the groups comparable other than the presence of disease in cases or the absence of disease in controls?	Were cases and controls matched appropriately?	Were the same criteria used for identification of cases and controls?	Was exposure measured in a standard, valid and reliable way?	Was exposure measured in the same way for cases and controls?	Were confounding factors identified?	Were confounding factors identified?	Were confounding factors identified?	Was the exposure period of interest long enough to be meaningful?	Was appropriate statistical analysis used?
Sait et al.	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Daggumati et al.	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Tripod et al.	Yes	Yes	Yes	Unclear	Yes	Yes	Yes	Yes	Yes	Unclear
Walls et al.	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Yes	Yes	Yes
Dollberg et al.	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes

Table 1b: Quality of individual studies (Randomized Control Trial)

Study/criteria	Was true randomization used for assignment of participants to treatment groups?	Was allocation to treatment groups concealed?	Were treatment groups similar at the baseline?	Were participants blind to treatment assignment?	Were those delivering treatment blind to treatment assignment?	Were treatment groups treated identically other than the intervention of interest?	Were those delivering treatment blind to treatment assignment?	Were treatment groups treated identically other than the intervention of interest?	Were participants analyzed in the groups to which they were randomized?	Were outcomes measured in a reliable way?	Was the trial design appropriate, and any deviations from the standard RCT design (individual randomization, parallel groups) accounted for in the conduct and analysis of the trial?
Baxter et al.	Yes	No	Yes	No	Unclear	Yes	Yes	Yes	Yes	Yes	Yes
Messner et al.	Yes	No	Yes	No	Unclear	Yes	Yes	Yes	Yes	Yes	Yes
Ito et al.	Yes	No	Yes	No	Unclear	Yes	Yes	Yes	Yes	Yes	Yes
Marcesan et al.	Yes	No	Yes	No	Unclear	Yes	Yes	Yes	Unclear	Yes	Yes
Kim et al.	Yes	No	Yes	Yes	Unclear	Yes	Yes	Yes	Yes	Yes	Yes
Camargo et al.	Yes	No	Yes	Unclear	Unclear	Yes	Yes	Yes	Yes	Yes	Yes
Heller et al.	Yes	No	Yes	Unclear	Unclear	Yes	Yes	Yes	Yes	Yes	Yes
Puthussery et al.	Yes	No	Yes	Unclear	Unclear	Yes	Yes	Yes	Yes	Yes	Yes

RCT: Randomized control trial.

Table 2: Characteristics of studies included in systematic review

Serial number	Study	Study design	n	Patient characteristics	Intervention	Result
1	Baxter <i>et al.</i>	RCT	37	Mean patient age 4.2 years (13 months–12 years)	Lingual frenectomy with CO ₂ laser and myofunctional exercises	Likert scale
2	Messner <i>et al.</i>	RCT	21	Patient age 1–12 years 19 men and 11 women 4 patients under 2 year old, 26 over 2 year old 3 patients have a family history of Ankioglosia	Frenuloplasty conventional, 26 th in general anesthesia, 4 in local anesthesia	Clinical assessment and questionnaire for 1 week, 1 month and 3 months post-operation
3	Salt <i>et al.</i>	Case control	21	Patient age 31–59 months 11 men, 10 women	Frenulotomy with Erbium: YAG laser	Assessment of speech aspects: Phonology Test according to DEAP TPT - Likert scale
4	Ito <i>et al.</i>	RCT	5	Patient age 3–8 years 4 men, 1 female	Frenuloplasty 4 patients in general anesthesia, frenulotomy 1 patient in local anesthesia Frenulectomy	The fifth item "Speech for age" dari Japan Society of Logopedics and Phoniatrics
5	Daggumati <i>et al.</i>	Case control	46	The average age of the patient was 40.97 months 50% have a family history 32 men, 14 women	Laser frenulectomy with Galbiati G25 diode laser (AlGaAs) 4 W, fiber 320 μm in local anesthesia Frenectomy	Likert score
6	Tripodi <i>et al.</i>	Case control	90	Patient age 6–12 years 45 men, 45 women	Laser frenulectomy with Galbiati G25 diode laser (AlGaAs) 4 W, fiber 320 μm in local anesthesia Frenectomy	Clinical assessment and questionnaire
7	Marcesan <i>et al.</i>	RCT	10	Patient age 2–33 years 8 men, 2 women	Frenotomy	Evaluated by a speech therapist
8	Kim <i>et al.</i>	RCT	37	Patient age 3–7 years 23 men, 14 women	Frenuloplasty 19 patients, 4 flaps 18 Frenotomy	PRES REVT U-TAP Likert-type scale, including with speech pathologists
9	Walls <i>et al.</i>	Case control	104	Mean patient age 3 years 62 men, 42 women	Frenotomy	Semi-spontaneous speech (reporting travel), automatic speech (counting from 1 to 20) You can mention the day and month of the week Mention of images (25 images to assess brazilian Portuguese accents and 25 to assess disturbed sounds due to frenulum alterations Alveolar sound t, d, n, s, z, J, l How to speak and the mobility of the tongue is recorded for post-Action analysis Diga arara baixinho ("Say arara softly") Speech evaluations are made by two different speech pathologists and are compared for consignment evaluations
10	Camargo <i>et al.</i>	RCT	13	Patient age 7.3–47.7 years with an average of 9.8 years 11 men, 2 women	Frenectomy	Questionnaire, 1 day, 7 days, 1 month post operation with 5 categories
11	Heller <i>et al.</i>	RCT	16	Patient age 3.1–9.9 years by mean 5.56 year old 9 men, 7 women	Four-flap Z frenuloplasty 11 patients, horizontal vertical frenectomy 5 patients	Standardized articulation test applied to all study participants by 2 speech pathologists who did not know the criteria for group requirements The test consists of 32 images of words in Hebrew consisting of 2–3 silabels
12	Puthussery <i>et al.</i>	RCT	10	Patient age 3–30 years	Laser phrenectomy, both local anesthetist and general anesthesia, and hospitalized Frenectomy	
13	Dollberg <i>et al.</i>	Case control	23	Patient age 4–8 years 17 men, 6 women	Frenectomy	

TPT: Toddler phonology test, DEAP: Diagnostic evaluation of articulation and phonology, PRES: Preschool receptive-expressive language scale, REVT: Receptive and expressive vocabulary test, U-TAP: Urmal test of articulation and phonation, RCT: Randomized control trial.

results in terms of improving the number of people by resulting in 4.05 with range (3–5), the increase in the number of sequences with the result of 3.95 with range (2–5), which approaches the intelligence of speaking in groups that do not have a tie, and in the group no treatment shows more errors.

In contrast to the report in the Daggumati *et al.* study that compared the control group (n =31) and the treatment group (n = 46) reported that there was no significant difference between the two groups in terms of speech quality and tongue mobility with a value of p = 0.484.

Assessment with questionnaire after Ankioglosia handling (Table 3)

Evaluation of speech difficulties using questionnaires was reported in three studies, of these three studies all reported improved speech quality in subjects performed frenectomy, where each study performed a frenectomy using a laser. Messner *et al.* reported frenectomy results in 30 subjects aged

1–12 years, with 28 subjects continuing the evaluation and 2 subjects not continuing the evaluation, with speech delays in pre-surgery of 3.3 increasing to 4.2 in post-operative where a scale of 1 indicated significant delay and 5 indicated conformity. Frenectomy can also improve phonation. This is indicated by a study conducted by Tripodi *et al.* which conducted research on 90 subjects divided into 45 male subjects and 45 female subjects. After a 12-month post-frenectomy evaluation, there was a significant increase in phonation changes. Puthussery *et al.* who conducted a study on 21 subjects conducted frenectomy intervention using laser dioxide carbon, after postoperative evaluation, only two parents did not agree with the improvement in terms of speech of their children.

Other assessments after the treatment of Ankioglosia (Table 3)

Seven studies showed different assessments in assessing speech quality after a frenectomy. Salt *et al.* in addition to conducting an evaluation with Likert Score also conducted an evaluation with the Phonology

Table 3: Assessment results after treatment of ankyloglossia

Serial number	Study	n	Evaluation	Speech results	Conclusion
1	Baxter <i>et al.</i>	37	Likert scale	Frustration when communicating reduced $p < 0.001$ Easier for parents $p = 0.001$ Easier for others $p = 0.001$ It's easier for children to talk faster $p = 0.004$ Issued the word $p < 0.002$ Previously difficult to sound $p < 0.001$ Speech delay and production of new words in children $p = 0.008$ Still stuttering $p = 0.500$ Less muttering $p = 0.008$ Used less baby talk $p = 0.031$	Increase
2	Daggumati <i>et al.</i>	46	Likert scale	Difficulty speaking with a Likert score of 1.48. $p = 0.484$	Not increase
3	Walls <i>et al.</i>	104	Likert scale	Significant speech improvement with a Likert score of 4.52 (0.61)	Increase
4	Salt <i>et al.</i>	21	Likert scale Sub-tes dari DEAP	Intelligibility according to parents with a result of 4.05 with a range (3–5) Intelligibility according to clinicians with a result of 3.95 with a range (2–5) PCC with an average increase of 77.95, range (47–99)	Increase
5	Messner <i>et al.</i>	21	Assessment questionnaire after Ankyloglossia handling	Preoperative point 3.3 increased to an average postoperative point of 4.2 ($p < 0.01$)	Increase
6	Tripodi <i>et al.</i>	90	Assessment questionnaire after Ankyloglossia handling	The 12 months evaluation gave good significant results against phonation changes	Increase
7	Puthussery <i>et al.</i>	21	Assessment questionnaire after Ankyloglossia handling	Only 2 parents disagreed with the increased speech after frenectomy	Increase
8	Ito <i>et al.</i>	5	The fifth item, "Speech for Age" oleh Japan Society of Logopedics and Phoniatics	Misarticulation and sounds expressed based on the international phonetic alphabet Substitution from a total of 19 preoperative to 1 after post-evaluation of 1–2 years Omissions from a total of 5 perioperative to 1 after post-operative evaluation 1–2 years Distorsi dari 13 pre-operative menjadi 11 setelah evaluasi post op 1–2 tahun	Increase
9	Marcesan <i>et al.</i>	10	Evaluated by a speech pathologist	8 out of 10 subjects experience increased speech to be more efficient	Increase
10	Kim <i>et al.</i>	37	Test Urimal untuk artikulasi dan fonasi/U-TAP	Articulation test (consonant) the rate of improvement (%) increases by 21.44 ± 23.49 on simple frenectomy and 22.94±38.69 on 4 flaps of Z-plasty	Increase
11	Camargo <i>et al.</i>	13	Mention of images (25 images to clarify the Brazilian Portuguese accent and 25 images to clarify the most influential sounds due to frenulum alterations) Alveolar sound (t, d, n, s, z, J, l) The way of speech and mobility of the tongue are recorded for post-analysis Diga arara baixinho ("Mention arara in a gentle way")	There was no significant difference in consonant production between preoperative and postoperative with values $p = 0.35$ and $p = 0.77$	Not Increase
12	Heller <i>et al.</i>	16	Speech evaluations are made by two speech pathological people who are then compared for consistency	91% of the total 11 patients had significant improvements in articulation in patients who performed 4 flap S of Z-frenuloplasty. 60% of the total 5 patients did not give results, 40% gave significant results in patients performed horizontal-to-vertical frenuloplasty	Increase
13	Dollberg <i>et al.</i>	22	Standardized articulation tests were given to all study participants by two speech pathologists who were unaware of the group's tasks The test consists of 32 images consisting of 1–3 syllables of the word Hebrew	There was no significant difference in consonant articulation between pre- and post-operative	Not increase

DEAP: Diagnostic evaluation of articulation and phonology, PCC: Percentage consonants correct, U-TAP: Urimal test of articulation and phonation.

Subtest with the Diagnostic Evaluation of Articulation and Phonology by assessing the Percentage Consonant Correct (PCC). This assessment was carried out on three categories of subjects with very significant results. PCC in the tongue tie treatment group with an average increase of 77.95% (range 47–99), in the nontongue tie group the PCC value was 72.62 (range 39–96), but in the group without treatment the average value of PCC did not reach 70%.

Ito *et al.*, who conducted research on 5 subjects with the assessment of The fifth item, "Speech for Age" by the Japan Society of Logopedics and Phoniatics which assesses misarticulation and sounds that appear based on the International Phonetic Alphabet on 3 aspects of speech or mentions, namely Substitution, Negligence, and Distortion. Substitute assessment in 5 subjects with a total point of 19 on preoperative decreased in the postoperative period to reach 1 point

after evaluation of 1–2 years. For omissions point with a total point of 5 on the pre-operative to 1 post-operative after evaluation 1–2 years. Distortion with a total point of 13 perioperative from 5 subjects to 11 postoperative points after 1–2 years of evaluation. This shows significant improvements in speech, especially in the aspects of misarticulation and sound.

Marcesan *et al.* reported the results of a study on 10 subjects who underwent a phrenectomy and conducted post-operative evaluation with the assessment method evaluated by speech language pathologists (SLPs), 8 out of 10 subjects experienced an efficient improvement in speech. Heller *et al.*, who conducted a study on 16 subjects on articulation assessed objectively by 2 pathological speech people. 11 people performed 4 flaps Z-Frenuloplasty and 5 people performed horizontal vertical frenuloplasty. 91% of the total 11 subjects in group 4 flap Z-Frenuloplasty

experienced significant articulation improvements, while in the horizontal vertical group frenuloplasty only 40% gave significant articulation improvement results. This shows that the 4 flap technique is more effective than the vertical horizontal technique in terms of articulation improvement.

Discussion

There is no simple, definitive tool that can be used to clinically determine about Ankyloglossia. This makes it difficult to identify the target population for Ankyloglossia studies and gives a clear indication for the phrenotomy. To this end, the relationship between breastfeeding problems, speech articulation problems, Ankyloglossia, and phrenotomy needs to be examined more closely. Some studies have tried to use more objective or standardized measures during the initial assessment phase but the practicality and usefulness of such actions in clinical settings remains questionable [24], [25].

Ankyloglossia procedure is a surgical and non-surgical therapy. The main surgical therapy is *frenectomy*. Non-surgical therapy is done for the symptoms caused by Ankyloglossia such as consultation with a pediatrician for symptoms caused when breastfeeding such as maternal breast pain, breastfeeding position, or stretching on the tongue. Other non-surgical therapies are physical therapy, talk therapy, complementary therapies such as craniosacral therapy, naturopathy, and orofacial myofunctional therapy [26].

The main surgical procedures performed are frenectomy, frenulectomy with or without myotomy, and Z-plasty. Frenectomy usually gives good results in babies. Whether it is using conventional frenectomy techniques, using laser diodes or using electrocauters. The technique of excision of frenulum (frenulectomy) or frenuloplasty, which combines the elevation of a flap or Z-plasty can be performed and is more over the general anesthetic. In infants using the Z-plasty technique and conventionally give the same good results. However, Z-plasty will give good results in giving free movements and to prolong the motion of the tongue [27], [28].

Speech motor function is strongly associated with facial bone structure and muscle mobility within the oral cavity such as the tongue [29]. At present, there is no hard evidence to explain that Ankyloglossia can cause speech problems. The last few studies have explained only a little about the relationship between Ankyloglossia and speech difficulties, but the conclusions presented are not expressly stated.

In a case control study conducted by Dollberg *et al.*, with the help of 2 speech pathologists who

assessed speech clarity with standard articulation tests, it concluded there was no significant difference in speech outcomes between the treatment group and the control group. This study was conducted in Hebrew (Silabel in Hebrew) which made the results less applicable to children who spoke English and other languages [23].

Case control studies conducted by Daggumati *et al.* showed that children with Ankyloglossia who underwent surgical intervention had the same speech quality compared to children who were treated conservatively without surgery. According to a telephone survey of babysitters using the Likert Score, there was no significant difference in speech difficulties and tongue mobility between children who were surgically treated and who were treated conservatively in Ankyloglossia. This data suggest that a conservative approach to Ankyloglossia governance may be a usable approach for children with speech problems related to Ankyloglossia. However, surgical procedures are still recommended because they have minimal risk; however, ease of operation is not always the right indication to intervene [15].

One of the assessments of speech quality is the assessment of consonant sounds, in the case control study with the Likert score evaluation conducted by Daggumati *et al.*, which compared consonant sounds in the treatment group (n = 46) and control groups (n = 31) The percentage of pre-operative stage consonant sounds (75%) and post-operative (77%) in the treatment group. Comparisons between consonant duration values in the pre- and postoperative stages showed only a slight difference. This showed no significant difference between the treatment group and the control group. Consonant duration values did not vary significantly among subjects in both the preoperative and postoperative stages [20].

Baxter R *et al.* reported the results of a study of 37 subjects on several indicators of speech difficulties, after a post-operative 1-week evaluation 76% of parents reported an increase in speech, at a post-operative 1-month evaluation 89% of parents reported an increase in speech in their children, these results decreased significant improvements to speech difficulties after a frenectomy. This research encourages the relevant health professions to treat Ankyloglossia to get maximum results on speech improvement [11].

The study conducted by Heller *et al.* compared two different types of frenuloplasty techniques on articulation assessment. The evaluation of speech is carried out by 2 different speech pathologists. Articulation errors are rated as zero, light, medium, or heavy with sounds on the mention of letters: S, Z, T, D, L, ZH, CH, J, and DG. Posterior sounds, such as "K" and "G." Broadly speaking, the action of frenuloplasty gives significant results in terms of increased articulation, although these results are more obtained in the frenuloplasty procedure with the 4 flap

Z-frenuloplasty Technique compared to the Horizontal-vertical frenuloplasty Technique [21].

The study by Walls *et al.*, in 104 neonates who underwent a phrenotomy due to breastfeeding difficulties maintained better speech outcomes when compared to individuals who resisted Surgical Intervention. Parents said there was a statistically significant improvement in speech outcomes after a three-year evaluation for patients undergoing a phrenotomy when compared to untreated patients. In addition, there was no statistical difference in speech outcomes between the frenotomy patients and the control group. Overall, frenotomy subjects during the baby experienced an increase in speech outcomes with an average Likert score of 4.52 [19].

In the Salt *et al.* study which divided three study groups, namely, treatment tongue tie (TTT), untreated tongue tie (UTT), and non-tongue tie (NTT) reported no significant difference in speech results found between children with UTT and NTT. The study compared children with TTT with UTT and NTT control groups at various measures, including improved speech clarity assessed by parents and physicians, and found no significant differences in speech outcomes between the three groups. But significant improvements were found in the TTT group with intelligibility according to parents and physicians of 4.05 and 3.95, respectively. Increased PCC with an average increase of 77.95, range (47–99) [13].

An increase in obvious phonation changes after surgery using lasers combined with speech therapy significantly gave satisfactory results [30]. This was shown in a study conducted by Tripodi *et al.* that conducted studies on 90 subjects. The use of lasers also makes healing less time than after scalpel dissection. The use of lasers in the procedure of frenectomy is also recommended by Puthussery *et al.*, where the use of this laser can reduce pain and postoperative swelling. Study Puthussery *et al.* reported that of the 21 subjects only 2 parents of subjects who did not agree with the improvement in the quality of speech in their children after a frenectomy [22].

Messner *et al.* who conducted a study on 15 children who successfully followed up with the age of 2 years or older who had undergone frenotomy reported a preoperative point Likert score of 3.3 increased to 4.2 post-operative. Phrenulotomy/frenuloplasty may be considered for infants 2 years of age or younger with significant Ankyloglossia, as speech and social/mechanical problems can go through an undeveloped phase. Phrenuloplasty is recommended for children of speech age with articulation difficulties. The same was also reported by Marcesan *et al.*, in 10 subjects where 8 out of 10 subjects experienced increased speech to be more efficient after a frenectomy [17].

Speech problems in children with Ankyloglossia are considered articulation disorders caused by limited

mobility of the tip of the tongue. Speech sounds that may be affected by impaired mobility of the tip of the tongue include lingual sounds and sibilants, such as t, d, n, l, s, r, z, and th [31], [32]. The same phenomenon was observed in cases in studies conducted by Ito Y *et al.*, with the exception of the sounds l and th, which do not exist in Japanese. Thus, it can be concluded that the freedom of movement of the tongue facilitated by the lingual frenulum will improve the ability to speak. At the point of substitute, omissions and distributions all decreased after a frenectomy [14].

Ankyloglossia, also known as tongue-tie, is a congenital oral disorder characterized by abnormal short lingual frenulum. The lingual frenulum, according to the International Affiliate of Tongue-Tie Professionals, is the remaining midline of tissue between the ventral surface of the tongue and the base of the mouth. When it interferes with normal functioning, this condition is called “symptomatic Ankyloglossia” [33]. Due to the limitations of the movement of the tip of the tongue, articulation problems are mainly in consonants such as s, z, t, d, r, l, j, ch, and th [34]. Research conducted by Kim *et al.* also showed that consonant articulation test results were lower than vocal results. In 37 subjects who followed the study until follow up it was reported that the Articulation test (Consonants) Improvement rate (%) increased by 21.44 ± 23.49 in simple phrenectomy and 22.94 ± 38.69 on 4 flaps of Z-Plasty. It is also shown that the 4 Flap Z-plasty procedures give better results compared to simple frenectomy [18].

Some SLPs, state that children with Ankyloglossia often have normal speech skills. In English, even sounds that require the most tongue movement, such as /l/ and /th/, can be produced with minimal distortion, with the tip of the tongue pressed down and up towards the alveolar ridge for the letter (/l/), or the tongue highlighted out for mention (/th/). Similarly, other sounds that require tongue elevation, such as /s/ and /z/ can also be produced effectively with the tip of the tongue downwards. As a result, Ankyloglossia should not have a dramatic impact on speech function in most cases [35].

Limitations

Although most studies use a large number of samples, the number is not well applied in all studies, so the number of study subjects varies greatly from 5 to 104 subjects. And there are 2 studies that do not explain the gender of the study subject. The age when performing frenectomy procedures in patients is very diverse, there are even studies that perform frenectomy procedures when they are babies so they have not evaluated the overall difficulty of speech before taking action. The types of speech difficulties evaluation in pre-operative and post-operative also vary, only few studies use the same parameters in assessing speech difficulties.

Conclusion

From the above review, we concluded that the procedure of frenectomy in subjects with Ankioglossia as a large extent gives good results in terms of speech quality.

Improved articulation and mention of consonant phonation improves after a frenectomy. Frenectomy with the 4 flap Z-Plasty Technique gives better results compared to conventional and horizontal frenectomy vertical frenuloplasty. Of the 13 studies reviewed, only three reported no significant differences in the quality of preoperative speech and postoperative frenectomy. Changes in the consonant phonation aspect significantly give good results after a frenectomy. We recommend an Ankioglossia assessment when still a baby and performed frenotomy if found it, because from the above study found good results after frenotomy in infants who have an Ankioglossia disorder.

To reinforce the results of research in this area, more long-term studies are needed on the correlation of Ankioglossia and speech difficulties and the more pronounced effects of frenectomy on changes or improvements in speech quality.

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