

# Interventions in the Alteration on Lingual Frenum: Systematic Review

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## Abstract

**Introduction** Altered lingual frenum modifies the normal tongue mobility, which may influence the stomatognathic functions, resulting in anatomical, physiological and social damage to the subject. It is necessary that health professionals are aware of the process of evaluation, diagnostics and treatment used today, guiding their intervention.

**Objective** To perform a systematic review of what are the treatment methods used in cases of lingual frenum alteration.

**Data Synthesis** The literature searches were conducted in MEDLINE, LILACS, SciELO, Cochrane and IBECs, delimited by language (Portuguese, English, Spanish), date of publication (January 2000 to January 2014) and studies performed in humans. The selection order used to verify the eligibility of the studies were related to: full text availability; review the abstract; text analysis; final selection. Of the total 443 publications, 26 remained for analysis. The surgical approach was used in all studies, regardless of the study population (infants, children and adults), with a range of tools and techniques employed; speech therapy was recommended in the post surgical in 4 studies. Only 4 studies, all with infants, showed scientific evidence.

**Conclusion** Surgical intervention is effective for the remission of the limitations caused by the alteration on lingual frenum, but there is a deficit of studies with higher methodological quality. The benefits of speech therapy in the post surgical period are described from improvement in the language of mobility aspects and speech articulation.

## Keywords

- ▶ lingual frenum
- ▶ therapeutics
- ▶ speech therapy
- ▶ surgical procedures
- ▶ operative

## Introduction

The lingual frenum is a middle fold of mucous membrane extending from the posterior-gum surface of the tongue, covering the lingual surface of the anterior alveolar crest.<sup>1</sup> In some cases, the lingual frenum may be changed, called ankyloglossia.<sup>2,3</sup> This alteration is characterized by a short and thick frenum<sup>4–6</sup> which may compromise tongue mobility.<sup>6–9</sup> However, there are varying degrees of commitment ranging from just a short and dense fold, until anterior

insertion,<sup>3,10</sup> causing controversy over its prevalence and clinical impact.

The etiology of the altered lingual frenum is still unknown, may be associated with risk factors such as being male (2.6:1)<sup>11</sup> and a positive family history.<sup>6,12</sup> Ankyloglossia can be considered as a relatively common disorder and studies demonstrate a prevalence ranging from 0.1 to 10.7%.<sup>6,13</sup>

When lingual frenum is altered and tongue mobility is affected, the subject may have problems related to feeding, such as sucking during breastfeeding, chewing and swallowing;

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articulation of speech; dental changes; and social functions.<sup>1,5,8,14</sup> Given the consequences related to lingual frenum alteration, often is needed to intervene and adequate tongue mobility and its functions, as well to improve the individual's quality of life.

Considering the various possibilities of intervention, including speech therapy intervention, to be performed in cases of ankyloglossia and the lack of studies on the same, it is believed a systematic review contributes to new thoughts on the theme. Thus, the objective is to conduct a systematic review of the evidence on the types of interventions used in the lingual frenum alteration and its evolution.

## Review of Literature

### Search Strategy

The question that guided the review was "What types of interventions are performed in the lingual frenum alteration?" From the formulation of guiding question, a bibliographic survey was conducted on the subject, interventions used in altered lingual frenum, in January 2014, in the electronic databases MEDLINE, LILACS, SciELO, Cochrane and IBECs. The search strategy applied followed recommendations of the latest version of "Cochrane Handbook for Systematic Reviews of Interventions"<sup>15</sup>. The choice of searching bases was due to its wide use by the community health sciences, and since they are source of internationally recognized references.

For the bibliographic search the following terms and combinations were used: lingual frenum x myofunctional therapy; lingual frenum x speech therapy; lingual frenum x therapeutics; ankyloglossia x myofunctional therapy; ankyloglossia x speech therapy; ankyloglossia x therapeutics; surgical procedures x lingual frenulum; surgical procedures x ankyloglossia; surgery x lingual frenulum; surgery x ankyloglossia. Search terms appear in the Medical Subject Headings (MeSH) and Health Sciences Descriptors (DeCS). "Ankyloglossia" is not listed in DeCS and "lingual frenulum" does not appear in any of the mentioned, only as synonymous in Portuguese of "lingual frenum," however we decided to keep it, as it is commonly used by Speech Therapy to replace the term "lingual frenum." Searches were delimited by language (English, Portuguese and Spanish), date of publication (January/2000 to January/2014) and studies performed in humans.

### Selection Criteria

The publications resulting from this search were analyzed by two researchers independently, following the following exclusion criteria: repeated articles and articles without full text available. All stages of the study were conducted independently by the researchers. When there was disagreement between the researchers, were included only the texts on which the final decision was consensual. In the sequence the articles were selected as according the execution of two-step selection. First, the articles had their abstracts analyzed, also independently and blindly considering as markers: if there were specific approach in lingual frenum, the type of study

(case study/case series, clinical trial/randomized, longitudinal or transverse), and the intervention type mentioned (surgery, speech therapy, or both). It was excluded texts about craniofacial anomalies (syndromes), literature reviews, systematic reviews, opinions of experts and articles in which intervention on the frenum was not the purpose of the study. In the second stage, the selected articles have undergone a complete revision of the text, to verify if they actually met the proposed inclusion criteria. At this stage were also excluded case studies and case series, because the methodology followed the recommendations of the Oxford Center Medicine,<sup>16</sup> using studies up to level 3 due to the scientific impact of the same.

### Data Analysis

The articles selected for review were analyzed according to these aspects: study design, sample characteristics, type of intervention, intervention characteristics, main results and conclusions.

## Discussion

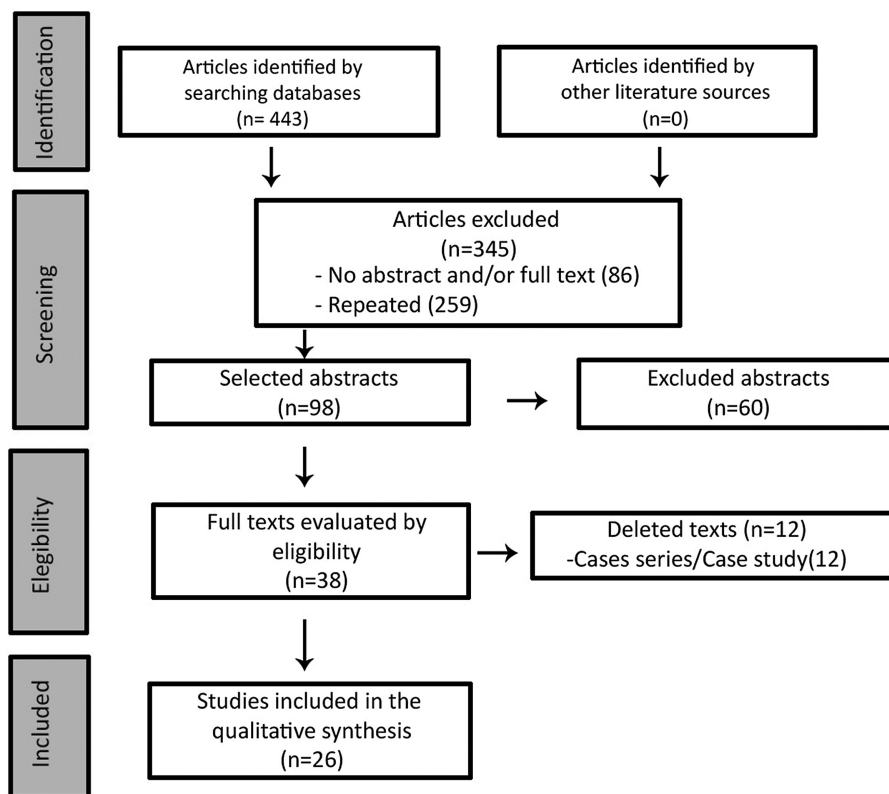
The electronic search in database resulted in the identification of 443 publications, 259 were excluded because they were repeated and the other 86 were excluded for not having abstract and / or full text. In total 98 full-text articles had their abstracts analyzed by the researchers, from these 60 were excluded because they did not treat directly approach the lingual frenum. Thus, remaining 38 articles, 12 were excluded for being case study or case series. In the end, 26 articles were reviewed, verifying the type of study and the intervention used. All stages of the selection process and analysis of the texts are represented in ►Fig. 1, based on the recommendations of the PRISMA.<sup>17</sup>

Analyzing the selected articles, it was found that there was greater scientific production on the searched topic as from the year 2002, and significant increase of publications in 2010, with an average of 3 publications per year. Most studies come from the United Kingdom<sup>3,13,14,18-22</sup> and United States.<sup>4,5,8,12,23-26</sup> The rest is divided between Brazil,<sup>9</sup> Ireland,<sup>27</sup> Korea,<sup>6</sup> Finland,<sup>7</sup> Turkey,<sup>2</sup> Australia,<sup>28,29</sup> Canada,<sup>30</sup> Israel<sup>10</sup> and Cuba.<sup>1</sup>

From the analysis of publications, one can verify that the studies had diverse objectives and characteristics. Thus, the study design, sample and type of intervention are shown in ►Table 1. The studies analyzed were grouped according to population and objective, described in more detail below.

### Studies with Infants

Studies show the increasing number of publications on the population of neonates and infants, due to breastfeeding difficulties associated with ankyloglossia. Some studies<sup>18,19,24</sup> mention the promoting of breastfeeding due to its advantages and the need for intervention in cases of difficulty to do it, avoiding discontinuation. The ankyloglossia can difficult the attachment in the areola, generating inadequate pressure to milk ejection, resulting in long breastfeeding sessions and little weight gain.<sup>18</sup> Also leads to



**Fig. 1** Representation of the selection and analysis process of the publications.

consequences for mothers, described as: sore nipples, little milk production and mastitis.<sup>26</sup>

Many studies are intended to assess breastfeeding, but few<sup>10,21,25,29,30</sup> used validated protocols and / or objective measures to quantify the quality of breastfeeding and felt pain during. The rest used questionnaires answered by the mothers of children, before the procedure and during follow up, investigating through reports which difficulties exist and if they persisted after surgery.<sup>3,8,13,18–20,22,24,26,28</sup> There was a study<sup>12</sup> that clinically evaluated breastfeeding before surgery, but without using protocol. In the lactation literature, surgical decision should be based on symptomatic complaints of the mother, little child's weight gain and findings of the oral examination.<sup>24</sup> There is description about other forms of intervention, aiming to change the baby's position in the mother's breast and using maneuvers. When these conducts are not effective, there is a referral to the surgeon.<sup>3,13,18–20,22,24–26,28–30</sup>

One study<sup>19</sup> mentions that some lingual frenum can be broken with the eruption of the lower teeth or objects in the mouth, not affecting the child in long-term. Thus there is no need for surgical intervention in all cases, but attention to the relationship between feeding difficulties and altered frenum enable intervention in symptomatic cases. Some studies<sup>18,19,22</sup> defend the instrumentalization of breastfeeding consultants, performing a simple procedure, because in this way it would decrease the delay between identification-intervention, allowing mothers persist in breastfeeding.

### Studies about Surgical Procedures and Techniques

Some studies report surgery intervention using laser method.<sup>2,14</sup> One of them<sup>14</sup> used carbon dioxide laser in a vertical section, demonstrating that this method causes less pain and swelling postoperatively, as well as less bleeding, and improved healing compared with traditional methods. In another study<sup>2</sup> there's comparison of the use of diode lasers and Er: YAG laser. Patients undergoing surgery with diode required local anesthesia due to discomfort, but those using Er: YAG felt greater pain 3h after the surgery. Other studies<sup>6,23</sup> mention the blade method, but using different techniques. One study<sup>6</sup> reported cases using the technique of "z-plasty" combined with partial myotomy of the genioglossus. According to the authors, releasing the contracted portion of the muscle increases the tongue mobility and protrusion, improving speech. But the study does not mention which minimum age is indicated for this type of procedure. Another study<sup>23</sup> compared the horizontal-vertical techniques and "four flap z frenuloplasty." The results of the study show that the group of patients undergoing different technique had higher benefit.

### Studies Related to Speech Therapy

A few selected publications mention the interdisciplinary work in relation to speech intervention. The study of children aged 1 to 12 years<sup>4</sup> and 3 to 9 years<sup>23</sup> show that speech, specifically the articulation, were altered in almost every subject, and this is also a complain of parents.<sup>4</sup> Regarding the study<sup>5</sup> with teenagers and adults 14 to 68 years, 50% had

**Table 1** List of articles selected for qualitative analysis

| Author, year                   | Study design                   | Sample   | Intervention               |
|--------------------------------|--------------------------------|--|----------------------------|
| Puthussery et al <sup>14</sup> | Clinical Trial, Non-Randomized | n= 21 subjects<br>Sex: NI<br>Age: 3 to 30 years  | Surgical                   |
| Marchesan et al <sup>9</sup>   | Clinical Trial, Non-Randomized | n= 10 subjects<br>Sex: 8♂ 2♀<br>Age: 2 to 33 years   | Surgical                   |
| Wallace e Clarke <sup>18</sup> | Clinical Trial, Non-Randomized | n= 10 subjects<br>Sex: 8♂ 2♀<br>Age: 2 to 31 days  | Surgical                   |
| Hogan et al <sup>19</sup>      | Randomized Clinical Trial      | n= 57 subjects<br>(CG 29; EG 28)<br>Sex: NI<br>Age: 3 to 70 days   | Surgical                   |
| Glynn et al <sup>27</sup>      | Clinical Trial, Non-Randomized | n= 48 subjects<br>Sex: 33 ♂ 15 ♀<br>Age: 3 to 192 months   | Surgical                   |
| Choi et al <sup>6</sup>        | Clinical Trial, Non-Randomized | n= 106 subjects<br>Sex: NI<br>Age: 1 to 10 years   | Surgical                   |
| Klockars et al <sup>7</sup>    | Clinical Trial, Non-Randomized | n= 317 subjects<br>Sex: 216♂ 101♀<br>Age: 0 months to 18 years   | Surgical                   |
| Heller et al <sup>23</sup>     | Clinical Trial, Non-Randomized | n= 16 subjects<br>Sex: 9♂ 7♀<br>Age: 3 to 9 years  | Surgical<br>Speech therapy |
| Lalakea e Messner <sup>5</sup> | Clinical Trial, Non-Randomized | n= 35 subjects<br>(CG 20; EG 15)<br>Sex: EG 11♂ 4♀<br>CG: NI<br>Age: EG 14 to 68 years<br>CG: 14 to 48 years | Surgical<br>Speech therapy |
| Hong et al <sup>8</sup>        | Clinical Trial, Non-Randomized | n= 341 subjects<br>Sex: 227♂ 114♀<br>Age: 1 day to 24 weeks  | Surgical                   |
| Aras et al <sup>2</sup>        | Clinical Trial, Non-Randomized | n= 16 subjects<br>Sex: 8♂ 8♀<br>Age: 18 to 27 years  | Surgical                   |
| Amir et al <sup>28</sup>       | Clinical Trial, Non-Randomized | n= 46 subjects<br>Sex: 29♂ 17♀<br>Age: 3 to 98 days  | Surgical                   |
| Ballard et al <sup>12</sup>    | Clinical Trial, Non-Randomized | n = 123 subjects<br>Sex: NI<br>Age: NI   | Surgical                   |
| Mettias et al <sup>20</sup>    | Clinical Trial, Non-Randomized | n= 63 subjects<br>Sex: NI<br>Age: mean 4 weeks   | Surgical                   |
| Steehler et al <sup>24</sup>   | Clinical Trial, Non-Randomized | n= 367 subjects<br>(CG 65; EG 302)<br>Sex: 216 ♂ 151♀<br>Age: mean 18 days                                   | Surgical                   |
| Buryk et al <sup>25</sup>      | Randomized Clinical Trial      | n= 58 subjects<br>(CG 28;EG 30)<br>Sex: 38 ♂ 20♀<br>Age: 1 to 35 days  | Surgical                   |

**Table 1** (Continued)

| Author, year                   | Study design                   | Sample  | Intervention               |
|--------------------------------|--------------------------------|---|----------------------------|
| Miranda e Milroy <sup>3</sup>  | Clinical Trial, Non-Randomized | n= 62 subjects<br>Sex: NI<br>Age: 12 to 36 days                         | Surgical                   |
| Geddes et al <sup>29</sup>     | Clinical Trial, Non-Randomized | n= 24 subjects<br>Sex: NI<br>Age: 1 to 131 days                         | Surgical                   |
| Srinivasan et al <sup>30</sup> | Clinical Trial, Non-Randomized | n= 27 subjects<br>Sex: 18 ♂ 9♀<br>Age: 2 to 71 days                     | Surgical                   |
| Dollberg et al <sup>10</sup>   | Randomized Clinical Trial      | n= 25 subjects<br>(CG 11; EG 14)<br>Sex: NI<br>Age: 1 to 21 days        | Surgical                   |
| Messner e Lalakea <sup>4</sup> | Clinical Trial, Non-Randomized | n= 30 subjects<br>Sex: 19 ♂ 11♀<br>Age: 1 to 12 years                   | Surgical<br>Speech therapy |
| Navarro e López <sup>1</sup>   | Clinical Trial, Non-Randomized | n= 29 subjects<br>Sex: NI<br>Age: 5 to 11 years                         | Surgical<br>Speech therapy |
| O'Callahan et al <sup>26</sup> | Clinical Trial, Non-Randomized | n = 299 subjects<br>Sex: NI<br>Age: between 2 and 323 days              | Surgical                   |
| Sethi et al <sup>13</sup>      | Clinical Trial, Non-Randomized | n= 85 subjects<br>Sex: 35 ♂ 17♀<br>Age: 3 to 120 days                   | Surgical                   |
| Berry et al <sup>21</sup>      | Randomized Clinical Trial      | n= 60 subjects<br>(CG 28; EG 30)<br>Sex: 40 ♂ 20♀<br>Age: 0 to 4 months | Surgical                   |
| Griffiths <sup>22</sup>        | Clinical Trial, Non-Randomized | n= 215 subjects<br>Sex: NI<br>Age: 0 to 3 months                        | Surgical                   |

Abbreviations: NI, Not Informed; ♂ = male; ♀ = female.

**Table 2** List of articles with the highest score according to the PEDro scale<sup>31</sup>

|                              | External validity<br>(Max = 1) | Internal validity<br>(Max = 8) | Interpretable outcomes (Max = 2) | Total points (Max= 11) |
|------------------------------|--------------------------------|--------------------------------|----------------------------------|------------------------|
| Hogan et al <sup>19</sup>    | 1                              | 4                              | 2                                | 7                      |
| Buryk et al <sup>25</sup>    | 1                              | 6                              | 2                                | 9                      |
| Dollberg et al <sup>10</sup> | 1                              | 8                              | 1                                | 10                     |
| Berry et al <sup>21</sup>    | 1                              | 8                              | 2                                | 11                     |

complaints about alteration in speech. There is also mention of the fact that some individuals with ankyloglossia develop speech normally, compensating the tongue mobility without the need of treatment, others need therapy due to the flaws in the articulation.<sup>4</sup> Another study<sup>9</sup> corroborates this finding, noting that some professionals refer patients before surgery to speech therapy, but this will only be effective if the alteration is not severe, because it is a mechanical disorder.

In the mentioned studies,<sup>4,5,9,23</sup> all subjects underwent tongue exercises after surgery as protrusion, tongue position in the papilla, against the cheeks and lateralization with food.<sup>4,5,23</sup> One study<sup>23</sup> also added the articulation of consonants in the list of exercises, this also shows that the group which performed the 4-flap technique demonstrated greater improvement in articulation than the other. The major effects of lingual exercise are related to tongue mobility.<sup>23</sup> There was

a study<sup>5</sup> which were recommended tongue mobility exercises, but there was no follow-up with a speech therapist.

To assess the subjects before and after surgery is critical to increase the scientific evidence, resulting in greater assertiveness in cases of lingual frenum alteration.<sup>9</sup>

### Scientific Evidence

To check the scientific evidence of the studies, we used the PEDro scale.<sup>31,32</sup> The purpose of the scale is to help researchers identify if the clinical outcomes of therapies applied meet the criteria exposed. 11 checklist items investigate as to internal validity, external validity and results that can be interpreted statistically. All 26 studies were analyzed by this scale, however only 4 scored more, as described in **Table 2**. Studies have many similarities as: study design, population of neonates and infants, use of surgical intervention (frenotomy), main results of less pain felt by mothers and improve in breastfeeding.

### Final Comments

From the selected studies, all resort to surgical option to treat cases of ankyloglossia. In the population of neonates and infants, the most used is the frenotomy by using cold instrument (scissors or scalpel), without the use of anesthetic. In the population of children and adults, techniques and instruments differ among authors. All results show that surgery is the most effective for improvement of symptoms due to ankyloglossia condition. However, the speech does not always fit the expected pattern, which justifies the work together with professional speech therapist, for better results. The theme is scientific important, face to ankyloglossia consequences in aspects of oral functions. It's needed an interdisciplinary team with doctors, dentists and speech therapists trained for assessment, diagnosis and intervention when necessary.

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