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Ankyloglossia and speech disorders: integrative literature

Anquiloglossia e alterações na fala: revisão integrativa da literatura

Revieanquiloglosia y trastornos del habla: revisión integradora de la literatura

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Abstract

Objective: To characterize the speech disorders resulting from ankyloglossia, through an integrative literature review. **Methods and Procedures:** Bibliographic survey carried out in February 2020, delimited language (English, Portuguese, Spanish) and age (from 6 years old). Articles available in four electronic databases were selected: PubMed, SciELO, Scopus, Web of Science. Keywords used: lingual frenum; speech disorders; ankyloglossia. Manuscripts published from 2010 to 2020 were included based on metadata analysis that considered title and abstract to identify the relevance for this research. Studies published over ten years ago, those that did not allow access to the full text, manuscripts repeated due to overlapping of descriptors or diverging from the topic were excluded. **Results and Discussion:** 276 articles were initially identified; after the inclusion and exclusion criteria were applied, 27 studies were considered. The results showed that subjects with lingual frenulum alterations, mainly ankyloglossia, use varied compensatory strategies of lips, tongue and mandible for the production of the phonemes 't', 'd', 'l', 'n', 's', 'z', 'r' and consonant clusters, which may present distortion, substitution and/or omission, as their production is hindered by the short frenulum. Otorhinolaryngologists, orthodontists and speech therapists are recommended to conduct careful clinical examination, allowing for diagnosis, in order to obtain satisfactory results in less time and indication of surgical interventions, when necessary. **Conclusion:** The integrative literature review shows the relationship between ankyloglossia and speech disorders.

Keywords: Lingual frenum; Speech disorders; Ankyloglossia.

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Authors' contributions:
BAS: participated in the design of the study, data collection, analysis and interpretation, and writing of the manuscript.
MLB: participated as supervisor in the design of the study, data analysis and interpretation, and writing of the manuscript.

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Resumo

Objetivo: Caracterizar as alterações na fala decorrentes da anquiloglossia, por meio de revisão integrativa da literatura. **Métodos e Procedimentos:** Levantamento bibliográfico realizado em fevereiro de 2020, delimitado segundo os idiomas inglês, português, espanhol e idade a partir de 6 anos. Foram selecionados artigos disponíveis em quatro bases eletrônicas: PubMed, SciELO, Scopus, Web Of Science. Palavras-chave utilizadas: freio lingual; distúrbios na fala; anquiloglossia. Foram consideradas publicações de 2010 a 2020 mediante análise de metadados, a partir do título e resumo, para identificar pertinência à pesquisa. Foram excluídos estudos publicados há mais de dez anos, que não permitiram acesso ao texto integral, repetidos por sobreposição dos descritores, discrepantes do tema. **Resultados e Discussão:** Foram localizados 276 artigos, que após aplicados os critérios de inclusão e exclusão resultaram em 27. Os resultados encontrados **indicam** que sujeitos com alterações no **frênulo lingual**, principalmente na anquiloglossia, utilizam estratégias compensatórias variadas de lábios, língua e mandíbula para a produção dos fonemas 't', 'd', 'l', 'n', 's', 'z', 'r' e de grupos consonantais, que poderão apresentar distorção, substituição e/ou omissão, por serem de difícil produção com frênulo curto. Aos profissionais otorrinolaringologistas, ortodontistas e fonoaudiólogos é recomendada realização de exame clínico cuidadoso, que possibilite diagnóstico com objetivo de obter resultados satisfatórios em menor tempo e indicação de intervenções cirúrgicas, quando necessárias. **Conclusão:** A revisão integrativa da literatura aponta para a relação entre anquiloglossia e alterações na fala.

Palavras-chave: Freio lingual; Distúrbios na fala; Anquiloglossia.

Resumen

Objetivo: Caracterizar las alteraciones del habla derivadas de la anquiloglosia, a través de una revisión integrativa de la literatura. **Métodos y Procedimientos**: Encuesta bibliográfica realizada en febrero de 2020, delimitada según inglés, portugués, español y edad a partir de 6 años. Bases de datos electrónicas seleccionadas: PubMed, SciELO, Scopus, Web Of Science. Palabras clave utilizadas: frenillo lingual; trastornos del habla; anquiloglosia. Las publicaciones de 2010 a 2020 se consideraron mediante análisis de metadatos, desde el título y el resumen. Se excluyeron los estudios publicados hace más de diez años, que no permitían el acceso al texto completo, repetidos por superposición de descriptores, discrepantes con el tema. **Resultados y Discusión**: Se localizaron 276 artículos, que luego de aplicar los criterios de inclusión y exclusión resultaron 27. Los resultados encontrados indican que los sujetos con alteraciones en el frenillo lingual, principalmente en anquiloglosia, utilizan variadas estrategias compensatorias de labios, lengua y mandíbula para la producción de los fonemas 't', 'd', 'l', 'n', 's', 'z', 'r' y grupos consonánticos, que pueden presentar distorsión, sustitución y/u omisión, porque son difíciles de producir con un frenillo corto. Se recomienda a los otorrinolaringólogos, ortodoncistas, logopedas profesionales que realicen examen clínico cuidadoso, que permita un diagnóstico con el objetivo de obtener resultados satisfactorios en menos tiempo e indicación de intervenciones quirúrgicas, cuando sea necesario. **Conclusión**: La revisión integrativa de la literatura apunta a la relación entre la anquiloglosia y los trastornos del habla.

Palabras clave: Frenillo lingual, Trastornos del habla, Anquiloglosia.

Introduction

The lingual frenulum, also known as frenulum linguae or tongue web, is a mucosal structure that connects the ventral part of the tongue to the floor of the mouth [1,2]. This mucosal fold goes from a more fixed part to a region with greater lingual mobility, and, when impaired, projection of the tongue becomes limited. [3]

According to the protocol published in 2010 [4], the lingual frenulum may be classified, based on its alterations, as: short, when correctly inserted, extending from the lower half of the tongue to the floor of the mouth; anterior, when the superior insertion is fixed from the inferior half towards the tip of the tongue; short and anterior, when there is a combination of the characteristics of the short and the anterior frenula [5].

Ankyloglossia, popularly known as "tongue--tie", is a malformation of the tongue, characterized by an abnormally short and/or thick lingual frenulum. The term "tongue-tie" became known for portraying both the condition of restricted mobility and the lingual frenulum itself [6].

Ankyloglossia is currently defined as a congenital oral anomaly resulting from residual embryological tissues that did not undergo apoptosis as expected during embryonic development, remaining on the underside of the tongue and limiting its movements [13].

Regarding the etiopathogenesis, ankyloglossia is reported in literature as an isolated finding in children[7]. However, there are associations between ankyloglossia and several syndromes, such as X-linked cleft palate, hemifacial microsomia, Beckwith-Wiedemann syndrome, van der Woude syndrome, Opitz syndrome, and orofaciodigital syndrome [8,9,10].

The use of cocaine during pregnancy is also a factor that seems to predispose the newborn to ankyloglossia. According to researchers [11], babies born to mothers addicted to cocaine are three times more likely to develop ankyloglossia, as compared to those of healthy mothers.

Other authors have indicated [12], based on a protocol developed by the Academy of Breastfeeding Medicine, that ankyloglossia may be considered partial or complete. Partial ankyloglossia occurs when the lingual frenulum is short and inelastic, hindering the mobility of the tongue; complete ankyloglossia is when the tongue is tethered to the floor of the mouth [11].

The Lingual Frenulum Evaluation Protocol for Infants, also known as Neonatal Tongue Screening Test [14], mandatory according to the Federal Law number 13.002/2014 [15], is a procedure used in every Brazilian hospital and maternity hospital to identify ankyloglossia in the children born in their facilities. In general, ankyloglossia occurs in 4% to 16% of newborns, with higher frequency in male individuals, in a ratio of 2.5:1 [16].

An abnormally short lingual frenulum may hinder orofacial functions, favouring oral breathing as well as inadequate chewing and swallowing patterns. It also results in difficulty breastfeeding, teething problems, social problems, and speech alterations, the more frequent being: distortions of the alveolar fricative ('s', 'z'), linguodental ('t', 'd', 'n'), alveolar lateral approximant ('l'), and simple alveolar vibrating phoneme in all positions ('r') [7,17].

For speech to be properly produced, there must be a balance between the anatomofunctional structures of the stomatognathic system and the individual's articulatory or motor abilities, allowing them to perform the movements that contribute to its production [17]. Precision of the points of articulation is influenced by presence and position of the teeth, mobility and position of lips, tongue, cheeks and jaw, in addition to adequacy of the intraoral space for phonemic articulation and resonance [18].

The most frequent manifestations in speech that may be caused by lingual frenulum alterations are: articulatory imprecision; distorted alveolar flap causing phoneme omission, substitution or distortion; unclearly produced consonant clusters; and also decreased opening of the mouth during the execution of speech [19,20,21,22].

In speech therapy practice, it is also possible to find distortion of alveolar phonemes and dialectal variants of the 'r' without any impairment of the lingual frenulum. Commonly, in these cases, the orofacial evaluation evidences motor inabilities or incoordination of the tongue during the performance of oral praxis, which in turn are related to the movements necessary for the articulation of sounds, showing the importance of speech therapy aiming at eliminating the identified phonological processes [23].

Speech disorders, in general, have a negative impact on individuals' social life, whether they are children, adolescents or adults, influencing their

self-esteem and how they relate with the environment. Individuals may be target of discrimination simply because their speech is not according to the expected by interlocutors or due to the impairment in their speech intelligibility. These reasons put both their health and quality of life at risk, thus hindering their socialization. In view of these considerations, it is important that lingual frenulum alterations are diagnosed in a timely manner, so their negative impact in the individual's communication is eliminated and/or reduced. The attention of a multiprofessional team that considers both functional and psychosocial deficits may indicate the relevant conducts [1,24].

In recent years, the lingual frenulum has been subject of interest in academic and clinical environments, in several areas of healthcare, regarding the early identification of alterations and respective interventions. Current studies have been focusing mainly on the implications of ankyloglossia in breastfeeding. In this study, however, the interest lays the possible speech impairments in children, adolescents and adults, resulting from abnormally short frenula.

Objective

To characterize speech impairments resulting from ankyloglossia through an integrative literature review.

Methods

Bibliographic search

The study focusing on an integrative literature review was guided by the following question: How are speech impairments resulting from ankyloglossia characterized?

The integrative review has the aim to establish well-defined concepts regarding data search in literature, analysis and interpretation of the results, in addition to provide a comprehensive knowledge of full studies for academic and clinical practice. This study adopted the six stages indicated for integrative literature reviews: 1) selection of a research question; 2) literature search or sampling; 3) data collection; 4) critical analysis of the manuscripts included; 5) discussion of the results, and 6) presentation of the integrative review [(25)].

Broad databases were used with the aim of capturing the greatest possible number of publications on the theme. Hence, manuscripts available on the databases Scientific Electronic Library Online (SciELO), PubMed, SciVerse Scopus, and Web of Science were selected and analyzed in February 2020.

The strategy for the bibliographic search on the databases used the descriptors: "lingual frenum", "speech disorders" and "ankyloglossia". The interlocutor AND and the equivalent descriptors in Portuguese were also used, as established by the Medical Subject Headings (MeSH). The descriptors were crossed as it follows: freio lingual AND distúrbios da fala; freio lingual AND anquiloglossia; distúrbios da fala AND anquiloglossia; lingual frenum AND speech disorders; lingual frenum AND ankyloglossia; speech disorders AND ankyloglossia.

Selection criteria

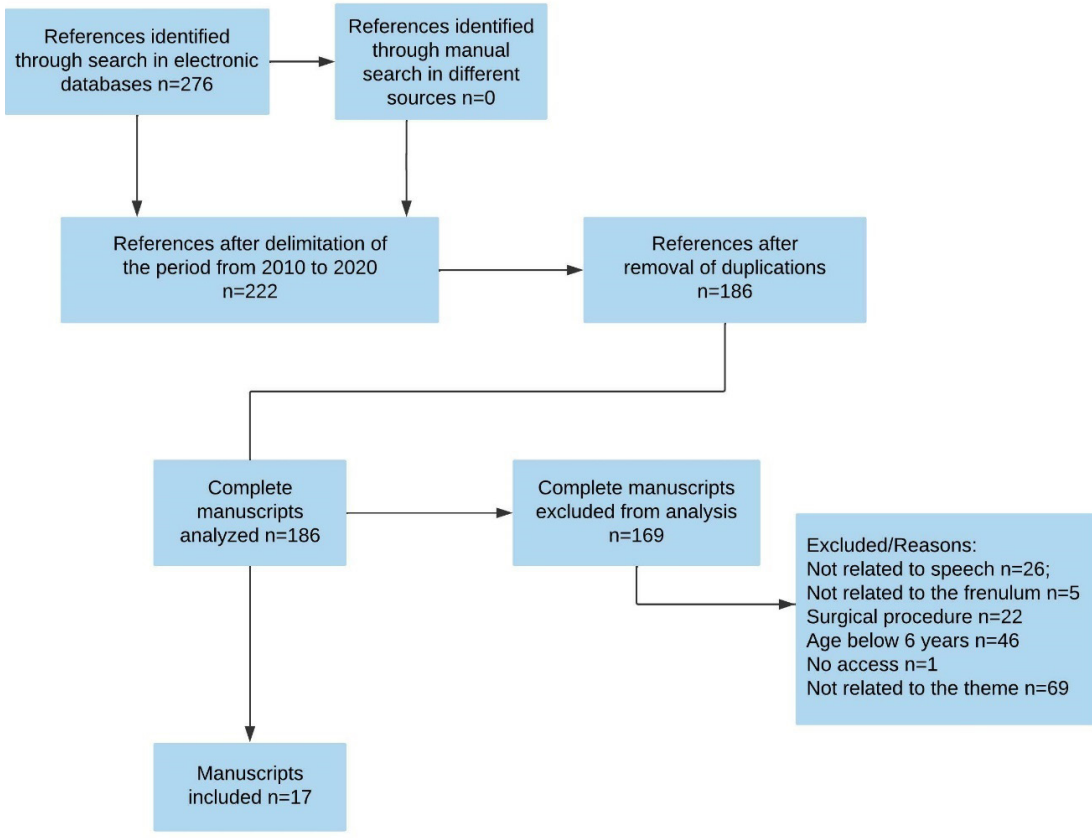
The inclusion criteria for this study were: manuscripts published between 2010 and 2020 through metadata analysis, in English, Portuguese or Spanish, including individuals aged 6 years or older, with speech disorders and lingual frenulum alterations. The exclusion criteria were: studies published before 2010, which could not be accessed in full, were duplicated due to overlapping keywords, those that included subjects younger than 6 years, that did not meet the central theme, did not focus on speech disorders related to lingual frenulum alterations and/or were limited to the research of surgical procedures for ankyloglossia. Both authors participated in every step of the study.

Procedures

The authors conducted a careful analysis of the selected publications by reading title and abstract of the manuscripts on the theme, in order to identify the relevance of the content. The studies initially selected were then accounted for and studied in greater depth, with the aim to identify the relationship between speech and lingual frenulum alterations.

The data search retrieved 276 records, that were inserted into a reference manager (EndNote). When the date of publication was limited to the period between 2010 and 2020, 222 manuscripts were identified. Thirty-six duplications were subsequently removed, leaving 186 studies to be analyzed. The metadata analysis was then conducted, by

reading titles and abstracts to identify the relevance of the manuscripts for this study. Seventeen manuscripts remained for full reading. Figure 1 presents the flow chart of the steps described, according to the *Preferred reporting items for systematic reviews and meta-analyses* (PRISMA) [26].



Source: Research data.

Figure 1. Flow diagram of the selection process

Results

Data regarding the manuscripts included in the study were extracted using the Excel software, version 2011, from Microsoft Office, and are arranged on Chart 1 according to: title, year of publication, journal, data basis, authors, study design, sample, and main results.

Chart 1. Manuscripts included in the study

Title	Year of publication	Journal	Data basis	Authors	Design of the study	Sample	Main results
Ankyloglossia-related changes in the stomatognathic system **(37)**	2012	RGO (Porto Alegre)	Scielo	Morisso et al.	Cross-sectional study	21 participants: 16 male and 5 female; Age range: from 5 to 16 years	Articulatory alterations were found in 95.24% of the sample. The most hindered phonemes were /t/, /d/, /n/, /l/, /s/, /z/. The frequent structural and functional alterations of the stomathognatic system found in individuals with ankyloglossia suggest that their cooccurrence is possible, but surgical intervention is not always necessary.
Attention to rhotacism language problem by oral surgery and vibrostimulatory therapy: a case report **(27)**	2013	Int. J. Odontostomatol.	Scielo	Nevárez et al.	Case report	1 child: male Age: 11 years old	Speech evaluation considered three aspects: phoneme sound level, syllable level, and stimulability score of the phonemes: /s/, /z/, /k/, /g/, /r/. Speech performance was measured in three periods: before surgery, four weeks, and six months after surgery. Combined surgery and functional therapy was a better alternative than only surgery. Combined therapies improved speech and swallowing abilities, as well as the patient's self-esteem.
Lingual frenectomy and alveolar tap production: An acoustic and perceptual study **(34)**	2013	Logopedics Phoniatrics Vocology	Scopus	Camargo et al.	Cross-sectional study	13 Brazilian Portuguese-speaking subjects Age ranged between 7:3 and 47:7 years Mean age: 19:8 years	26 productions of the consonant sound /r/ on the stressed syllable of the word "arara" were analyzed. After the frenectomy, the number of productions of the phoneme did not increase, however, the number of alveolar productions did. Surgery improved the mobility of the tongue, while the alveolar production stayed practically the same.
Surgical Speech Disorders **(38)**	2014	Facial Plastic Surgery Clinics of North America	Web of Science	Shen et al.	Literature review	Age range: infants and children	Diagnosis of speech disorders results from the work of surgeons and speech-pathologists combined. Speech impairments related to ankyloglossia are observed in the production of linguodental (/t/, /d/) and fricative (/z/, /s/ and /th/) phonemes.
Tongue tie: The evidence for frenotomy **(35)**	2014	EarlyHumanDevelopment	Web of Science	Brookes et al.	Literature review	Wide age range: infants, children, adolescents and adults	Children with untreated lingual frenulum alterations can present more articulation errors than those submitted to frenotomy. Reports suggest that prophylactic frenotomy may promote speech development and intelligibility; however, evidence is insufficient for the indication of surgery.
What an Otolaryngologist Should Know About Evaluation of a Child Referred for Delay in Speech Development **(28)**	2014	Jama Otolaryngology--Head & Neck Surgery	Web of Science	Tonn et al.	Literature review	Age range: children	For an otorhinolaryngologist, it is important to understand about speech disorders in children and their causes, so they can indicate the adequate health professional to the family, with the aim to optimize the child's learning. Altered lingual frenulum is among the main causes of speech delay in 2-year-old children or older, although it may not be the primary cause of the delay. The ability to elevate the tip of the tongue up to the alveolar ridge to produce the phonemes /t/, /d/, /l/, /n/, and to project the tongue forward for the /th/ (in English) do not require an extensive projection of the tongue; thus, ankyloglossia is rarely the origin of a speech impairment. The otorhinolaryngologist must be familiarized with speech development milestones in infancy, know the common causes of speech delay, understand the difference between speech and language, and treatment options.
Ankyloglossia and quality of life **(33)**	2015	World JournalofDentistry	Scopus	Roopavathi et al.	Case report	Adolescent, female; 15 years old	Four weeks after frenectomy, the patient reported satisfaction with the improvements in the mobility of the tongue for swallowing food and producing words with the phonemes /t/, /d/, /n/, which contributed to her self-esteem.

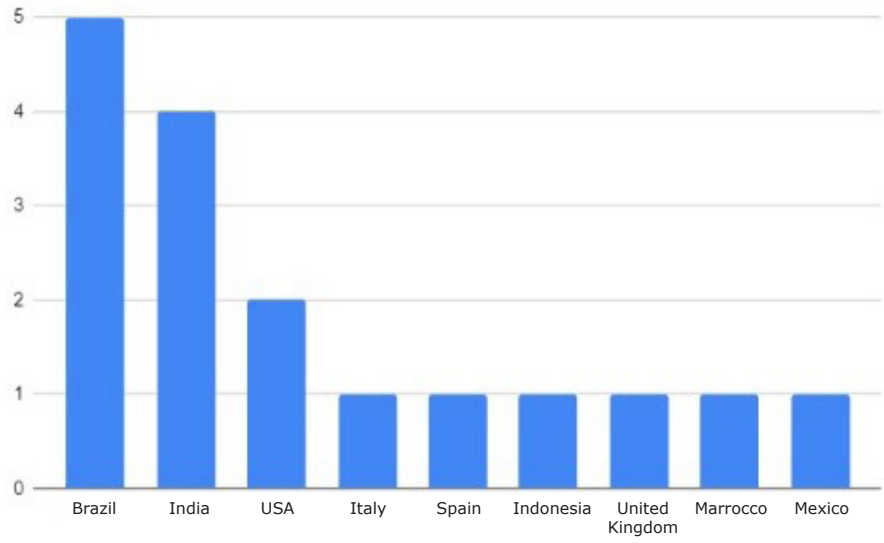
Title	Year of publication	Journal	Data basis	Authors	Design of the study	Sample	Main results
Ankyloglossia with cleft lip: A rare case report **(30)**	2015	J IndianSocPeriodontol	PubMed	Jangid et al.	Case report	Child, male; 12 years old	Teenager with cleft lip and ankyloglossia, with thick lingual frenulum, inserted 3 mm away from the tip of the tongue. Correction of the ankyloglossia improved the phonetics, oral hygiene, and general personality development. The parents were instructed to find a speech pathologist, to improve the speech further.
Interventions in the Alteration on Lingual Frenum: Systematic Review **(36)**	2016	IntArchOtorhinolaryngol	Web of Science	Miranda et al.	Systematic review	Wide age range: infants, children, adolescents and adults	All studies included in the research showed that surgery, regardless of technique, is effective for improvement of the symptoms related to ankyloglossia. Speech, however, not always adjust to the expected pattern, which justifies the need to combine speech therapy for better results.
Alterações de fala relacionadas às alterações do frênulo lingual em escolares **(24)**	2016	Rev. CEFAC	Scielo	Suzart et al.	Cross-sectional study	Cross-sectional study Research group: with lingual frenulum alterations Control group: without lingual frenulum alterations	Orofacial myofunctional evaluation - MBGR protocol. Found statistically significant difference between the groups assessed regarding tone of the tongue and speech. From the 52 children evaluated, 26 (50%) had lingual frenulum alterations, 15 (57.7%) of which were girls. In the group with lingual frenulum alterations: 21 (80.8%) presented reduced tongue tone, 20 (76.9%) presented low tongue posture in the oral cavity, 16 (61.5%) presented articulation problems. Among those with low tongue posture during speech production, 4 (20%) had short anteriorized frenulum, 2 (10%) had anteriorized frenulum, and 14 (75%) had short frenulum. No other statistically significant differences were observed between the groups. The speech alterations observed were substitution of the alveolar liquid phoneme /l/ by /r/, and systematic acoustic distortion of the mild /r/, in consonant clusters with /r/ - specifically /tr/ and /dr/ - and asystematic distortion of /s/ and /z/. It was not possible to determine whether the phonetic alterations were the same for different lingual frenulum alterations.
Labial ankyloglossia: A rare case report **(31)**	2016	ContemporaryClinicalDentistry	Web of Science	Bahadure et al.	Case report	Male; 8 years old	Child with labial ankyloglossia, presenting as one of the main complaints speech fluency difficulties, as he was unable to pronounce the phonemes /t/, /d/, /z/, /s/, /n/, /l/, /r/ and /th/. Lingual frenectomy was performed after labial frenectomy. Fluent speech after 3 years of speech therapy.
Ankyloglossia as an oral functional problem and its surgical management **(8)**	2018	DentMedProbl	Web of Science	Belmehdi et al.	Case report	2 participants; 15-year-old girl and 13-year-old boy	Complaint of restricted tongue movement and speech difficulties; unable to precisely produce /l/, /t/ /d/, /n/, /s/ and /th/ and to move the tip of the tongue towards the upper and lower lips. After frenectomy and immediate speech therapy, a marked improvement was observed in the mobility of the tongue.
The effectiveness of frenotomy in the treatment of ankyloglossia: A case report from Adam Malik General Hospital Medan-Indonesia **(39)**	2018	Bali Medical Journal	Web of Science	Farhat et al.	Case report	Female; 6 years old	Physical exam showed lingual frenulum attached too close to the tip of the tongue, hindering the production of speech sounds that require elevation or extension of the tip of the tongue: /s/, /z/, /t/, /d/, /l/, /r/. According to the case report, frenectomy alone was not able to resolve speech difficulties. The patient presented considerable speech improvement after functional reeducation.

Title	Year of publication	Journal	Data basis	Authors	Design of the study	Sample	Main results
Ankyloglossia inheritance: like father, like son **(32)**	2019	RevPediatrAten Primaria	Web of Science	Ruiz Guzman, Luis et al.	Cross-sectional study	326 groups of siblings: in 133 of them nobody underwent frenectomy in 96 frenectomy was performed in one of the siblings in 91 two siblings underwent frenectomy in 6 three siblings underwent frenectomy Prevalence of ankyloglossia in siblings: 44.9%	Subdiagnosis of ankyloglossia is conditioned by the lack of knowledge of some of its consequences: malposition of the teeth, spinal problems, speech disorders, respiratory problems and apnea, among others.
Compensatory strategies for the alveolar flap [ɾ] production in the presence of ankyloglossia **(40)**	2019	Rev. CEFAC	Scielo	Martinelli et al.	Comparative observational analytic cross-sectional study	88 participants, 44 with ankyloglossia (experimental group - EG) and 44 without ankyloglossia (control group - CG) Age range: from 7 to 42 years. Groups were paired by age and sex: 17 female and 27 male, mean age 13.6 years	Individuals with ankyloglossia presented reduced opening of the mouth, vertical jaw movement, jaw and tongue deviations, depressed center of the tongue, greater use of the laterals of the tongue, deformation of the body of the tongue, and variation of the point of articulation in the production of the alveolar flap. Strategies that included the participation of the lips for production of the alveolar flap and variation of the mode of articulation were observed in only some individuals with ankyloglossia.
Proposal for a myofunctional therapy protocol in case of altered lingual frenulum. A pilot study **(41)**	2019	European Journal of Pediatric Dentistry	Web of Science	Saccomanno et al.	Cross-sectional study	5 male and 1 female children Age range: between 4:5 and 11:7 years Mean age: 7:7 years	The group treated with myofunctional therapy and prescription of exercises by the speech pathologist (to be done at home) yielded positive results. A decrease in the articulatory distortions was observed, especially for the phonemes /t/, /d/, /l/, /n/, /s/ and for consonant clusters. Participation of the lips and mandibular lateralization during verbal articulation were reduced.
Pedodontist's Role in Managing Speech Impairments Due to Structural Imperfections and Oral Habits: A Literature Review **(29)**	2020	Int J ClinPediatr Dent	PubMed	Bommangoudar et al.	Literature review	Several studies including infants, children and adolescents	The production of sounds involves neuromotor components of the oral structure, which is evaluated regarding its level of maturity through the oral diadochokinetic test. Pronunciation of the phonemes /t/, /d/, /n/ requires the tip of the tongue in contact with the palate; pronunciation of /l/, /s/, /z/ requires that the tip of the tongue is facing up, but it may also the pointing down. The /r/ is a phoneme that is usually altered. The movements of the tongue towards the superior teeth are extenuating due to the traction of the lingual frenulum. A person with ankyloglossia often finds it difficult to produce rapid speech clearly. In these cases, a surgical approach should be followed by speech therapy.

Source: Research data.

From the 17 manuscripts included, 5 (29.41%) were from Brazilian researchers, and the remaining were from foreign institutions: 4 (23.53%) from India, 2 (11.76%) from the United States, 1 (5.88%) from Spain, 1 (5.88%) from Italy, 1 (5.88%) from Indonesia, 1 (5.88%) from the United Kingdom, 1 (5.88%) from Morocco, and 1 (5.88%) from Mexico (Graph 1).

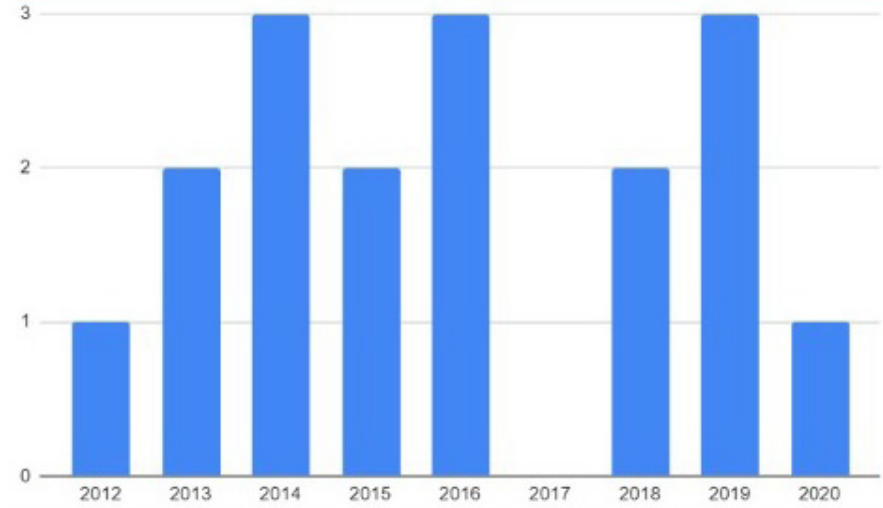
Graph 1. Number of manuscripts published by country



Source: Research data.

From 2010 to 2020, the distribution of the 17 manuscripts included in this research was as it follows: 2012, 1 (5.88%) study; 2013, 2 (11.76%); 2014, 3 (17.65%); 2015, 2 (11.76%); 2016, 3 (17.65%); 2018, 2 (11.76%); 2019, 3 (17.65%); and 2020, 1 (5.88%) (Graph 2).

Graph 2. Number of annual publications



Source: Research data.

The manuscripts included mainly participants from 6 to 12 years of age, even though the selection criteria did not limit studies with adolescent and adult participants.

In general, the studies indicate the presence of speech disorders related to ankyloglossia. Although not all of them described the impairments, most systematically mentioned the phonemes /t/, /d/, /l/, /n/, /r/, /s/, /z/, consonant clusters with /r/, and /th/ (θ) [8,24,27,29,33,37,38,39,40,41].

Discussion

The manuscripts included in this study address the multiple aspects regarding ankyloglossia and its implications to speech.

Ankyloglossia is more prevalent in male individuals [11,16]. However, a study found lingual frenulum alterations in 57.7% of its female participants [24].

The causes of ankyloglossia are not sufficiently known, as it can occur with X-linked, dominant or, in some cases, recessive inheritances [32].

Given that this hereditary malformation affects more than 10% of the population, this study indicates the need to research the transmission mechanisms in larger samples, so that inheritance and pathogenesis can be better understood from an epidemiological point of view [32].

Ankyloglossia may be underdiagnosed due to lack of knowledge of some of its consequences: malposition of the teeth, spinal alterations, speech disorders, respiratory problems and apnea, among others [32].

Although ankyloglossia is an uncommon developmental abnormality, it can be associated with other congenital defects, such as cleft lip and palate. Whatever the anomaly may be, it must be treated effectively and in due course, for the wellbeing of the patient [34].

On the other hand, frequent structural and functional alterations of the stomatognathic system found in individuals with ankyloglossia suggest that other impairments are possible, but surgical intervention is not always necessary [37].

Speech therapy is important after interventions in the lingual frenulum to help the child learn the new points of articulation necessary to correctly produce speech sounds. A surgical procedure, by itself, does not correct speech impairments [39].

Literature shows that individuals with ankyloglossia have presented reduced vertical jaw movement, jaw and tongue deviations, depressed center of the tongue, and variation of the point of articulation in the production of the alveolar flap, differing from the characteristics observed in individuals without ankyloglossia, which were not addressed in this study [40].

Some studies have shown that almost all of their samples had speech unintelligibility and articulatory imprecision in phonemes that require the elevation of the tongue, due to the short lingual frenulum [8,24,37].

Among individuals with ankyloglossia, compensatory strategies were observed in productions considered auditory normal, evidencing the importance of visual and auditory perceptive evaluations carefully conducted by a speech pathologist [39].

The lingual frenulum alteration is mechanic and, therefore, the outcomes of speech therapy are enough if the alterations are not severe. Otherwise, surgical intervention should be indicated in the first place [35,36].

Qualitative improvements are reported for speech and articulation after lingual frenulum interventions[24,35]. Particularly, there is a reduction of articulatory distortions, especially for phonemes 't', 'd', 'l', 'n', 's', 'z', 'r' and for consonant clusters. Compensatory movements, such as participation of the lips and mandibular lateralization during verbal articulation, decrease in comparison to the initial evaluation [35].

Timely diagnosis and intervention for ankyloglossia are essential for the subsequent morphofunctional development of children and adolescents. Health professionals, such as otorhinolaryngologists, orthodontists and speech-language pathologists, should conduct careful and elaborate clinical exams that allow for the diagnosis, so that satisfactory results are obtained in less time and surgical interventions are indicated, when needed. Early intervention provides a better chance for the individual to acquire normal anatomofunctional and communication abilities [8,27].

Some studies included in this research [27,33,36] showed that surgery, regardless of the technique, is effective to improve impairments related to ankyloglossia. However, other studies indicate that surgery alone is not enough to promote improvements in speech pattern, and therefore speech rehabilitation is necessary after the surgical

procedure [8,31,39]. The results indicate the importance of this diagnosis to establish a conduct in cases of speech disorders, with joint participation of dentists, otorhinolaryngologists, and speech-language pathologists [27,28,30,31,36].

According to a study, otorhinolaryngologists may help pediatricians in the complete evaluation of a child with delayed speech development, whether the delay results from an anatomical malformation caused by a short frenulum or not [28]. Another study reported the role of orthodontists in the treatment of speech impairments due to structural alterations and oral habits. These researchers suggested that the orthodontist may observe neuromotor abilities related to speech and refer the individual to the speech-language pathologist [29].

In 2016, a rare case of ankyloglossia was described, where the researchers observed that the frenulum was inserted on the tip of the tongue, which in turn was attached to the lower labial frenum along the floor of the mouth and between the proclined mandibular central incisors. After surgical procedure, the patient was instructed to carry out regular tongue exercises for 3 to 5 minutes, once or twice a day, for 3 to 4 weeks, and to keep adequate oral hygiene. The patient presented improvements in the mobility of the tongue and in speech intelligibility [31].

Studies have shown that untreated children had double the articulation errors of those that underwent surgery. According to the researchers, all the recommended surgical techniques are successful in treating ankyloglossia and require a qualified professional, with broad knowledge and understanding of the several etiologies of complications associated with the liberation. This is due to the importance to provide the ideal post-op care to achieve good clinical outcomes and the patient's general satisfaction. However, speech does not always fit the expected pattern, which justifies the importance of working with a speech-language pathologist to obtain better results [35,36].

The monitoring of a speech-language pathologist is considered fundamental in determining the success of therapy, as individuals with ankyloglossia need intervention, correction, orientation, and supervision in the execution of oral myofunctional and speech exercises during the therapy sessions and at home [33].

The development of an integrative review of the focused literature added relevant information and knowledge from an interdisciplinary and interprofessional point of view regarding the central theme. The discussion, however, indicates a limitation, or rather, the need to further investigate speech-pathology practices for the assessment and rehabilitation of speech disorders resulting from ankyloglossia.

New studies should be conducted, especially focused on speech therapy after surgical intervention.

Conclusions

The results obtained in this review showed that orofacial functions and speech may be impaired depending on the degree of alteration of the lingual frenulum.

The results allow the conclusion that individuals with frenulum alterations, especially ankyloglossia, use several compensatory strategies with lips, tongue, and jaw to produce the phonemes 't', 'd', 'l', 'n', 's', 'z', 'r' and consonant clusters, which may present distortions, substitutions and/or omissions, as their production is hindered by the short lingual frenulum.

The integrative literature review points to a relationship between ankyloglossia and speech disorders.

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