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MÔNICA CARMINATTI

FRÊNULO LINGUAL ALTERADO: IMPLICAÇÕES NA FALA E EFEITOS DA FRENECTOMIA E TERAPIA MIOFUNCIONAL NOS ASPECTOS GERAIS E FUNCIONAIS

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Tese apresentada ao Programa de Pós-Graduação em Odontologia, Nível Doutorado, Universidade Federal do Rio Grande do Sul, como pré-requisito final para a obtenção do título de Doutor em Clínica Odontológica – Odontopediatria.

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Orientador: Prof. Dr. Fernando Borba de Araujo
Coorientadora: Profa. Dra. Erissandra Gomes

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Banca Examinadora

Dr. Fernando Borba de Araujo (Orientador) - UFRGS

Dra. Erissandra Gomes (Coorientadora) - UFRGS

Dra. Irene Queiroz Marchesan - CEFAC

Dra. Monalise Costa Batista Berbert - UFCSPA

Dra. Tathiane Larissa Lenzi - UFRGS

Dra. Fabiane Piva (Suplente) - ULBRA

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RESUMO

A alteração no frênulo lingual pode causar limitação dos movimentos da língua e consequentemente prejuízos na fala. A indicação cirúrgica nas alterações de frênulo lingual deve considerar os aspectos morfológicos e funcionais da língua e a terapia miofuncional pode ser necessária no pós-cirúrgico. Foram conduzidos três estudos para esta tese: o primeiro estudo teve como objetivo investigar a associação entre a anquiloglossia e as alterações de fala; o segundo estudo avaliou a efetividade da frenectomia associada ou não à terapia miofuncional nos aspetos gerais e funcionais da língua de crianças com alteração no frênulo lingual, comparando-as com crianças sem alteração no frênulo lingual, entre seis e 12 anos; e o terceiro estudo investigou as características da fala de crianças com alteração do frênulo da língua. No primeiro estudo desta tese, foi realizada uma revisão sistemática a partir das bases de dados do *PubMed*, *Embase*, *LILACS*, *SCOPUS*, *Web of Science*, *Science Direct*, *Scielo* e da literatura cinza. Dois revisores selecionaram independentemente os estudos, extraíram os dados e avaliaram o risco de viés. A qualidade da evidência foi avaliada utilizando a *Quality Assessment Tools*. Os resultados mostraram que dos 47 estudos potencialmente elegíveis, sete foram incluídos na revisão. Estes foram publicados entre 1999 e 2019 e, utilizaram diferentes instrumentos para o diagnóstico da anquiloglossia e para avaliar a fala. Dos três estudos que investigaram a associação, um não encontrou associação entre as classificações da anquiloglossia e alterações de fala e dois mostraram implicações significativas da anquiloglossia na fala. Concluiu-se que a qualidade da evidência foi considerada muito baixa. Atualmente, a evidência é insuficiente para conclusões definitivas sobre as implicações da anquiloglossia na fala. O segundo estudo desta tese foi um ensaio clínico randomizado controlado realizado com 40 crianças com diagnóstico de alteração no frênulo lingual e 20 com frênulo lingual normal (GCO). Inicialmente (T0), foi aplicado o Protocolo de Avaliação do Frênulo de Língua em todas as crianças. As crianças diagnosticadas com alteração no frênulo lingual submetidas à frenectomia também foram avaliadas 15 dias (T15) e 30 dias (T30) após o procedimento, com o mesmo protocolo. Em T15 as crianças submetidas à frenectomia foram randomizadas em grupo que recebeu exercícios de mobilidade de língua (GE) e grupo que não recebeu exercícios de mobilidade de língua (GC). Ao considerar a realização dos exercícios, o GE melhorou significativamente

para a elevação e mobilidade de língua e outros aspectos da fala como abertura de boca, posição da língua e imprecisão na fala. Considerando o efeito do tempo, na diferença entre T0 e T30, o GE melhorou significativamente comparado ao GC no aspecto da mobilidade da língua (p=0,016), na medida da abertura máxima da boca (AMB) (p=0,024) e AMB com o ápice da língua tocando a papila incisiva (p=0,026). Concluiu-se que a frenectomia associada a terapia miofuncional trouxe benefícios para a mobilidade de língua das crianças. O terceiro estudo desta tese foi um recorte do delineamento do ensaio clínico, em que participaram 34 crianças. Utilizou-se a prova de fala do Protocolo para Avaliação de Frênulo de Língua, acrescido da descrição das estratégias de reparo. A análise dos dados de fala foi realizada por meio de avaliação perceptivo-auditiva. Os resultados mostraram que a maioria das crianças apresentou distorções nas consoantes alveolares [ɾ], [s] e [t] e grupos consonantais com [ɾ]. Entre as demais características analisadas, foi observada redução da abertura de boca. A principal estratégia de reparo utilizada foi a redução de encontro consonantal, seguida da não-realização da líquida não-lateral em posição de coda. Concluiu-se que a maior frequência de alterações de fala foram as de origem fonética, no entanto, estratégias de reparo também foram observadas.

Palavras-chave: Freio lingual, Anquiloglossia, Frenectomia, Terapia Miofuncional, Transtornos da Articulação, Distúrbios da Fala

ABSTRACT

Alteration lingual frenulum may restrict tongue movements and consequently cause speech disorders. Referrals for the surgical treatment of anomalies in lingual frenulum should take into consideration the morphological and functional aspects of the tongue and myofunctional therapy may be needed after surgery. Three studies were conducted for this thesis. The first study aimed to investigate the association between ankyloglossia and speech disorders. The second study assessed the effectiveness of frenectomy, whether associated or not with myofunctional therapy, in improving the general and functional aspects of the tongues of children with alteration on lingual frenulum. They were also compared against children with typical lingual frenulum, between the ages of 6 to 12 years old. The third study investigated the speech characteristics of children with alteration on lingual frenulum. In the first study of this thesis, a systematic review was conducted of the PubMed, Embase, LILACS, Scopus, Web of Science, Science Direct, Scielo and gray literature databases. Two reviewers independently selected studies, extracted data and assessed the risk of bias. The quality of evidence was assessed using quality assessment tools. The results showed that, of the 47 potentially eligible studies, seven were included in the review. These were published between 1999 and 2019 and used different instruments for the diagnosis of ankyloglossia and speech assessment. Of the three studies that investigated the association, one found no association between ankyloglossia classifications and speech disorders while two showed a significant impact of ankyloglossia on speech. It was concluded that the quality of evidence was very low. Currently, there is insufficient evidence with which to draw definitive conclusions about the ramifications of ankyloglossia for speech. The second study of this thesis was a randomized controlled clinical trial with 40 patients diagnosed with atypical lingual frenulum and 20 participants with typical lingual frenulum (CGO). Initially (T0), we used the Tongue Frenulum Evaluation Protocol to assess all the children. Subjects diagnosed with alteration on lingual frenulum and referred to frenectomy were also evaluated 15 days (T15) and 30 days (T30) after the procedure, using the same protocol. At T15, children who had undergone a frenectomy were randomized into two groups: one with tongue mobility exercise therapy (SG) and another without tongue mobility exercise therapy (CG). As for the former group, SG tongue lift and mobility improved significantly, as well as other aspects of speech such as mouth opening,

tongue position and speech accuracy. Regarding the effect of time and the difference between T0 and T30, the SG improved significantly when compared to CG in tongue mobility (p=0.016), maximum mouth opening (MMO) (p=0.024) and MMO with the tip of the tongue touching incisive papilla (TTIP) (p=0.026). We concluded that frenectomy associated with myofunctional therapy benefitted the tongue mobility of children. The third study was a further analysis of one facet of the clinical trial included in this thesis, involving 34 children. We used the speaking test of the Tongue Frenulum Evaluation Protocol and described strategies of speech repair. The analysis of the speech data was performed by means of auditory-perceptual evaluation. Results showed that most speech distortions in the sample group were associated with the alveolar consonants [ɾ], [s] and [t] as well as consonant clusters with [ɾ]. Among the other characteristics analyzed, the function of mouth opening presented a narrower range of motion in the sample. The major repair strategy used by the participants was to simplify consonant clusters, followed by the omission of non-lateral liquid sounds in final position. We concluded that the highest frequency of speech disorders were phonetic in origin. Nevertheless, strategies of speech repair were also observed.

Keywords: Lingual frenum, Ankyloglossia, Frenectomy, Myofunctional therapy, Articulation disorder, Speech disorders

SUMÁRIO

INTRODUÇÃO

A língua humana é uma estrutura muscular complexa, considerada uma das mais importantes estruturas do corpo humano, que participa ativamente de todas as funções orofaciais (STONE *et al.*, 2018). No entanto sua musculatura ainda é pouco compreendida devido a sua complexa anatomia (SANDERS *et al*., 2013). A língua é considerada um hidróstato muscular, e por esse motivo seu volume é constante, constituído por grupos musculares dispostos em várias direções e com capacidade de realizar uma diversidade maior de movimentos comparados a um sistema muscular esquelético, o que os torna particularmente complexos de estudar (KIER; SMITH, 1985).

No ventre da língua encontra-se o frênulo lingual que é uma estrutura dinâmica tridimensional, formado por uma prega central de fáscia, que se estende pelo assoalho da boca, variando na morfologia dentro de um espectro (MILLS *et al.*, 2019). Com base em estudo embriológico, a alteração no frênulo ocorre quando tecidos remanescentes, que deveriam ter sofrido apoptose durante o desenvolvimento embrionário, permanecem na face inferior da língua (KNOX, 2010), causando alterações de inserção e/ou comprimento, podendo comprometer a mobilidade da língua (KROL; KEELS, 2007). O ponto de fixação do frênulo na língua e no assoalho da boca não se modifica ao longo do tempo e sua constituição histológica não permite que se rompa espontaneamente ou seja alongado por meio de exercícios (MARTINELLI *et al.*, 2014).

Alguns estudos genéticos realizados em humanos têm sugerido que a alteração do frênulo da língua tem caráter hereditário (KLOCKARS, 2007; MOROWATI, 2010) ocorrendo mais comumente no sexo masculino (MESSNER *et al*., 2000; RUFFOLI *et al.*, 2005; ATA *et al.*, 2019), com uma proporção de 3:1 (LALAKEA; MESSNER, 2003; KLOCKARS, 2007). A prevalência varia entre 0,1% e 18% (SUTER; BORNSTEIN, 2009; BRAGA *et al.*, 2009; HILL, 2019), justificada pelas diferenças da população estudada e critérios diagnósticos utilizados (SUTER; BORNSTEIN, 2009; HILL, 2019). Ainda, um estudo aponta que em neonatos, a prevalência é maior (1,7% a 10,7%) do que em estudos que investigam crianças, adolescentes ou adultos (0,1% a 2,08%) (INGRAM *et al.*, 2005).

Quando avaliado, o frênulo lingual pode ser diagnosticado como normal ou

alterado, de acordo com os critérios utilizados pelo avaliador (MARCHESAN, 2010). Diante do diagnóstico de frênulo lingual alterado, há uma grande variação anatômica relacionada à sua fixação, que pode estar anteriorizada ou quanto à sua extensão, quando ele é curto, resultando em diminuição da mobilidade da língua (BRITO *et al.*, 2008). Desta forma, os parâmetros para a avaliação estabelecidos têm sido baseados na observação da mobilidade e na aparência da língua associadas à inserção, bem como à fixação do frênulo lingual (KHAIRNAR, 2014; TSAOUSOGLOU *et al.*, 2016; ZAGHI *et al.*, 2019).

Na literatura são encontrados alguns protocolos que avaliam o frênulo lingual (INGRAM et al., 2005; MARTINELLI; MARCHESAN; BERRETIN-FELIX, 2013) em bebês apresentando itens referentes a aparência dos frênulos e avaliação dos movimentos. Instrumentos que se propõem a classificar a alteração do frênulo de língua em crianças maiores também têm sido utilizados para a avaliação (KOTLOW, 1999; RUFFOLI *et al.*, 2005; MARCHESAN, 2010), no entanto não há um protocolo validado para o exame (SEGAL *et al.*, 2007). O uso de protocolos para avaliação é importante para estabelecer parâmetros que permitam o estudo do caso e a definição do tratamento, bem como possibilita a padronização dos exames realizados por diferentes profissionais (GENARO *et al.*, 2009).

No Brasil, pesquisas têm sido realizadas com o instrumento proposto por Marchesan (2010), o Protocolo de Avaliação do Frênulo da Língua (ANEXO), para avaliar o frênulo lingual com base em escores, aplicável para crianças em idade escolar. Além disso, há um consenso entre fonoaudiólogos brasileiros para a utilização deste protocolo na prática clínica. Marchesan (2010) é uma das maiores responsáveis por conduzir a área da Motricidade Orofacial brasileira a conquistar espaço mundial, principalmente no tema de frênulo lingual. O protocolo proposto pela autora é composto por uma anamnese que contém histórico de queixas gerais, realizada com os responsáveis; e um exame clínico, o qual é dividido em provas gerais e funcionais. Nas provas gerais, é verificada a elevação da língua, o percentual referente à medida da abertura máxima da boca e abertura máxima da boca com o ápice da língua tocando papila incisiva e a fixação do frênulo no assoalho da boca e na face inferior da língua. A partir das características de fixação do frênulo no assoalho da boca e na face inferior da língua, o frênulo é classificado em: fixação do frênulo anteriorizada; frênulo de tamanho curto; frênulo curto e com fixação anteriorizada e quando é observada a

fusão do frênulo no assoalho da boca é classificada como anquiloglossia. Na avaliação funcional é analisada a mobilidade da língua, o tônus, a posição durante o repouso e a função da fala. A avaliação fonoaudiológica, por meio do uso deste protocolo específico, permite a classificação do frênulo lingual alterado em curto, anteriorizado, curto e anteriorizado e anquiloglossia, além de avaliar a interferência da alteração nas funções orais (MARCHESAN, 2010).

Os movimentos da língua são muito refinados na espécie humana (SANDERS *et al*., 2013) e quando há limitação em sua mobilidade, as funções orais poderão ser prejudicadas (KHAIRNAR, 2014; TSAOUSOGLOU *et al.*, 2016; NASCIMENTO *et al.*, 2019), dentre elas, a fala (LALAKEA; MESSNER, 2003; MARCHESAN, 2004; CUESTAS *et al.*, 2014; SUZART; ITO, 2015; CARVALHO, 2016; NASCIMENTO *et al.*, 2019). No entanto, embora a relação da alteração do frênulo da língua com a fala seja relatada desde o tempo do filósofo Aristóteles (PATEL *et al.*, 2018) ainda existem muitos questionamentos sobre a temática.

Os fonoaudiólogos que trabalham com crianças buscam formas específicas de avaliar o frênulo da língua para detectar se este dificulta as funções orais, especialmente a produção correta da fala (MARCHESAN, 2010). Os movimentos da língua durante a fala estão entre as atividades motoras mais complexas e parecem ser únicos entre os mamíferos (SANDERS *et al*., 2013). Cada consoante e cada vogal exige um formato e uma posição diferente da língua no interior da cavidade oral, ou seja, a língua está em constante movimento durante a fala e deve ter agilidade e precisão suficientes para sua adequada produção (XING *et al.*, 2016). Entender a especificidade das alterações de fala, bem como os fatores que interferem na sua produção, amplia o campo de atendimento do fonoaudiólogo e melhora a qualidade de vida dos pacientes.

Há estudos que observaram as alterações de fala relacionadas às alterações de frênulo lingual de origem fonética. As distorções com maior ocorrência nestes estudos são no grupo consonantal com [r] (especificamente o [tr] e [dr]) e [l] (TEJA-ÁNGELES *et al.*, 2014; SUZART; CARVALHO 2016), no flape alveolar [r] (GONÇALVES; FERREIRO, 2006; BRAGA *et al.*, 2009; CAMARGO *et al.*, 2013; CUESTAS *et al.*, 2014; TEJA-ÁNGELES *et al.*, 2014; ITO *et al.*, 2015 SUZART; CARVALHO, 2016) e nas alveolares [s] [z] (BRAGA *et al.*, 2009; CAMARGO *et al.*, 2013; TEJA-ÁNGELES *et al.*, 2014; ITO *et al.*, 2015), [t], [d], [l] e [n] (GONÇALVES; FERREIRO, 2006; BRAGA *et*

al., 2009; TEJA-ÁNGELES *et al.*, 2014; CUESTAS *et al.*, 2014). Observa-se, também, adaptações/compensações na articulação dos sons, como imprecisão articulatória, velocidade aumentada de fala, abertura da boca reduzida, desvios de lábios e de mandíbula, e posição baixa de língua na cavidade oral, com participação atípica de suas margens laterais (MARCHESAN, 2004; CAMARGO, 2013; SUZART; CARVALHO, 2016).

Ainda há alguns estudos que verificaram a presença de omissões (MARCHESAN, 2004; BRAGA *et al.*, 2009; ITO *et al.*, 2015) e substituições envolvendo o fonema /r/ (MARCHESAN, 2004; ITO *et al.*, 2015) na fala de crianças com alteração de frênulo lingual, as quais constituem alterações fonético-fonológicas. Sabe-se que a organização linguística e a produção dos fonemas podem ser influenciadas pela capacidade motora e pela integridade dos órgãos fonoarticulatórios (WETZNER, 2005; GONÇALVES; FERREIRO, 2006; BRAGA *et al.*, 2009; MONTEIRO *et al.*, 2009; SILVA *et al.*, 2009; MARINI *et al.*, 2011; MEZZOMO *et al.*, 2011; GUBIANI *et al.*, 2015) e quando essas estruturas apresentam alterações de tonicidade e de praxia, podem interferir na produção dos fonemas (CASARIN *et al.*, 2006). Assim, a imprecisão articulatória também pode interferir no nível fonológico, uma vez que a fala requer coordenação complexa e planejamento de movimentos de lábios e de língua para a produção dos sons (MARINI *et al.*, 2011).

Outros estudos relatam que as alterações na fala são raras ou que é difícil afirmar que a causa das alterações advém do frênulo lingual (DOLLBERG *et al.*, 2011, WEBB *et al*., 2013). A diversidade de opiniões, assim como as divergências entre os autores, provavelmente provém da inexistência de parâmetros comuns para avaliação (MARCHESAN, 2010).

As cirurgias de frênulo lingual também são motivo de discussões em relação à indicação ou não e em que momento deve ser realizada ou qual a técnica mais eficaz. A frenectomia (excisão completa do frênulo, incluindo sua inserção ao osso adjacente), a frenotomia (incisão do freio, resultando em uma remoção parcial) e a frenuloplastia (corte ou remoção do frênulo por meio de métodos variados para a correção da situação anatômica) são as principais opções de tratamento cirúrgico para as alterações de frênulo lingual (KNOX, 2010; CHAUBAL, DIXIT, 2014). Estudos têm destacado que a indicação de intervenção cirúrgica deve considerar a avaliação dos aspectos morfológicos e funcionais da língua e esta deve promover melhora nesses

aspectos (OLIVI *et al.*, 2012; CUESTAS *et al.*, 2014). Não há evidências científicas que favoreçam um ou outro método cirúrgico (SUTER; BORNSTEIN, 2009; KHAIRNAR, 2014). No entanto, a frenectomia é o procedimento que tem sido utilizado para a liberação do frênulo lingual (KNOX, 2010). Uma revisão sistemática observou em alguns estudos que utilizaram técnicas cirúrgicas variadas, melhora nos aspectos relacionados à mobilidade de língua, como elevação e protrusão, após a intervenção (SUTER; BORNSTEIN, 2009). Entretanto, outro estudo observou após o procedimento cirúrgico que o melhor resultado foi para o movimento de protrusão e o pior para o de elevação de língua (ITO *et al.*, 2015). Em alguns casos, principalmente os de maior gravidade, pode ocorrer a necessidade de um período maior para a recuperação do movimento de elevação da língua. Para esses casos, exercícios de mobilidade de língua poderiam ser indicados (SUTER; BORNSTEIN, 2009). Outra revisão sistemática aponta que a cirurgia é eficaz para melhorar os sintomas devido à alteração do frênulo lingual. No entanto, a fala nem sempre se enquadra no padrão esperado, o que justifica o trabalho em conjunto com o fonoaudiólogo para obter melhores resultados (MIRANDA; CARDOSO; GOMES, 2016) .

A intervenção fonoaudiológica no pós cirúrgico das alterações de frênulo lingual é documentada em alguns estudos (NAVARRO; LOPES, 2002; MESSNER; LALAKEA, 2002; OLIVI *et al.*, 2012; HELLER *et al.*, 2005; WEBB *et al.*, 2013; BELMEHD *et al.*, 2018). Um estudo de série de casos observou que a substituição e a omissão podem melhorar após a cirurgia e evoluir para distorção, no entanto exercícios de língua no pós operatório e terapia de fala seriam necessários para as crianças (ITO *et al.*, 2015). A recomendação de exercícios isotônicos de língua e de técnicas para a produção dos fonemas demonstra ganhos em relação à melhora da mobilidade proporcionando maior efeito terapêutico (MESSNER; LALAKEA, 2002; HELLER *et al.*, 2005). Entretanto, as evidências científicas relacionadas ao benefício da indicação de mioterapia após a correção cirúrgica de frênulo lingual são escassas (SUTER; BORNSTEIN, 2009).

A inconsistência ou não regularidade nos achados justifica o aprofundamento dos estudos sobre a condição do frênulo e da fala, bem como entre esta condição e as habilidades práxicas da língua e formas de tratamento. Considerando os apontamentos acima e as divergências encontradas na literatura sobre a implicação do frênulo lingual na fala, a primeira das três propostas de estudo para esta tese é realizar

uma análise das evidências disponíveis para a discussão sobre este tema, uma vez que as alterações de fala podem provocar potenciais implicações nas relações do indivíduo com o meio (LALAKEA; MESSNER, 2003; BRAGA *et al.*, 2009), sua autoimagem e aprendizagem (RABELO *et al*., 2011; GOULART, CHIARI, 2014), repercutindo de maneira negativa na saúde e qualidade de vida (RABELO *et al*., 2011; GOULART, CHIARI, 2014). Uma análise aprofundada da qualidade metodológica dos estudos existentes pode auxiliar na tomada de decisão na prática clínica.

A justificativa para a segunda proposta de estudo desta tese, na literatura consultada, a maioria dos artigos publicados sobre tratamento das alterações de frênulo lingual em crianças apresentam limitações metodológicas que incluem tamanho da amostra, histórico não homogêneo em relação à idade, técnica cirúrgica variada no mesmo estudo, características dos aspectos anatômicos e/ou funcionais ausentes (MESSNER; LALAKEA, 2002; LALAKEA; MESSNER, 2003; HELLER *et al.*, 2005; WEBB *et al.*, 2013; ITO *et al.*, 2015; BELMEHD *et al.*, 2018). A investigação é importante uma vez que um número crescente de pacientes e profissionais de saúde buscam informações baseadas em evidências para o tratamento das alterações de frênulo lingual em crianças; no entanto, há escassez de evidências de qualidade nos estudos (SUTER; BORNSTEIN, 2009; OLIVI *et al.*, 2012; JIN *et al.*, 2018).

O argumento para a terceira proposta de estudo para esta tese é, como mencionado anteriormente, que as distúrbios de fala relacionadas às alterações de frênulo lingual mais estudadas são as de origem fonética. No entanto, há escassez de estudos investigando as alterações de fala no nível fonológico em crianças com alteração no frênulo lingual, as quais podem estar relacionadas às dificuldades no aspecto fonético (MARCHESAN, 2004; SUZART, CARVALHO, 2016). A análise de alterações fonológicas em crianças com alteração de frênulo lingual justifica-se pelas possíveis dificuldades na tradução do conhecimento fonológico em uma ação motora adequada para a realização do fone/sílaba apropriado.

2 OBJETIVOS

Esta tese teve como objetivos:

- Realizar uma revisão sistemática para verificar se há associação entre alterações de frênulo lingual e as alterações de fala;
- Realizar um ensaio clínico randomizado controlado para avaliar a efetividade da frenectomia associada ou não à terapia miofuncional nos aspetos gerais e funcionais de crianças com alteração no frênulo lingual, comparando-as com crianças sem alteração no freio lingual, entre seis e 12 anos;
- Por meio do recorte do estudo com delineamento de ensaio clínico, investigar quais são as características da fala de crianças entre seis e 12 anos que apresentam alteração do frênulo da língua.

3.1 ARTIGO 1

Artigo formatado nas normas de *International Journal of Paediatric Dentistry*

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ANKYLOGLOSSIA AND SPEECH DISORDERS: A SYSTEMATIC REVIEW

Running Title: Ankyloglossia and Speech Disorders

Mônica Carminatti[1]
Rafaela Soares Rech[2]
Erissandra Gomes[2]
Fernando Borba de Araujo[1]

[1]Post-Graduate Program in Pediatric Dentistry, Federal University of Rio Grande do Sul, Brazil
[2]Faculty of Dentistry, Federal University of Rio Grande do Sul, Brazil

Author contributions:
M.C. involved in idea, performed the methodology and wrote the manuscript; R.S.R. performed the methodology and proofread the manuscript; E.G. involved in idea and proofread the manuscript; F.B.A. proofread the manuscript.

Corresponding author:
Prof. Erissandra Gomes
Faculty of Dentistry, Federal University of Rio Grande do Sul
Ramiro Barcelos 2492, 90035-003, Porto Alegre, RS, Brazil
Phone number: +55 51 3308 5176
E-mail: erifono@hotmail.com

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Conflict of interest
The authors declare no conflict of interest

ABSTRACT

Ankyloglossia - also referred to as tongue-tie - is a congenital anomaly that may limit tongue movements and consequently affect speech. However, there is no consensus on the precise nature of these repercussions. The aim of this systematic review was to investigate the association between ankyloglossia and speech disorders. The PubMed, Embase, LILACS, Scopus, Web of Science, Science Direct and Scielo databases were queried, as well as gray literature. No language or date filters were applied. Two reviewers independently selected studies, extracted data and assessed the risk of bias. The quality of evidence was evaluated by using Quality Assessment Tools. From 938 potentially eligible studies, 47 were full text and 7 have been included in this review. These studies were carried out in America, Asia and Europe and they were published between 1999 and 2019. Additionally, different tools were used to diagnose and assess speech affected by ankyloglossia. Of the three studies that studied this association, one found no relation between the classifications of ankyloglossia and speech disorders while two found a significant effect of ankyloglossia on speech. The quality of evidence was considered very low. Current evidence is insufficient to draw definitive conclusions about the effects of ankyloglossia on speech.

Keywords: Ankyloglossia; Lingual frenulum; Articulation disorder; Speech disorders; Systematic review

INTRODUCTION

Ankyloglossia - also referred to as tongue-tie - is a congenital anomaly that occurs when tissue that should have undergone apoptosis during embryonic development remains attached to the underside of the tongue, causing changes in attachment and/or tongue length[1]. Males are more predisposed to this condition than females[2], at a ratio of 3:1[3,4]. Prevalence varies between 0.1% and 18%[5-7], depending on the population studied and the diagnostic criteria used[5,7]. Restrictions in tongue movement caused by ankyloglossia can affect different oral functions in various ways[3,8-14]; speech is one of these[3,8,12,15].

Although the relationship between ankyloglossia and speech disorder has been reported since Aristoteles' time[16], there are still many questions on the subject. Regarding changes in speech production, some studies have reported altered articulation or strategies of adaptation or compensation[6,8,12,15]. However, others have found different results as to the impact of ankyloglossia on oral functions[5,6,17-21]. This diversity of opinions, as well as the differences between authors, probably stems from an absence of common parameters for assessment[22].

Our study is therefore justified since speech problems may potentially cause negative repercussions on self-image[3,6] and learning[23,24], as well as on general health and quality of life[17,23,24]. Thus, an examination of the available evidence concerning the issue of whether or not ankyloglossia affects speech is necessary. Moreover, decision-making in clinical practice can be hindered by the ongoing absence of a thorough assessment of the methodological quality and level of evidence of existing studies.

No systematic review was found in health care literature addressing the question as to whether ankyloglossia is associated with speech disorders in children, adolescents or adults.

Thus research context, the objective of this systematic review was to determine whether there is an association between ankyloglossia and speech disorder.

METHODS

Protocol and registration

This systematic review was conducted according to the Cochrane Collaboration[25] reported according to PRISMA (Preferred Reporting Items for

Systematic Reviews and Meta-Analyses) guidelines[26] and submitted for registration on PROSPERO (International prospective register of systematic reviews: http://www.crd.york.ac.uk/PROSPERO/).

Eligibility criteria

Inclusion criteria took into account observational study designs (cohort, case-control, cross-sectional, case reports and case series) with pediatric, adolescent and adult populations of both sexes diagnosed with ankyloglossia. Furthermore, some form of speech assessment must have been performed. No language or date filters were applied. Comments, letters, books, editorials, announcements, clinical trials, reviews, literature reviews, systematic reviews, conference abstracts and duplicate studies were excluded.

Studies addressing exposure to ankyloglossia were taken into consideration, regardless of the use of a comparison group or not. In the case of studies which included associated types of exposure, only data concerning participants exposed to ankyloglossia were considered for this review. The main result of our study was the occurrence of speech impediments, protocol-based assessments, clinical assessments, questionnaires and clinical history reviews.

Sources of information and research strategy

In order to formulate our proposition, the PECO concept was used (population, exposure, comparator and outcomes) and the following structured question was framed: Is there is an association between ankyloglossia and speech disorders?

Our search strategy was initially established for the PubMed database using MeSH (Medical Subject Headings), DeCS (Health Sciences Descriptors) and EMTREE (Embase Subject Headings) keywords concerning exposure of interest and results. The Boolean operator OR is used to combine the concept of the terms in each CEE; the AND operator is used to combine the different PECO concepts (population, exposure, comparator, outcome). A sensitive search strategy was adapted for other databases: Embase, Latin American Literature of Health Sciences of the Americas and Caribbean - LILACS, Scopus, Web of Science, ScienceDirect and The Scientific Electronic Library Online - SciELO. The Gray Literature Report was used to search for gray literature. The full search strategy and search terms are presented in Table 1. Studies indexed up to July 2019 were included.

In order to reduce possible selection bias, the references of included studies and other bibliographic resources on the theme of ankyloglossia in children, adolescents and adults, with a speech outcome, were taken into account as sources of additional data. Research results from multiple databases were cross-referenced to detect and eliminate duplicates using Endnote X7 (Thomson Reuters, Philadelphia, Pennsylvania).
Study selection and data extraction

Firstly, two reviewers (M.C. and R.S.R.) independently examined the results of the electronic search by reading the title and abstract in order to identify studies that met the eligibility criteria. Decisions were listed as "eligible", "deleted" or "uncertain". Any disagreement between the reviewers was discussed.

Eligible or uncertain articles were read independently and in full by the two reviewers (M.C. and R.S.R.) and selection criteria for inclusion were put into practice. The reasons for the exclusion of these texts at this phase were recorded. A third reviewer (E.G.) was consulted on any disagreements regarding eligibility.

The following information was extracted from eligible studies: author(s), year of publication, country of origin, purpose, study design, number of subjects, population characteristics, exposure characteristics, outcome measures and results.

Assessment of risk of bias

A blind and independent assessment of the quality of the research studies was carried out by two authors (M.C. and R.S.R.) using the quality assessment tools[27] specific to observational studies. The methodological quality of evidence was classified as good, fair or poor, in accordance with the 14 criteria recommended in the instrument. If the answer to questions 7, 8, 9, 10, 11 and 14 was "yes", the study was classified as having a low risk of bias. This classification was also extended to studies that satisfied at least 50% of the tool items. Disagreements between the reviewers were resolved by the third reviewer (E.G.).

Summary of Results

Included articles were moved to a data extraction base, following a standard form in Excel© (Microsoft Corporation, USA). Data were summarized in tables, according to the nature of the outcome measures. For quantitative measurements of the results, average values, frequency, standard deviation and p-values were recorded,

whenever possible. Besides this, data were summarized in tables when the methods were similar enough to allow this. No meta-analysis was performed due to the heterogeneity of the findings and a lack of access to the raw data from the speech samples of participants.

RESULTS

Study selection

A total of 1264 articles were retrieved from databases and literature searches. After eliminating duplicates, 938 records remained. Upon analyzing the titles and abstracts, 891 studies were excluded. Therefore, 47 studies met the inclusion criteria at the full-text phase of the review. After reading the full studies, seven[6,8,12,17,18,19,21] met the criteria for data extraction and analysis (Figure 1). The other 40 articles were excluded for various reasons, which included no exposure factor (ankyloglossia) and/or outcome (speech disorders). Similarly, we also excluded studies investigating the effect of surgery on changes in the lingual frenulum as well as research designs and methodology incompatible with the inclusion criteria.

Characteristics of included studies

The characteristics of the population, lingual frenulum and methodological design of the selected studies are shown in Table 2. These seven studies were conducted in America, Asia and Europe and were published between 1999 and 2019. Six were cross sectional studies[6,8,12,17,18,21] and one was a case-control study[19]. The sample sizes ranged from 23 to 1402 subjects whose ages ranged from 20 days to 62 years and 10 months. With regard to the lingual frenulum characteristics, while three studies utilized classification predicated on point of attachment[6,8,12], another study assessed normal or impaired tongue protrusion but did not refer to a classification tool[21]. One more study used a scale of severity to analyzed the length of the lingual frenulum[18] while two other studies did not report on this particular characteristic[17,19]. Measures of speech were presented in various ways: through the use of protocols, questionnaires, chart reviews and clinical histories. Only three studies mentioned the evaluators of speech function[6,12,19].

The aims, outcome measures and the results of the included studies are presented in Table 3. Concerning the outcomes presented by the authors, only four

described the frequency of speech changes in patients diagnosed with ankyloglossia[17-19,21] two studies reported both the association between speech disorders and ankyloglossia as well as frequency, with the aid of findings regarding statistical significance[8,12]. Another study investigated the association between speech changes and ankyloglossia classifications and reported a lack of statistical significance[6].

Authors have suggested a relation between ankyloglossia and speech, often observing the omission of the phoneme /r/, the substitution of this phoneme[8], the substitution of /l/ with /r/[12], and distortions of {R}, consonant clusters with /R/[8,12], /s/ and /z/[8,12]. Speech was classified as distorted and/or inaccurate, and descriptions of altered speech production included decreased spacing between maxilla and mandible, excessive anterior or lateral mandibular movements, or excessive salivation[8]. The substitution the phoneme /l/ with /r/ consonant and distortion in consonant clusters with /r/ was associated with a short lingual frenulum whereas distortions of /s/ and /z/ were associated with anterior lingual frenulum[12].

Risk of bias assessment studies

All seven studies were classified as being of poor quality (Table 4). Most of the studies did not clearly specify or define the populations of their studies[6,8,12,18,19,21]. No study justified sample size. Five studies presented problems regarding selection, recruitment and the inclusion and exclusion criteria of their methodology[8,18,19,21]. Researchers from two studies did not perform an adjusted statistical analysis of the association they established between ankyloglossia and speech impediments[8,12].

DISCUSSION

The methods used to diagnose ankyloglossia varied among studies. However, the three surveys performed in Brazil[6,8,12] used the same criteria for the diagnosis of this anomaly. This finding is related to a consensus among Brazilian speech language pathologists to use a classification schema that describes the lingual frenulum as normal, short, anterior or anterior and short[8]. Other studies have used different methods that comprehend range of motion[18] the ability to protrude the tongue beyond the red border of the lower lip and palpation of the floor of oral cavity[21]. The diagnostic criteria used in the studies were not validated and there is no prospective comparison of their methods with a proposed standard. None of the studies evaluated the internal

or external validity of their diagnostic methods[28].

The substantial difficulty in comparing studies that address problems and results associated with ankyloglossia arises from an absence of common parameters for assessment and diagnosis, as well as lack of awareness about the consequences of deformities in the lingual frenulum[5,16,22]. Given that ankyloglossia is a multidisciplinary problem which involves dentists, speech language therapists and doctors, a universally accepted method of assessment, duly standardized and validated, is imperative[5]. Upon assessing a lingual frenulum, similar classifications and professional recommendations would make it easier for patients and their relatives to feel confident and safe in accepting treatment decisions. Patients can often feel insecure if presented with contradictory opinions about the same problem[29]. Future research should focus on developing universal tools for classifying ankyloglossia and its effect on speech acquisition[30].

All of the studies in our review used diverse methods of measurement to assess speech disorders. Brazilian studies used the speech aspect item from an orofacial motor assessment instrument[12] and a children's language test[6]. Other studies used articulation tests[19], questionnaires[17] or performed a review of patient medical history[18] and records[21]. The heterogeneity among these assessment tools made comparing results a challenging task.

As for the individual characteristics of the studies that were included in this review, Sanchez-Ruiz *et al*.[17] used a questionnaire to conduct interviews with parents or guardians over the telephone. In this manner, they traced the frequency of speech disorders accompanying cases of ankyloglossia. Karabulut *et al*.[18] analyzed the clinical data of subjects who had undergone a frenotomy procedure for ankyloglossia. Daggumati *et al.*[21] performed a retrospective review of medical records to identify patients diagnosed with ankyloglossia. The retrospective nature of studies such as Daggumati *et al.* [21], Karabulut *et al.*[18] and Sanchez-Ruiz *et al.*[17] may imply methodological flaws such as incomplete data and variability in patient-reported information. Moreover, the study[17] that used a questionnaire by telephone to assess outcome by telephone measured the perception parents or guardians had of the speech problems of their children. Additionally, recall bias may have influenced the results. What is more, while one study investigated the frequency of speech disorders in children over one year of age [18], another investigated children three years or older[17];

these are age ranges in which speech errors such as omission and substitution are common - regardless of deformities of the tongue - because these children are still in the phase of language acquisition.

Studies in which speech was evaluated by speech language therapists such as Braga *et al*.[6] did not find an association between lingual frenulum classifications and speech impediments. However, their study did not have a control group and this makes the results less convincing. Dollberg *et al*.[19] analyzed the speech intelligibility of children who had been diagnosed with ankyloglossia as infants; results were compared to those of a group of children with no history of ankyloglossia. The authors found that children with a history of ankyloglossia produced more speech errors that the latter group. Nevertheless, even though a control group was included, no comparison was made with a third, untreated group with a history of ankyloglossia in order to prove association. Furthermore, the patient sample was small, making results too inconsistent for affirmations of quality.

There is a lack of specific standardization regarding tools that evaluate speech, as can be seen from the different methods used in the studies. Therefore, there is a need for greater investment in common speech assessment protocols for patients with ankyloglossia; this implies not only design, but also translation, adaptation and validation procedures for different countries. Specific protocols favor improved common clinical decision-making and recommendations by experts and, by extension, better clinical practice based on evidence of quality.

Two Brazilian studies[8,12], suggest an association between ankyloglossia and speech, but they are of low methodological quality and were not able to present a consistent conclusion. When considering the types of changes in speech, omissions[8], substitutions[12] and distortions were observed[8,12]. The most affected phonemes were /r/, /s/ and /z/. Marchesan [8] also reported an association between altered lingual frenulum and speech disorders, but did not identify the functional aspect that was observed. Neither was an age group defined, which may have been an important confounding factor in the analysis of results. Suzart *et al*.[12] noted an association between study and control groups with respect to tongue posture and articulation. However, there was no justification given for the sample size and the assessor was not blinded to the exposure status of the participants.

This review highlights the vast differences in ankyloglossia management

strategies, which are affected by factors such as profession, geography, personal experience and number of cases. Furthermore, when considering the divergence of the speech results among studies, one must take into account the different instruments used for evaluations. Due to a lack of a standardized assessment protocol, and depending on the method used, evaluators may have considered one or more characteristics a speech impediment (such as omissions, substitutions, distortions or functional inaccuracies). No sample size calculations, missing or inconsistent blinding, non-existent control groups, heterogeneity in the diagnosis of ankyloglossia and non-standard speech assessments are highlighted in our analysis of the included studies. Therefore, upon analysis, the quality of evidence in these studies was classified as poor.

Due to the limited number of studies included and the heterogeneity of the results, a quantitative evaluation was not explored. Although the findings of this review present evidence of low quality, it is still considered relevant for discussion, especially to alert researchers about this subject, which has been widely spread even without published studies of quality to answer the research question.

Therefore, with the aim of building reliable evidence, further research with adequate methodological quality is needed for assessing the association between ankyloglossia and speech disorders. This should be supported by standardardized and validated diagnostic methods. In conclusion, there is insufficient evidence to say whether a diagnosis of ankyloglossia implies speech disorders in children, adolescents and adults.

Why this paper is important for pediatric dentists

However, there is still insufficient evidence to prove that there is an association between this anomaly and speech disorders in children, adolescents and adults.

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Table 1. Database and word truncation

Database	Search descriptors	Article numbers Date
Pubmed	((((((((((("Lingual Frenum"[Mesh]) OR "Lingual Frenums"[Mesh]) OR “Lingual Frenulum” [Mesh]) OR "Lingual Frenulums"[Mesh]) OR "Ankyloglossia"[Mesh]) OR "Ankyloglossias"[Mesh]) OR "tongue"[Mesh]) OR “Tongue Tie” [Mesh]) OR “Tongue Ties” [Mesh]) OR "Tongue Diseases"[Mesh]) OR "Partial Ankyloglossia"[Mesh]) OR "Partial Ankyloglossias"[Mesh])) AND ((((((“Articulation Disorders"[Mesh]) OR "Articulation Disorder"[Mesh]) OR "Speech Articulation Disorder"[Mesh]) OR "Speech Disorders"[Mesh]) OR "Speech Sound Disorder"[Mesh]) OR "Speech Sound Disorders"[Mesh])	643 07/31/2019
Embase	('tongue frenulum':ti,ab,kw OR ankyloglossia:ti,ab,kw OR tongue:ti,ab,kw OR 'tongue disease':ti,ab,kw OR 'partial ankyloglossia':ti,ab,kw) AND ('articulation desorder':ti,ab,kw OR 'speech articulation disorders':ti,ab,kw OR 'speech disorder':ti,ab,kw OR 'speech sound disorder':ti,ab,kw)	21 07/31/2019
Lilacs	(tw:((lingual frenum) OR (lingual frenums) OR (lingual frenulum) OR (lingual frenulums) OR (ankyloglossia) OR (ankyloglossias) OR (tongue) OR (tongue tie) OR (tongue ties) OR (tongue diseases) OR (partial ankyloglossia) OR (partial ankyloglossias))) AND (tw:((articulation disorder) OR (articulation disorders) OR (speech articulation disorder) OR (speech disorders) OR (speech sound disorder) OR (speech sound disorders)))	102 07/31/2019
Scopus	(TITLE-ABS-KEY ("Lingual Frenum" OR "Lingual Frenums" OR "Lingual Frenulum" OR "Lingual Frenulums" OR ankyloglossia OR ankyloglossias OR "Tongue Tie" OR "Tongue Ties" OR "Tongue Diseases" OR "Partial Ankyloglossia" OR "Partial Ankyloglossias") AND TITLE-ABS-KEY ("Articulation Disorders" OR "Articulation Disorder" OR "Speech Articulation Disorder" OR "Speech Disorders" OR "Speech Sound Disorder" OR "Speech Sound Disorders"))	281 07/31/2019
Web of science	TS=("Lingual Frenum" OR "Lingual Frenums" OR "Lingual Frenulum" OR "Lingual Frenulums" OR "Ankyloglossia" OR "tongue" OR "Tongue Tie" OR "Tongue Ties" OR "Tongue Diseases" OR "Partial Ankyloglossia") AND TS=("Articulation Disorders" OR "Articulation Disorder" OR "Speech Articulation Disorder" OR "Speech Disorders" OR "Speech Sound Disorder" OR "Speech Sound Disorders")	163 07/31/2019
Science direct	("Lingual Frenum" OR "Lingual Frenulum" OR "Ankyloglossia" OR "Tongue Tie") AND ("Articulation Disorders" OR "Speech Articulation Disorder" OR "Speech Disorders" OR "Speech Sound Disorder")	3 07/31/2019
Scielo	((Lingual Frenum) OR (Lingual Frenums) OR (Lingual Frenulum) OR (Lingual Frenulums) OR (Ankyloglossia) OR (Ankyloglossias) OR (tongue) OR (tongue tie) OR (tongue ties) OR (Tongue Diseases) OR (Partial Ankyloglossia) OR (Partial Ankyloglossias)) AND ((Articulation Disorder) OR (Articulation Disorders) OR (Speech Articulation Disorder) OR (Speech Disorders) OR (Speech Sound Disorder) OR (Speech Sound Disorders))	51 07/31/2019
Grey Literature Report	"Lingual Frenum" OR "Lingual Frenums" OR "Lingual Frenulum" OR "Lingual Frenulums" OR "Ankyloglossia" OR "tongue" OR "Tongue Tie" OR "Tongue Ties" OR "Tongue Diseases" OR "Partial Ankyloglossia" OR "Articulation Disorders" OR "Articulation Disorder" OR "Speech Articulation Disorder" OR "Speech Disorders" OR "Speech Sound Disorder" OR "Speech Sound Disorders"	0 07/31/2019

Table 2. Characteristics of included studies

			Features		
Author / Year	Parents	Design	n	Participants	Exposure ranking: ankyloglossia Classification
Braga et al., 2009[6]	Brazil	Transverse	260	Age: 6 to 12 years (mean age 8.4 ± 1.5 years) Sex: 120 (46%) male 140 (54%) female	Total with diagnosis: 47 (18%) Rating: 28 (60%) short, 12 (25%) anterior, 7 (15%) short and anterior
Daggumati et al., 2019[21]	USA	Transverse	470	Age: Study group: 6.04 years; Control group: 5.97 years Sex: 52 (67.5%) male 25 (32.45%)	Total with diagnosis: 220 (46.8%) Rating: Not reported
Dollberg et al., 2011[19]	Israel	Case - control	23	Age: 4 to 8 years Sex: 17 (73.9%) male, 6 (26.1%) female	Total with diagnosis: 15 (65.2%) Rating: Not reported
Karabulut et al., 2008[18]	Turkey	Transverse	127	Age: 20 days to 7 years (mean 15.6 months) Sex: 92 (72%) male, 35 (28%) female	Total with diagnosis: 127 (100%) Rating: 72 (56.7%) mild degree, 55 (43.3%) moderate degree
Marchesan, 2004[8]	Brazil	Transverse	1402	Age: 5 years 8 months - 62 years 10 months Sex: 687 (49%) male, 715 (51%) female	Total with diagnosis: 127 (9%) Rating: 21 (16.5%) short, 106 (83.5%) anterior
Sanchez-Ruiz et al., 1999[17]	Spain	Transverse	72	Age: 2 weeks - 13 years (Mean 3.03 ± 3.41 years) Sex: Not reported	Total with diagnosis: 72 (100%) Rating: Not reported
Suzart et al., 2016[12]	Brazil	Transverse	52	Age: 8 years 6 months - 10 years 11 months (Mean 9.78 years) Sex: 11 (42.3%) male, 15 (57.7%) female	Total with diagnosis: 26 (50%) Rating: 19 (73.1%) short, 5 (19.2%) short and anterior, 2 (7.7%) anterior

Table 3. Description of the objective outcome measures and results of included speech studies by author and year of publication

Author / Year	Aim	Outcome measure Assessor	Speech results Frequency of speech problems in association with ankyloglossia
Braga et al., 2009[6]	Check the prevalence of alteration on lingual frenulum and its implications in speech of children of school age	Phonological naming test for children ABFW Assessor:speech language pathologist	Frequency: 34 (72%); 6 (85%) short and anterior, 21 (75%) short- and 7 (58%) anterior Association: p=0.3865: protrusion (p=0.7921), omission (p=0.2332), distortion (p = 0.856), blocked articulation (p=0.3793), rapid speech (p=0.2407)
Daggumati et al., 2019[21]	Determine whether there are differences in caregiver perception of the speech of patients who underwent frenotomy compared to patients who were not treated surgically.	Review of medical records Assessor: not stated	Frequency: 220 (100%) Association: not stated
Dollberg et al., 2011[19]	Determine whether articulation and speech intelligibility affect children who have undergone frenotomy in early childhood compared to children without ankyloglossia.	Standardized articulation test Assessor: speech language pathologist	Frequency: consonant articulation 7.1 $\pm$ 6.9 errors, word precision 14.5 $\pm$ 10.0 errors, speech intelligibility 1.7 $\pm$ 0.36 errors, sentence 0.46 $\pm$ 1.6, speech fluency 1.6 $\pm$ 0.5 errors Association: not stated
Karabulut et al., 2008[18]	Define the characteristics of patients referred to surgery for ankyloglossia.	Review of medical history Assessor: not stated	Frequency: 8 (6.2%) Association: not stated
Marchesan, 2004[8]	Classify the different types of lingual frenulum and associated speech disorders.	Analysis of collected samples of speech Assessor: not stated	Frequency: 62 (48.81%) 12 (57%) Short, 50 (47.2%) anterior Association: p<0.001
Sanchez-Ruiz et al., 1999[17]	Understand the relationship between the lingual frenulum with speech and other oral functions, and evaluate surgical referrals and the results of clipping the frenulum.	Review of medical history questionnaires over the telephone Assessor: not stated	Frequency: 19 (38%) Association: not stated
Suzart et al., 2016[12]	Characterize and compare the speech disorders related to alteration on lingual frenulum.	Evaluation of spontaneous speech; rating elicited speech by pointing to figures, used in the Assessment Protocol for Orofacial Motricity, MBGR Assessor: speech language pathologist	Frequency: 24 (92.3%); 20 (76.9%) with low tongue posture, 16 (61.5%) blocked articulation, 6 (23.1%) with phonetic speech problems Association: tongue posture (p=0.02), blocked articulation (p=0.01), phonetic speech problems (p=0.05)

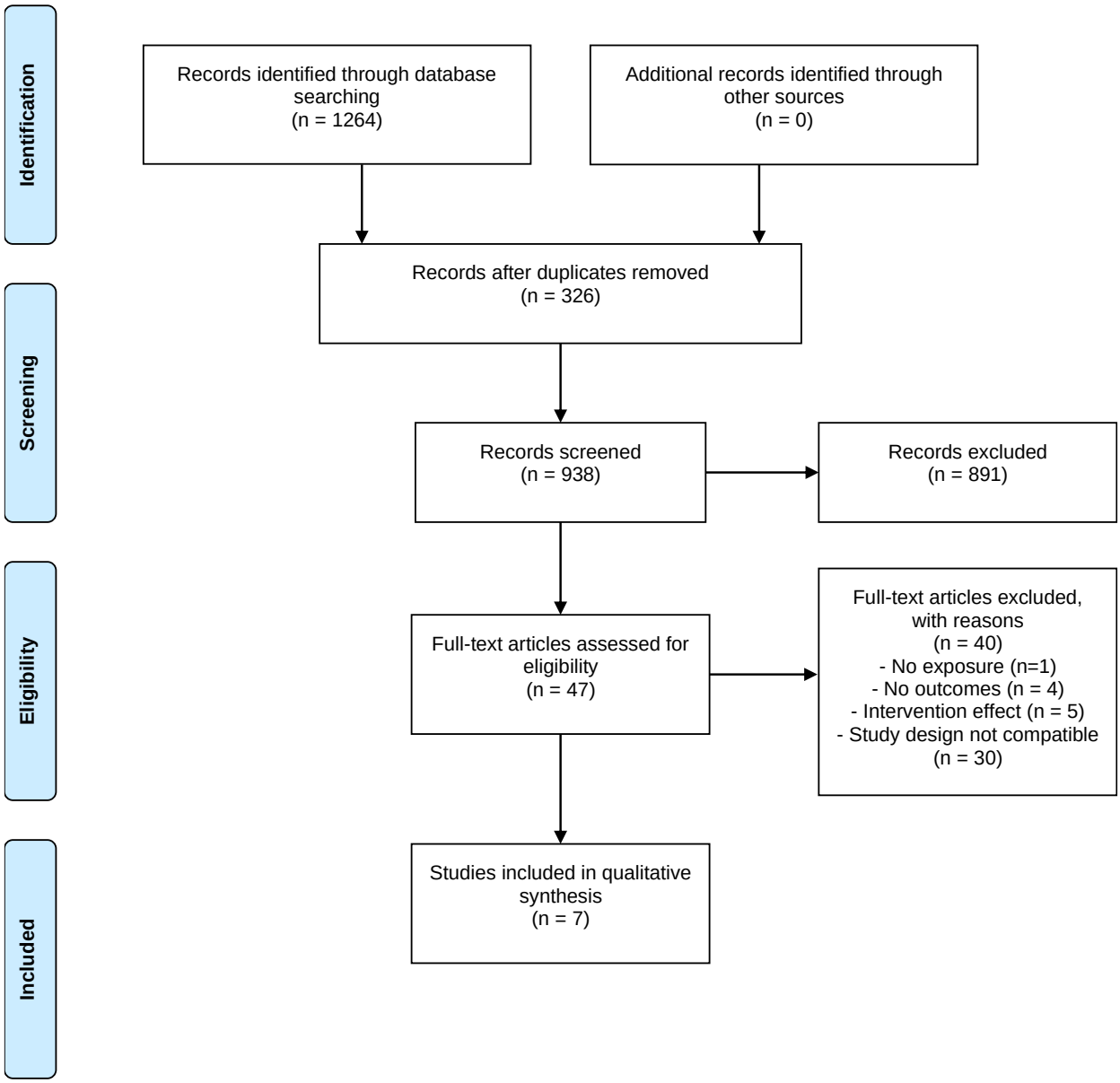
Table 4. Bias risk analysis of included studies

Autor/Ano	Critéria*														Quality Rating (Good, Fair, or Poor)
	1	2	3	4	5	6	7	8	9	10	11	12	13	14	
Braga et al., 2009	Y	N	CD	Y	N	Y	NA	Y	Y	NA	Y	N	NA	N	Poor
Daggumati et al., 2019	Y	N	CD	N	N	Y	NA	Y	N	NA	Y	N	NA	N	Poor
Dollberg et al., 2011	Y	N	CD	N	N	Y	NA	N	N	NA	N	N	NA	N	Poor
Karabulut et al., 2008	Y	N	CD	N	N	Y	NA	N	N	NA	N	N	NA	N	Poor
Marchesan et al., 2004	Y	N	CD	N	N	Y	NA	NA	Y	NA	N	N	NA	N	Poor
Sánchez-Ruiz et al., 1999	Y	Y	CD	N	N	Y	NA	Y	Y	NA	Y	N	NA	N	Poor
Suzart et al., 2016	Y	N	CD	Y	N	Y	NA	N	Y	NA	Y	N	NA	N	Poor

Legend: Y= Yes, N= No, CD= Cannot Determine, NA= Not Applicable, NR= Not Reporterd

*Critéria:
1. Was the research question or objective in this paper clearly stated?
2. Was the study population clearly specified and defined?
3. Was the participation rate of eligible persons at least 50%?
4. Were all the subjects selected or recruited from the same or similar populations (including the same time period)? Were inclusion and exclusion criteria for being in the study prespecified and applied uniformly to all participants?
5. Was a sample size justification, power description, or variance and effect estimates provided?
6. For the analyses in this paper, were the exposure(s) of interest measured prior to the outcome(s) being measured?
7. Was the timeframe sufficient so that one could reasonably expect to see an association between exposure and outcome if it existed?
8. For exposures that can vary in amount or level, did the study examine different levels of the exposure as related to the outcome (e.g., categories of exposure, or exposure measured as continuous variable)?
9. Were the exposure measures (independent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?
10. Was the exposure(s) assessed more than once over time?
11. Were the outcome measures (dependent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?
12. Were the outcome assessors blinded to the exposure status of participants?
13. Was loss to follow-up after baseline 20% or less?
14. Were key potential confounding variables measured and adjusted statistically for their impact on the relationship between exposure(s) and outcome(s)?

Figure 1. Flow diagram of article selection process.



3.2 ARTIGO 2

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Tipo de artigo: Artigo Original

THE EFFECTIVENESS OF LINGUAL FRENECTOMY AND MYOFUNCTIONAL THERAPY FOR CHILDREN: A RANDOMIZED CONTROLLED CLINICAL TRIAL

Running Title: Lingual frenectomy myofunctional therapy in children

Mônica Carminatti[1]
Gabriel Ferreira Nicoloso[2]
Priscilla Polinesi Miranda[3]
Erissandra Gomes[4]
Fernando Borba de Araujo[1]

[1]Post-Graduate Program in Pediatric Dentistry, Federal University of Rio Grande do Sul, Brazil
[2]Centro Universitário da Serra Gaúcha (FSG) and Dentistry Program, Inedi Faculty (CESUCA), Brazil
[3]Post-Graduate Program in Child and Adolescent Health, Federal University of Rio Grande do Sul, Brazil
[4]Faculty of Dentistry, Federal University of Rio Grande do Sul, Brazil

Author contributions: E.G. conceived the ideas; M.C., G.F.N. and P.P.M. collected the data; M.C. analysed the data and led the writing; F.B.A. and E.G. proofread the manuscript.

Corresponding author:
Prof. Erissandra Gomes
Faculty of Dentistry, Federal University of Rio Grande do Sul
Ramiro Barcelos 2492, 90035-003, Porto Alegre, RS, Brazil
Phone number: +55 51 3308 5176
E-mail: erifono@hotmail.com

Word count: 4.415

Conflict of interest

The authors declare no conflict of interest.

ABSTRACT

Background: Surgical referrals for anomalies in the lingual frenulum should take into consideration the morphological and functional aspects of tongue and the fact that myofunctional therapy may be necessary following surgery. Objective: To evaluate the effectiveness of lingual frenectomy, as a stand-alone treatment or in association with myofunctional therapy, on the general and functional aspects of children with anomalies in the lingual frenulum, by comparing them to children with typical lingual frenulum between the ages of six to 12 years old. Methods: A randomized controlled clinical trial with 40 patients diagnosed with altered lingual frenulum and 20 patients with typical lingual frenulum (CGO). Initially (T0), the Tongue Frenulum Evaluation Protocol was used to evaluate all of the children. The ones who were diagnosed with alteration on lingual frenulum and underwent a frenectomy were also assessed 15 (T15) and 30 days (T30) after the procedure, using the same protocol. At T15, participants had undergone frenectomy were randomly distributed between two groups: one which offered tongue mobility exercises (GE) and another which did not (GC). Statistical tests were used to draw a comparison between and within groups, at $p<0.05$. Results: Considering the time effect, GE improved significantly when compared to GC with respect to tongue mobility ($p=0.016$), maximum mouth opening (MMO) ($p=0.024$) and MMO with the tip of the tongue touching the incisive papilla (TTIP) ($p=0.026$). Conclusion: Frenectomy associated with myofunctional therapy benefitted the tongue mobility of the children.

Keywords: Tongue-tie, Lingual frenulum, Ankyloglossia, Frenectomy, Myofunctional therapy, Clinical trial

INTRODUCTION

The lingual frenulum is a mobile, three-dimensional structure composed of a central band of connective tissue called fascia which extends to the floor of the mouth. Lingual frenulum develop along a varying spectrum of morphology[1]. Atypical anatomy of the frenulum is due to a failure in cell apoptosis during embryogenesis, between the floor of the mouth and the tongue. This causes changes in attachment and/or length[2]. The point of attachment to the floor of the mouth does not change over time and its histological characteristics prevent spontaneous breaks or elongation through exercise[3]. The prevalence of anomalies ranges between 0.1% and 18%[4,5], and they seem to occur more frequently in men [6]. An altered lingual frenulum interferes in orofacial functions[7,8]. These facts justify the importance of proper diagnosis and treatment for altered lingual frenulum and the repercussions they cause.

Established assessment parameters have been based on the observation of tongue mobility and appearance, associated with the points of attachment of lingual frenulum[7,9]. Although tools that were intended to classify changes in the lingual frenulum have also been used for assessment[10,11], there is no validated instrument for the examination of children of preschool age or older[12].

Referrals for surgical intervention should follow careful consideration of the morphological and functional aspects of the tongue so as to promote improvement in these aspects[13,14]. There is no scientific evidence favoring any particular surgical method[4,7]. However, a frenectomy is the procedure that has been used to remove the lingual frenulum[2]. A systematic review found that, in some studies, various surgical techniques improved aspects of tongue mobility, such as elevation and protrusion, after intervention[4]. However, another study showed that, after the surgery, tongue protrusion showed the best response while lifting improved the least[15]. In some cases, particularly the most serious ones, a longer recovery period may be necessary before lifting motions of the tongue show progress. For these cases, tongue mobility exercises could be recommended[4].

Speech therapy after surgery for lingual frenulum anomalies has been documented[9,16-19]. One case series study noted that substitutions or omissions in speech can improve after surgery but then evolve into distortions. Distortions took longer to correct and remained in the speech of some children. As such, tongue

exercises after surgery and speech therapy would constitute necessary care[18,19] for children[20]. Approaches involving isotonic tongue exercises language and techniques for the production of phonemes have shown results in relation to improved mobility, thus demonstrating greater therapeutic effect[17,21].

An increasing number of patients and healthcare professionals have sought seeking evidence-based information for the treatment of altered lingual frenulum in children. Nevertheless, there is a dearth of published evidence of quality[4,22,23]. In the literature, most of the articles published on this topic present methodological limitations including sample size, non-uniform backgrounds in relation to population ages, different surgical techniques in the same study and no characterization of anatomical or functional features[4,19-22].

The objective of this study was to evaluate the effectiveness of frenectomy, with or without myofunctional therapy, on the general and functional aspects of children with alteration on lingual frenulum. These groups were also compared with children with typical lingual frenulum, between six to 12 years old.

METHODOLOGY

Sample

This was a randomized, single-blind, parallel-group controlled study, was performed on children between six and 12 years of age, of both sexes, who had been referred to the Faculty of Dentistry at the Federal University of Rio Grande do Sul (UFRGS), in Brazil. The research project was developed between May 2016 and October 2019, and subsequently approved by the Research and Ethics Committee at UFRGS, under approval number 21471/2012. A written informed consent form was filled out by parents and children before their participation in the study. The protocol was published in the Brazilian registry of randomized clinical trials (*Registro Brasileiro de Ensaios Clínicos* - REBEC), under code number RBR-7292kp number.

The study included children who did or did not present anomalies in their lingual frenulum. The participants of the former group should receive referrals for lingual frenectomy. Children with no upper and/or lower incisors, head or neck malformations, cognitive impairment, complaints of hearing loss, or previous or contemporaneous history of surgical procedures on their lingual frenulum were not included.

Logistics

The logistics of the assessments and interventions concerning the children in our study are shown in Figure 1.

Rating lingual frenulum

The children were examined by a speech language therapist (E.G.) who used the Tongue Frenulum Evaluation Protocol[11]. This tool is the only instrument published in Brazil. It has a score range and is currently under the process of being validated.

This protocol consists of an interview, to be carried out with the parents or guardians, and a clinical examination. This clinical assessment is divided into general and functional findings. The general findings include reduced tongue lift, and maximum mouth opening (MMO) measurements in millimeters, as well as MMO measurements with the tip of the tongue touching the incisive papilla (TTIP), in millimeters. These measures are also converted to percentages. Similarly, the types of lingual frenulum and their points of attachment were classified. The functional findings include tongue mobility, muscle tone, tongue position at rest, changes in the production of speech sounds and other aspects of speech. The following scores are used to rate the findings of the clinical examination: zero means no anomalies, while scores of 1 or 2 indicate the severity of an alteration. When the sum of the scores of the general tests is equal to or greater than 3, the lingual frenulum is considered changed. When the sum of the functional tests is equal to or greater than 25, anomalies in the lingual frenulum may be affecting orofacial functions, such as speech[11].

The speech assessment was also performed using the mentioned protocol and was composed of three parts: 1) informal speech (we asked the children to answer these questions and tasks: What's your name? How old are you? Do you study or work? Tell us about your school. Tell us about something interesting that happened to you; 2) counting from 1 to 20; then naming the weekdays and the months of the year; 3) pointing to a board and naming 50 figures. During this part of the evaluation omissions, substitutions and distortions were observed. Under the item "other aspects to be observed during speech", opening of the mouth, tongue position, jaw movement, speed, accuracy of speech as a whole and voice were all analyzed.

The protocol was used together with an anthroposcopic approach,

complemented by further image and footage analysis, and recorded with a digital Canon™ PowerShot SX240HS camera placed a meter away from the child. The examiner sat facing the child, who was asked to sit with their feet flat on the floor and their head in a habitual position. In each session, gloves were used by the assessor, as well as a Digimes™ digital caliper previously sterilized with 70% ethanol.

A diagnosis of anomaly of the lingual frenulum was reached based on the results of the Tongue Frenulum Evaluation Protocol [11].

Treatment for anomalies of lingual frenulum

Frenectomy

Prior to frenectomy, the operating dentist (G.F.N.) evaluated the patient's anatomy and the range of tongue lift, extension and lateralization. G.F.N., who has experience in this field, performed the surgical procedure on all patients and used the same frenectomy technique under local anesthesia delivered via lingual nerve. The apex of the tongue was previously transfixed with surgical to immobilize the tongue. The incision of lingual frenulum tissue was made with a surgical scalpel blade No. 15. Immediately after, the muscle fibers were dissected with straight, blunt-edged scissors to create a diamond-shaped surgical wound. Tongue lift, even if passively, was assessed in order to verify adequate movement for oral functions. The wound was then closed with silk sutures which were removed seven days after surgery (T7). Fifteen days post-procedure (T15), these children were randomly distributed into new groups.

Randomization Process

Children who had been diagnosed with anomalies of the lingual frenulum were randomly divided into two groups: children who had undergone frenectomy but were not offered isotonic tongue exercises (Comparison Group - CG); and children who had undergone frenectomy and also received postoperative therapy in the form of isotonic tongue exercises (Study Group - SG). The sequence was generated on the https://www.random.org/ website.

Mobility exercises

After randomization at T15, children belonging to SG, were instructed to perform isotonic tongue exercises between 15th and 29th day after the procedure. Based on literature[21,24], three exercises were given: running the tongue around

closed lips and teeth, touching four cardinal points outside of the mouth with the tip of the tongue and pronouncing the sounds /l/ and /n/ alternately. Patients were asked to perform the exercises three times a day to record frequency of practice in a table given to each SG participant. This table had been designed by the researchers. The 14-day table was complemented by a description of the proper way to do each exercise and patients were instructed to record the dates and frequency of each exercise each day. The children were also instructed not to mention doing these exercises to the speech language therapist who assessed them at T30. The speech language therapist (M.C.) who conducted the exercise portion of the study was not the same speech language therapist who examined the children at T0, T15 and T30.

If a child selected for the research was already in speech therapy elsewhere, a letter was sent to the professional in order to explain the inclusion of the patient in the study and to request that, between T0 and T30, no exercises for tongue mobility or stimulus of speech production be carried out, so as to avoid potential interference in our study results.

Intervention Effects

The Tongue Frenulum Evaluation Protocol[12] was used to examine CG and SG children before surgery at T0, T15 and T30. The same examiner (E.G.) assessed the participants in order to verify the effects of interventions. Only the patient history interview with the parents or guardians was performed pre-surgery. The group of children with typical lingual frenulum (CGO) were assessed at T0. The intervention assessor was blinded as to whether children belonged to the CG or SG group.

Statistical analysis

Quantitative variables were described as mean and standard deviation or median and interquartile range. For comparison between groups, the Student’s t-test or analysis of variance (ANOVA) was used, complemented by the Tukey test. In the case of asymmetry, the Mann-Whitney and Kruskal-Wallis tests were performed, respectively. The qualitative variables were described by absolute and relative frequencies. To compare the proportions between groups, the Pearson’s chi-square test or Fisher's exact test was applied. For simultaneous intra and inter-group comparisons, generalized estimates of equations (GEE) were used, complemented by the least significant difference (LSD) test. The level of significance was set at 5%

(p£0.05). Statistical analyzes were performed using IBM SPSS Statistics 21 (SPSS Inc., Chicago, IL).

RESULTS

Of the 62 children diagnosed with alteration on lingual frenulum, 6 were missing upper and or lower incisors, had undergone or were undergoing speech therapy at the time of our study or presented cognitive impairment. Therefore, they did not meet the inclusion criteria. Another 4 children were excluded because of their parents or guardians or because they themselves did not wish to participate in the study. During the follow-up period with and without exercise, 12 children were excluded due to abandonment or failure to do the exercises. They were included in the final analysis of the 40 children diagnosed with alteration on frenulum of the tongue that completed all stages of the study. Figure 2 shows the CONSORT flowchart diagram for children with alteration on lingual frenulum. In the CGO, 20 children with typical lingual frenulum were included, with the aid of the protocol[11].

Regarding the general characterization of the sample, groups did not differ according to sex (p=0.535), but there were more male subjects in all groups, with 14 (70%) in the CG, 11 (55%) in the SG and 11 (55%) in the CGO. Mean age was similar among the three groups (p=0.389) and was calculated at 8,11$\pm$1,91, for the CG, 8.70$\pm$1.93 for the SG and 8.92$\pm$1.90 for the CGO. There was no difference in patient complaint between the CG and the SG (p=0,126). No child from the GCO presented complaints during the patient history interview. However, complaints relating to speech were frequent, 12 (60%) in the CG and 9 (45%) in the SG, followed by complaints concerning a short frenulum by 6 (30%) in the CG and 8 (60%) in the SG. The variables related to medical history showed no differences among groups except in relation to speech impediment, an association which was observed when CG and SG were compared with CGO (p=0.001). There were no reports of speech problems in the CGO while 16 (80%) of the CG and 14 (70%) of the SG did report speech problems. Family history presented a comparative association among the groups (p=0.002). Whereas GCO reports related no occurrences in the family, 10 (50%) from the CG and 12 (60%) from the SG did relate family history of the occurrence.

The results of the functional and general tests of the three groups at T0 are shown in Table 1. There was no significant difference between the CG and SG in

relation to the results of the functional and general tests However, when these groups were compared with CGO, they presented lower scores on these tests, in accordance with the assessment protocol scale. An analysis of the frequency of anomalies in the CG and SG in functional tests, most were only able to reproduce imprecise tongue mobility movements or were simply unable to do them. Among the CG and SG subjects, disability was most frequent in the tongue vibration test, 32 (80.0%), followed by reduced tongue muscle tone 21 (52.5%) and non-visible tongue at rest 22 (55.0%). On all the speech tasks of the assessment protocol, the anomaly of the highest occurrence was distortions of phonemes in 30 (75%) children. Other atypical aspects of speech were observed among the children, such as reduced mouth opening 30 (75%), tongue position on the floor of the mouth 20 (50.0%) and anterior tongue position 24 (60.0%). Unclear speech was noted in 36 (90.0%) children.

Results of general and functional tests at T0, T15 and T30 concerning intervention effect over time and among groups are described in Table 2. With regard to the time effect between T0 and T15, the general and functional aspects of tests improved in both groups, with the exception of MMO in the CG and SG, which showed no significant progress ($p=0.245$), and a smaller improvement in other aspects of speech in the SG. Nevertheless, the SG went on to show further progress between T15 and T30, with statistical significance for improvements in tongue lift ($p<0.001$), tongue mobility ($p<0.001$), and other aspects of speech ($p<0.001$). The relationship between the percentage of MMO and MMO with TTIP presented significant improvement between T15 and T30 for the CG ($p<0.001$). However, this relationship was not observed in the SG. In the intra-group analysis, no association was observed among the variables of the general and functional tests. However, considering the effect of time on the groups between T0 and T30, the SG showed significant improvement when compared to the CG with regard to tongue mobility, MMO and MMO with TTIP.

DISCUSSION

Regarding the classification of anomalies of lingual frenulum, reports of short and anterior frenulum are the most frequent of the present study. However, others have noted a higher prevalence of short frenulum[5,25]. Since there is no consensus on diagnostic criteria, each study is defined by specific indicators which, in turn, are

related to factors such as profession, geography, personal experience and number of cases. These differences may affect classification[23].

The results of the general tests of the CG and SG at T0 showed low scores, in accordance with the assessment protocol scale. A systematic review discussed the common criteria among studies regarding abnormalities of lingual frenulum. The study were referred to the appearance of the frenulum (short and sometimes thick), the format of the tongue when extended (heart-shaped) and difficulties in mobility (the inability to extend the tongue beyond the lower jaw incisors, etcetera)[12]. So, there is an anatomical profile of what anomalies of the frenulum cause but, since the degree of irregularity is variable, the anatomy of tongue, the frenulum and other adjacent structures such as the floor of the mouth must be assessed[11].

The outcomes of the functional tests at T0 showed low scores for the CG and SG. Movement restriction occurs due to mechanical changes in the tongue[15]. In this study, when the tongue mobility test was carried out, most children CG and SG children were observed reproducing imprecise movements or failing to do them altogether. Studies have shown the occurrence of changes in mobility[8,25,26] and praxis[8,26] in individuals with alteration on lingual frenulum. These showed a significant association between the extension of the frenulum and mobility tests, as well as between praxis, attachment of the frenulum to the floor of the mouth and tongue mobility[8]. Another study reported that short and anterior frenulum presented greater changes in lift and vibration praxis of the tongue, and that suction tasks were also more difficult[5]. This is because the lingual frenulum is composed of a high amount of type I collagen fibers which have considerable tensile strength and can therefore restrict tongue movement[3]. In this study, the short and anterior frenulum were the most prevalent in the CG and SG, as well as an accompanying inability to vibrate the tongue.

The muscle tone of the tongue was diminished among more than half of the children in the CG and SG. This finding has been corroborated in other studies[8,25]. In children with alteration on frenulum, the resting position of the tongue will probably be on the floor of the mouth, due to difficulty in maintaining the tip of the tongue on the incisive papilla. On its own, this can cause a decrease in muscle tonus[25]. In the habitual posture of rest, the tongue was not visible in 55% of the children. One must keep in mind that, in order to verify the position of the tongue at rest, the subject must have their mouth open or half-open. At the time of the assessment, these

participants' lips were closed[25].

For more than half of the children with anomalies of the lingual frenulum, the tongue was positioned on the floor of the mouth and also exhibited an anterior posture during speech. This finding was similar to that of another study[25]. This is to be expected since differences in attachment and extension make it difficult for the tongue to move freely. For this reason, the tongue remains in a low position while articulating speech sounds[25].

The muscular and anatomical aspects of the tongue, such as strength, mobility, praxis, posture, morphology and frenulum, must be in harmony for proper stomatognathic performance[27]. The compensatory behaviour of patients with alteration on lingual frenulum may contribute to a wide variety of issues presenting as myofunctional disorder[28]. Anomalies in lingual frenulum may impair mobility which, in turn, is likely to result in limitations in oral functioning. The function of speech may suffer the greatest impact in this regard[25].

A distortion of phonemes was observed in 75% of the CG and SG. Similar results have been observed in other studies[5,14,19,20,25,29,30]. For speech to be produced properly, there must be a balance between all the anatomical and functional structures of the stomatognathic system and the motor subsystems involved in their production[31]. Phonetic speech disorder is the most commonly found speech symptom in children with alteration on lingual frenulum[25]. This occurs because the anterior third portion of the tongue, which is necessary for the articulation of sounds, can not rise to the alveolar region due to the mechanical impediment caused by the shortening and/or atypical attachment of the lingual frenulum[30].

Other aspects of speech have been observed, such as the articulatory imprecision - which was noted in 90% of the CG and SG - as well as reduced mouth opening during speech. Another study found similar results in individuals with alteration on lingual frenulum[29]. "Blocked articulation" is one of the factors that cause articulatory imprecision and often affects speech as a whole[25]. This is a type of compensation due to reduced tongue mobility. While the tongue tries to move through the correct points of articulation to produce speech sounds, the space between the upper and lower jaws narrows[5,29]. Reports have shown that shows that "blocked articulation" is another frequent speech symptom which suggests an atypical lingual frenulum[25].

As for the time effect, when participants returned for reassessment at T15, significant progress was noted in relation to their performance in general and functional tests after frenectomy, with the exception of some aspects already mentioned in this study. A similar result was found in another study also performed in Brazil, which evaluated 10 participants with the same assessment protocol. After frenectomy, all patients showed varying degrees of progress regarding orofacial function and tongue posture and mobility, whether or not there was complementary speech therapy[15]. A survey conducted in Japan evaluated the speech of children between three and eight years of age, with a diagnosis of anomalies in the lingual frenulum. The research, which documented the progress of these patients up to two years after frenectomy, outcomes similar to ours. After a speech articulation test was given, the authors found that the speech impediments in 4 of the 5 children (80%) had improved due to the surgical procedure[20]. Other studies assessing postoperative speech and tongue mobility corroborate the results of this study[17,21]. A systematic review[4] quoted four studies which compared aspects related to tongue mobility after surgery. These studies used different techniques for the surgical correction of the lingual frenulum. In all three studies, MMO and TTIP, as well as tongue extension, were measured. As we observed in our sample, after surgery all three of these measurements showed progress.

As regards the effect of time and exercises, upon assessment at T30, the SG showed significant improvement in tongue mobility and other aspects of speech, such as mouth opening, tongue position and speech accuracy. The CG did not show the same degree of progress in the same period. Similar results were found in studies[17,21] which used isotonic exercises after frenectomy. The authors observed that, in general, articulation improved but no mention was made about whether omissions, distortions and phonetic substitutions remained in speech. One case study reported that an eight year old child, after having undergone a frenectomy, still presented speech distortions over a two-year follow up. The authors identified speech problems involving omission, substitution and distortion, even after exercise therapy. Another case study states that effective rehabilitation therapy should include the functional therapy due to distortions of the phoneme /r/[32]. Postoperative tongue exercises and speech therapy are essential for individuals who have acquired compensatory speech habits due to the presence of an alteration on lingual frenulum[20].

Concerning the effect of the time difference between T0 and T30 on groups, the SG showed significant improvement in tongue mobility, MMO and MMO with TTIP, especially when compared to the CG. This study demonstrates that general and functional aspects improved to different extents after frenectomy and myofunctional therapy. However, when the CG and SG were compared with the CGO, the tongue mobility and speech function results of the first two groups still demonstrate a need for more myofunctional therapy in order to achieve outcomes similar to that of the CGO.

In our study, the effect of exercise after frenectomy showed promising results. Nevertheless, myofunctional therapy ended before orofacial muscles could fully adopt a new pattern of functioning to support tongue lift and mobility as well as normal speech. According to a study that also offered postoperative myofunctional exercises to children, improvements in tongue lift and extension are not fully apparent until after one to three months after surgery[17]. This is most likely related to the fact that tongue muscles take some time to adapt after the removal of the lingual frenulum. Thus, further postoperative research on children is needed for longer periods.

Why this paper is important to pediatric dentists

A frenectomy is an effective measure in improving tongue lift and mobility, as well as speech. Myofunctional therapy supports better progress in tongue lift and mobility, as well as in other aspects of speech, such as mouth opening, tongue position and speech accuracy.

This study demonstrates the importance of the multidisciplinary treatment of ankyloglossia by pediatric dentists and speech language pathologists.

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Table 1. Results of general and functional tests according to the groups at baseline (T0)

Variables	CG	SG	CGO	P-value	
				CG x SG	Intragroup
General tests					
Results tongue lift - mean ± SD	2.50 ± 0.68	2.65 ± 0.58	0 (0.0)	0.478	<0.001 *
Tongue lift forms a square / rectangle - n (%)	17 (85.0)	18 (90.0)	0 (0.0)		<0.001 *
Tongue lift forms a heart shape - n (%)	13 (65.0)	15 (75.0)	0 (0.0)		<0.001 *
Floor of mouth rises with tongue lift - n (%)	20 (100)	20 (100)	0 (0.0)		<0.001 *
Results measured with a caliper - median (P25 -P75)	1 (0 - 1)	1 (1 - 1)	0 (0 - 0)	0 152	<0.001 *
Measurement of MMO	42.5 (36.8 - 46.6)	39.9 (29.3 - 57.0)	44.01 (35.9 - 52.3)	0.137	0.056
Measure of MMO +TTIP	14.7 (0 - 25.8)	10 (0 - 27.0)	32.2 (25.4 - 39.8)	0.86	<0.001 *
Percentage of the relationship between AMB and MMO + TTIP measurements	34.6 (0 - 61.1)	24.0 (0 - 48.4)	75.5 (50.7 - 89)	0.133	<0.001 *
Results frenulum attachment - median (P25 -P75)	2.28 (1 - 3)	2.51 (1 - 3)	0.20 (0 - 1)	0.120	<0.001 *
Attachment to floor of mouth - n (%)					
Caruncle	1 (5.0)	1 (5.0)	16 (80.0)		
Inferior alveolar crest	19 (95.0)	19 (95.0)	4 (20.0)		
Attachment the underside of the tongue - n (%)					
Middle	1 (5.0)	0 (0.0)	20 (100)		
Between middle and tip	12 (60.0)	8 (40.0)	0 (0.0)		
Tip	7 (35.0)	12 (60.0)	0 (0.0)		
Results of clinical classification of the frenulum - median (P25 -P75)	2.0 (2 - 2)	2.0 (2 - 2)	0.0 (0 - 0)	1,000	<0.001 *
Final classification of the frenulum - n (%)					
Normal	-	-	20 (100)		
Short	7 (35.0)	7 (35.0)	-		
Short and anterior	12 (60.0)	11 (55.0)	-		
Ankyloglossia	1 (5.0)	2 (10.0)	-		

Overall result of the anatomical assessment - median (P25 -P75)	8.0 (4 - 9)	8.3 (6 - 9)	0.2 (0 - 1)	0.484	<0.001*
Functional tests					
Tongue mobility results - median (P25 - P75)	6.7 (3-15)	9.4 (3-15)	0.0 (0 - 1)	0.313	<0.001 *
Tongue muscle tone - median (P25 - P75)	1.0 (0 - 2)	1.2 (0 - 2)	0.0 (0 - 0)	0.622	<0.001 *
Tongue position at rest results - median (P25 - P75)	0.6 (0 - 2)	0.5 90-2)	0.1 (0 - 1)	0.685	0.020 *
Speech results - median (P25 - P75)	6.8 (0-12)	6.6 (0 - 12)	0.6 (0 - 6)	0.870	<0.001 *
Other aspects of speech results - median (P25 - P75)	3.6 (1-6)	3.4 (2 - 6)	0.9 (0 - 4)	0.393	<0.001 *
Results of general functionality tests - median (P25 - P75)	20.5 (8-29)	21.0 (14 - 35)	0.9 (0 - 10)	0.356	0.001*

Legend: CG = Comparison Group; SG = Study Group; CGO = control group; n = number; SD = Standard Deviation; P = Percentile; MMO = Maximum Mouth Opening; TTIP = Tongue Tip on Incisive Papilla
p <0.05 * - Kruskal-Wallis

Table 2. Results of general and functional tests as to the effect of intervention over time between CG and SG and the difference between initial and final results in groups

			Effect (p-value)		
Results	CG	SG	Time	Groups	Time x Groups
General tests					
Tongue lift			<0.001 *	0.945	0.367
T0	2.5±0.15 [b]	2.65±0.3 [c]			
T15	1.5±0.24 [a]	1.70±0.28 [b]			
T30	1.25±0.23 [a]	1±0.26 [a]			
T15 and T30 difference	-1.25 (-1.78 to -0.72)	-1.65 (-2.12 to -1.18)			
Results caliper measurements			<0.001 *	0.282	0.810
T0	0.90±0.07b [b]	1±0 [b]			
T15	0.55±0.11[a]	0.60±0.11 [a]			
T30	0.40±0.11[a]	0.55±0.11 [a]			
T15 and T30 difference	0.5 (-0.76 to -0.24)	-0.35 (-0.23 to 0.67)			
MMO measurements			0.245	0.925	0.024 *
T0	41.91±2.81 [a]	40.37±6.69 [a]			
T15	41.70±4.21 [a]	41.91±5.40 [a b]			
T30	41.24±4.71 [a]	42.99±5.27 [b]			
T15 and T30 difference	-0.46 (-2.58 to 1.65)	1.07 (2.34 -0.19)			
MMO + TTIP measurements			<0.001 *	0.582	0.026 *
T0	14.20±6.47 [b]	10.52±6.80 [b]			
T15	18.56±5.84 [a]	18.98±6.59 [a]			
T30	20.45±6.23 [a]	20.68±6.94 [a]			
T15 and T30 difference	1.89 (0.05 to 3.73)	1.69 (0.32 to 3.07)			
Percentage of the relationship between MMO and MMO + TTIP measurements			<0.001 *	0.418	0.067
T0	33.81±15.03 [c]	25.65±15.38 [b]			
T15	44.13±12.86 [b]	45.01±12.58 [a]			
T30	49.73±13.89 [a]	47.83±13.81 [a]			
T15 and T30 difference	5.61 (1.20 to 10.00)	2.81 (-1.09 to 6.73)			
Functional tests					
Tongue mobility			<0.001 *	0.949	0.016 *
T0	7.8±0.83 [b]	9±0.88c [c]			
T15	5.25±0.92 [a]	5.20±0.75 [b]			
T30	4.75±0.92 [a]	3.8±0.59 [a]			
T15 and T30 difference	-3.05 (-4.16 to -1.94)	-5.20 (-6.78 to -3.62)			
Speech					
T0	6.65±0.69[b]	7,00±0,67[b]	<0.001 *	0.588	0.774
T15	6.40±0.69[a]	6.65±0.75 [a]			
T30	6.25±0.68[a]	6.35±0.81[a]			
T15 and T30 difference	-0.40 (-0.82 to 0.02)	-0.65 (-1.44 to 0.14)			

Other aspects of speech			0.049 *	0.828	0.643
T0	3.65±0.27 [b]	3.95±0.18 [b]			
T15	3.50±0.26 [a]	3.80±0.33 [b]			
T30	3.45±0.26 [a]	3.35±0.35 [a]			
T15 and T30 difference	-0.20 (-0.50 to 0.10)	-0.60 (-1.23 to - 0.03)			

Legend: CG = Comparison Group; SG = Study Group; MMO = Maximum Mouth Opening; TTIP = Tip of Tongue on Incisive Papilla
* p <0.05 - Generalized Estimates of Equations (GEE) - Teste Least Significant Difference (LSD)

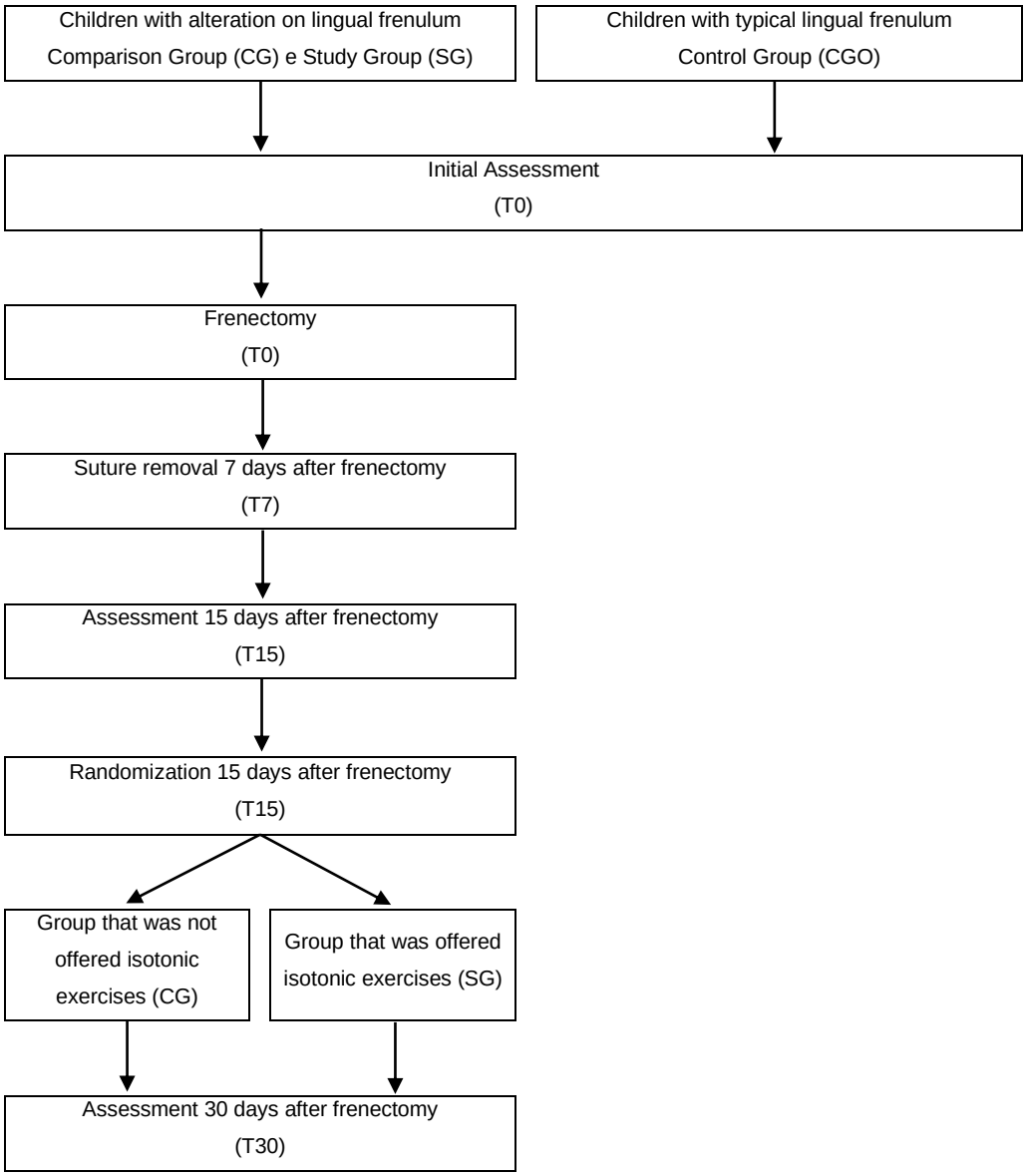

Figure 1. Flowchart of the logistics of the randomized controlled clinical trial

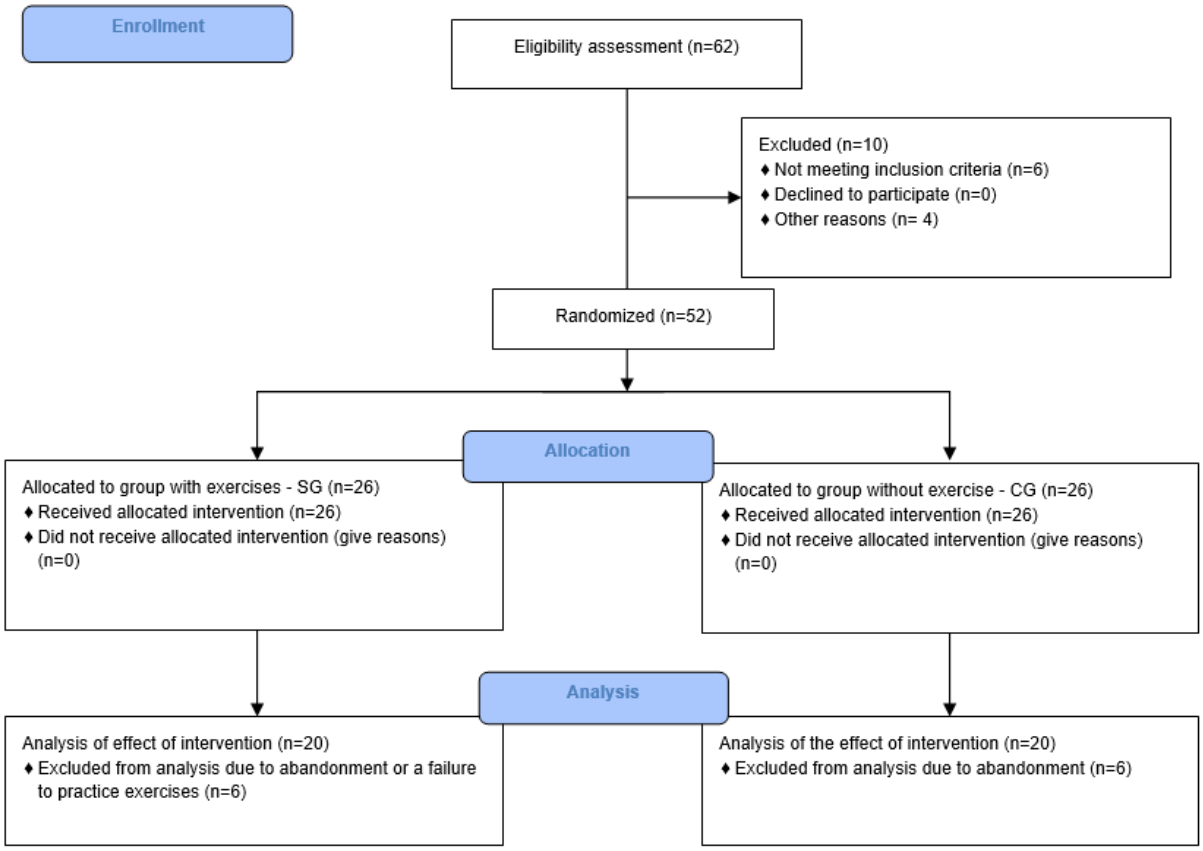

Figure 2. CONSORT 2010 flow diagram for the randomized study of abnormal lingual frenula and the effects of surgical intervention, with or without myofunctional therapy

3.3 ARTIGO 3

Artigo formatado nas normas de J*ournal of Speech, Language, and Hearing Research*

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THE SPEECH CHARACTERISTICS OF CHILDREN WITH ALTERATION ON LINGUAL FRENULUM

Short title: Speech and alteration on lingual frenulum

Mônica Carminatti[1]

Cristina Emília Schunemann[2]

Bárbara de Lavra Pinto Aleixo, PhD [3]

Brunah de Castro Brasil, PhD [3]

Erissandra Gomes, PhD [3]

Fernando Borba de Araujo, PhD[1]

[1]Post-Graduate Program in Pediatric Dentistry, Federal University of Rio Grande do Sul, Brazil

[2]Federal University of Rio Grande do Sul, Brazil

[3]Faculty of Dentistry, Federal University of Rio Grande do Sul, Brazil

Corresponding author:

Profa. Erissandra Gomes

Department of Oral Surgery and Orthopedics, School of Dentistry, Federal University of Rio Grande do Sul

Ramiro Barcelos 2492, 90035-003, Porto Alegre, RS, Brazil.

Phone number: +55 51 3308 5176

E-mail: erifono@hotmail.com

ABSTRACT

Objective: To investigate the speech characteristics of children between the ages of six and 12 years old with alteration on lingual frenulum. Method: We analyzed 34 speakers of Brazilian Portuguese, of both sexes, who come from the southern region of Brazil. We used the speech test of the Tongue Frenulum Evaluation Protocol and described strategies of speech repair. Two judges performed an auditory-perceptual analysis. A third judge resolved any disagreements. Data were analyzed descriptively through percentage distributions. Results: Speech distortions were found in all children of the patient sample. Most children showed distortions of the alveolar consonants [ɾ], [s] and [t] as well as consonant clusters with [ɾ]. Among the other characteristics analyzed, the function of mouth opening presented a narrower range of motion in the sample. The major repair strategy children used was to reduce consonant clusters, followed by omissions of non-lateral liquid sounds in final position. The co-occurrence of phonetic and phonological disorders was more frequent in children with short and anterior frenulum. Conclusion: The highest frequency of speech disorders were phonetic in origin. Nevertheless, strategies of speech repair were also observed.

Keywords: lingual frenulum, ankyloglossia, children, speech disorders, articulation disorders

Conflict of interest: The authors declare no conflict of interest

INTRODUCTION

The lingual frenulum is a mobile, three-dimensional structure composed of a central band of connective tissue called fascia which extends to the floor of the mouth. Lingual frenulum develop along a varying spectrum of morphology[1]. Some anatomical variations in frenulum morphology can lead to restricted tongue movement and an imbalance between stability and mobility[1]. Restrictions in tongue mobility affect tongue praxis[2-4], thus making some oral functions difficult[4,6,7], such as speech[4,7-9]. The prevalence of this congenital anomaly varies between 0.1% to 18%[2,10] and the rates regarding its impact on speech range from 48.8% to 73.3%[2,7,9].

The most studied speech disorders related to alteration on lingual frenulum have been phonetic in nature. The most frequent distortions occur in consonant clusters with [r] (specifically [tr] and [dr]) and [l][9,11], with the alveolar flap [r][2,8,9,11-14] and the alveolar sounds [s], [z][2,11,13,14] [t], [d] [l] and [n][2,8,11,12]. Limitations to broader and more sophisticated articulatory movements have also been observed, along with a smaller opening of the mouth during speech[7,9,13], excessive anterior and lateral mandibular movements, changes in the rate of speech and imprecise articulation[7,8,15]. Distortions in sound can be considered changes that primarily affect speech intelligibility without compromising the phonological contrasts of a language. Therefore, they do not affect the meaning of words[16], unlike omissions and substitutions.

Some studies have reported omissions[2,7,14] and substitutions associated with the phoneme /r/ [7,14] in the speech of children with alteration on lingual frenulum; as such, these are a combination of phonetic and phonological changes. However, these same studies did not analyze the phonological processes of the speech of children with atypical lingual frenulum, nor the strategies of repair that they use. It is

known that the linguistic organization and production of phonemes can be influenced by motor capacity as well as the integrity of the organs of phonation and articulation[2,3,12,16-20]. When these structures show changes in tone and praxis, they can impair phoneme production[21]. Thus, imprecise articulation can also interfere with the phonological aspects of speech, since it requires complex coordination and the planning of lip and tongue movements in order to production sounds[17].

Our analysis of phonological disorders in children with anomalies in lingual frenulum is justified by the possible difficulties in translating phonological knowledge into the appropriate motor actions for the realization of a phone or a correct syllable. In the literature, there are few studies which have investigated changes in the phonological aspect of speech in children with alteration on lingual frenulum, which may also be related to difficulties with the phonetic aspect[2,7,9,14,22].

The aim of this study was to investigate the speech characteristics of children between 6 and 12 years of age, with alteration on lingual frenulum. Moreover, the specific objectives were: 1) to characterize distortions and describe the phones which were affected with the greatest frequency in the speech of children with atypical lingual frenulum, considering the classification thereof; 2) verify and characterize the strategies of repair used in the speech these children, considering the classification thereof.

METHODS

Ethical considerations

This study was conducted between May 2016 and October 2019, with the approval of the Research and Ethics Committee at Federal University of Rio Grande do Sul (UFRGS), under approval number 2383064. Parents, caretakers and children filled out a written and informed consent form before participating in the study.

Sample

A cross-sectional study of a randomized controlled clinical trial, with a sample of 34 children of both sexes, between the ages of 6 and 12 years old. These participants are speakers of Brazilian Portuguese and come from the southern region of Brazil. They were referred to the Faculty of Dentistry at the Federal University of Rio Grande do Sul for the surgical treatment of their lingual frenulum.

The study included children with alteration on lingual frenulum who were referred for frenectomy. Children with no upper and/or lower incisors, with head or neck malformations, cognitive impairments or hearing complaints were excluded from the study. Similarly, children with a patient history of surgery for their lingual frenulum or who had undergone or were undergoing speech language therapy were also excluded from the study.

The diagnosis of anomalies in the lingual frenulum was based on the results of the Tongue Frenulum Evaluation Protocol[23].

Assessment of lingual frenulum

The evaluation of the lingual frenulum of the patient sample was carried out by a single assessor, a speech language pathologist (E.G.), who used the Tongue Frenulum Evaluation Protocol[23]. To date, this is the only specific instrument published in Brazil with scores for the assessment of lingual frenulum. It consists of two sections: patient history including general complaints to be used with parents or caretakers; a clinical examination, which is divided into general and functional tests. During general tests, the assessor checks tongue lift, the percentage corresponding to maximum mouth opening (MMO) and MMO plus a measure of tongue tip placement on the incisive papilla (MMO +TTIP), as well as the attachment of the frenulum to the floor of the mouth and to the underside of the tongue. Based on the

type of attachment to these structures, the lingual frenulum of the sample were classified into: anterior attachment; short frenulum; anterior and short frenulum and ankyloglossia (when the frenulum is fused to the floor of the mouth). In the functional evaluation, besides speech, tongue mobility, tone and position at rest are assessed.

To evaluate general aspects, a score of zero is given for no anomalies while a score of one or two is given to represent degree of abnormality. When the sum of the scores of the general test is equal to or greater than three, the lingual frenulum is considered abnormal. When the sum of the scores of the functional tests is equal to or greater than 15, this is indicative of possible interference in oral functions, including speech, due to an atypical lingual frenulum.

The aforementioned assessment protocol[23] was incorporated into an indirect observation approach called anthroposcopy. Moreover, the evaluation was recorded in images and videos with the help of a Canon™ digital camera *PowerShot SX240HS,* placed one meter away from the child being examined. During the examination, the assessor sat facing the child. Each participant was asked to sit with their feet flat on the floor and to hold their head in a habitual position. The assessor used gloves procedure and a digital Digimes™ caliper, previously sterilized with 70% alcohol.

Evaluation of speech

The speech assessment was also performed using the same assessment protocol[23] and had three parts: 1) informal speech (we asked the children to answer these questions and tasks: “What's your name? How old are you? Do you study or work? Tell us about your school. Tell us about something interesting that happened to you”; 2) counting from 1 to 20; then naming the weekdays and the months of the year; 3) pointing to a board and naming 50 figures. The first 25 words contain all

phones in the Brazilian Portuguese language and the other half include phones which are more likely to be affected by the lingual frenulum during speech production. These include, the simple alveolar vibrant sound in all positions and alveolar fricative sounds. At every phase of the three-step speech test, patients presented omissions, substitutions and distortions. Another part of the speech test is an item called "other aspects to be observed during speech". Here the assessor pays attention to opening of the mouth, tongue position, jaw movement, speed, accuracy of speech as a whole and voice.

Analysis of the speech data was performed by means of auditory-perceptual evaluation. The video recordings of the first three speech tasks of all the children were analyzed by two independent speech language pathologists and judges (B.L.P.A. and C.E.S.). A third judge (B.C.B.) - also a speech language pathologist. The judges did not have contact with the assessor who used the assessment protocol (E.G.).

The two judges received the videos of the children's tests on a USB stick, along with a form specially drafted for this study. The form contained the same topics of speech assessment as the assessment protocol[23] with added sections for the analysis of speech repair strategies at the syllabic and segmental levels. The term speech repair strategies refers to the different patterns presented by the child as they try to implement phonological processes[24,25].

Data were analyzed descriptively through percentage distribution using 21 Statistics SPSS (SPSS Inc., Chicago, IL).

RESULTS

Of the 34 children in the sample, 20 patients (58.8%) were male and 14 patients (41.2%) were female. The mean age was 8.7 ± 2.2 years. Regarding the

chief complaint brought up by parents or guardians during the patient history interview, 17 (50%) spontaneously reported noticing speech problems. All parents and guardians were explicitly asked if their child or ward presented speech defects and, in addition to those who had already mentioned this as their main complaint with no prompting, another 9 (26.5%) also reported noticing speech problems. Thus, a total of 26 (76.4%) spoke about noticing a speech impediment.

The frenulum were classified as short in 10 (29.4%) children, short and anterior in 21 (61.8%) children and ankyloglossia in 3 (8.8%) participants.

The results of a functional test of elicited, automatic speech and a naming task, matched to the classification of the lingual frenulum, are detailed in Table 1. There were omissions, substitutions and distortions in patient speech and some children could present a combination of two or more of these speech problems.

The frequency of speech distortions for specific sounds, matched to the classification of the lingual frenulum, are described in Table 2.

Other aspects or general characteristics observed in patient speech, associated with the classification of lingual frenulum, are described in Table 3. The possibility was considered that participants might present a combination of two or more features.

The speech impediments classified in the assessment protocol[23], such as omission and substitution, are described through strategies of repair, at the syllabic and segmental level, and associated with the classifications of lingual frenulum in Table 4.

DISCUSSION

In our study sample, speech repercussions were frequent, mainly in the group of children with short and anterior frenulum, followed by children with short frenulum.

This finding corroborated the results of another study[2]. However, a different study reported a higher frequency of speech disorders in children with short frenulum[9]. This difference in findings may be due to a higher prevalence of subjects with short frenulum in their patient sample[9] than in our sample.

With regard to speech characteristics, all the children in our sample presented speech distortions. This corroborates other studies which have also found that children with alteration on lingual frenulum show these distortions[2,7,9]. In the sample studied, half of the children presented concomitant distortions, omissions and substitutions. The latter two were classified in this study as speech repair strategies. In addition to distortions, the authors of the studies cited above also highlighted that omissions[2,7] and substituições[7,9] were common. Regarding the classification of lingual frenulum in our study, these phonetic and phonological disorders were frequent in children with short and anterior frenulum. An anterior attachment of the lingual frenulum tends to limit movements of the tongue and the more anterior the attachment, the greater the probability of speech disorders[2].

As for the frequency of distortions in relation to specific sounds, most children produced distorted alveolar taps, consonant clusters with [r], the alveolar fricative [s] and the alveolar plosive [t]. Our findings are similar to the published results of other studies investigating speech disorders in individuals with alteration on lingual frenulum. These other authors report the distorted production of consonant clusters with [r], the alveolar flap [r][2,8,9,11-13] and the alveolar sounds [s], [z][2,11,13,] [t], [d] [l] and [n][2,8,11,12]. In this study, all these changes were similarly common in children with frenulum classified as ankyloglossia, except for the distortion of the alveolar fricative [s].

The distortion of alveolar phones is due to difficulties in lifting the anterior third

196 portion off the tongue, a movement which is necessary for the proper articulation of
197 these sounds. This inability to raise the anterior third of the tongue to touch the
198 alveolar region is due to the mechanical impediment caused by the shortening and/or
199 atypical attachment of the lingual frenulum[12]. It can be said that distortion occurs
200 when the speaker seeks to adjust or compensate for their speech disorder, in an
201 attempt to improve intelligibility. Distortion is considered a phonetic change that does
202 not concern the phonological rules of a language and is characterized by difficulties
203 in motor skills involved in the production of sound, such as position, time, stress and
204 speed, resulting in a non-standard sound[16].

205 As regards the analysis of other oral aspects during speech production,
206 narrowed mouth opening was noted in our patient sample. This reduction in mouth
207 opening can affect the accuracy of speech as a whole[2]. It occurs when the child tries
208 to reach the correct articulation points by reducing the space between the jaws
209 during speech[2,9]. A clinical study found that subjects with short frenulum open their
210 mouths less when they speak[9].

211 Other types of compensation observed in our analysis of speech were forward
212 placement of the tongue or visible sides of the tongue and excessive mandibular
213 lateralization. All of these can also impair the accuracy of speech, according to
214 another study[15]. Findings in the literature support the conclusion that the more
215 anterior the attachment of the tongue, the greater the restriction of tongue
216 movements and, consequently, the greater the impact on speech[2].

217 Unusual movements of the lips, tongue and jaw during speech production,
218 when presented by individuals with alteration on lingual frenulum, are directly or
219 indirectly related to the restriction of the vertical movements of the tongue and do not
220 necessarily interfere with speech intelligibility[26,27]. In our study, we observed the

following unusual movements during speech: unilateral lifting of the upper lip, lip vibration and cheek inflation when attempting to produce the [r] sound.

With regard to the speech repair strategies, the most frequent one noted in our sample was the reduction of consonant clusters, followed by the omission of final, non-lateral liquid sounds. Less frequently, we also observed the substitution of lateral and non-lateral liquids, as well as the gliding of non-lateral liquids. The inability to use appropriate tongue movements for the proper production of phonemes such as liquids may result in net omissions and substitutions, as has already been mentioned in previous studies[2,7,9]. During our research, we observed that anatomical changes related to the lingual frenulum interfere with the phonetic aspect of speech. In turn, this interference also seems to affect, to a greater or lesser extent, the phonological aspect of speech[2,9,22]. In a case report about phonological disorder and alteration on lingual frenulum, the authors reported on the presentation of the phonological processes of substitution and simplification which were difficult to correct, particularly when liquid sounds were involved[22].

In this study, some of the children in our patient sample with short lingual frenulum or short and anterior lingual frenulum presented devoicing as a speech repair strategy. It is worth noting that, concerning speech repair strategies involving voicing, an alteration on lingual frenulum may not be a cause since there is no participation of the tongue in the production of this sound feature; this is related to the issue of mode of sound articulation[16]. For this reason, this type of disorder changes are characterized as purely phonological.

The results of our study allow us to infer that praxis difficulty originating from alteration on lingual frenulum function may interfere with phonological acquisitions, more specifically in children with the most anteriorly attached and short frenulum. We

emphasize that the correction of atypical lingual frenulum should be performed as early as possible, to prevent the setting of inadequate speech patterns and avoid having to relearn and naturalize new motor and linguistic patterns[14,28].

In this research, we observed that the highest frequency of speech disorders were phonetic in origin. However, speech repair strategies were also observed and most of these were influenced by the uncharacteristic motor aspect. All the children in our sample presented distorted speech, most often with the alveolar consonants [ɾ], [s] and [t] and consonant clusters with the [ɾ] sound. The major speech repair strategy used by the participants was to reduce consonant clusters, followed by the tendency to omit non-lateral liquids in final position. The co-occurrence of phonetic and phonological disorders was more frequent in children with short and anterior frenulum.

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Table 1. Results of the functional speech test associated with lingual frenulum classification

Characteristics n (%)	Total (n=34)	Classification of lingual frenulum alteration		
		Short (n=10)	Anterior and short (n=21)	Ankyloglossia (n=3)
Omissions				
Yes	20 (58.8)	5 (50.0)	14 (66.6)	1 (33.3)
No	14 (41.1)	5 (50.0)	7 (33.3)	2 (66.6)
Substitutions				
Yes	21 (61.7)	5 (50.0)	15 (71.4)	1 (33.3)
No	13 (38.3)	5 (50.0)	6 (28.5)	2 (66.6)
Distortions				
Yes	34 (100)	10 (100)	21 (100)	3 (100)
No	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)

Table 2. Frequency of specific distorted phones associated with lingual frenulum classification

Distorted production n (%)	Total (n=34)	Classification of lingual frenulum alteration		
		Short (n=10)	Anterior and short (n=21)	Ankyloglossia (n=3)
[t]	20 (58.8)	5 (50.0)	12 (57.1)	3 (100)
[d]	10 (29.4)	3 (30.0)	7 (33.3)	0 (0.0)
[n]	5 (14.7)	2 (20.0)	3 (14.2)	0 (0.0)
[s]*	19 (55.8)	4 (40.0)	14 (66.6)	1 (33.3)
[z]	16 (47.0)	2 (20.0)	13 (61.9)	1 (33.3)
[ʃ]	7 (20.5)	1 (10.0)	6 (28.5)	0 (0,0)
[ʒ]	9 (26.4)	1 (10.0)	8 (38.1)	0 (0,0)
[l]	11 (32.3)	3 (30.0)	6 (28.5)	2 (66.6)
[ɾ]*	24 (70.5)	7 (70.0)	14 (66.6)	3 (100)
[s]**	16 (47.0)	3 (30.0)	11 (52.3)	2 (66.6)
[ɾ]**	24 (70.5)	5 (50.0)	16 (76.1)	3 (100)
[ɾ]***	25 (73.5)	7 (70.0)	15 (71.4)	3 (100)
[l]***	14 (41.1)	3 (30.0)	9 (42.8)	2 (66.6)

Legend: *Simple onset; ** Final position; ***Complex onset

Table 3. Speech characteristics of the children associated with lingual frenulum classification

Characteristics n (%)	Total (n=34)	Classification of lingual frenulum alteration		
		Short (n=10)	Anterior and short (n=21)	Ankyloglossia (n=3)
Mouth opening				
Adequate	12 (35.2)	2 (20.0)	10 (47.6)	0 (0.0)
Reduced	22 (64.7)	8 (80.0)	11 (52.3)	3 (100)
Tongue position				
Adequate	16 (47.0)	6 (60.0)	10 (47.6)	0 (0.0)
Mouth floor	4 (11.7)	1 (10.0)	2 (9.5)	1 (33.3)
Anterior	10 (29.4)	3 (30.0)	6 (28.5)	1 (33.3)
Visible sides	11 (32.3)	1 (10.0)	8 (38.0)	2 (66.6)
Mandibular movement				
Adequate	26 (76.4)	9 (90.0)	16 (76.1)	1 (33.3)
Deviation to the right	4 (11.7)	1 (10.0)	2 (9.5)	1 (33.3)
Desviation to the left	4 (11.7)	0 (0.0)	4 (19.0)	2 (66.6)
Speech rate				
Adequate	26 (76.4)	7 (70.0)	17 (80.9)	2 (66.6)
Increased	4 (11.7)	1 (10.0)	2 (9.5)	1 (33.3)
Reduced	4 (11.7)	2 (20.0)	2 (9.5)	0 (0.0)
Voice quality				
Adequate	31 (91.1)	8 (80.0)	20 (95.2)	3 (100)
Altered	3 (8.8)	2 (20.0)	1 (4.7)	0 (0.0)

Table 4. Speech repair strategies in children associated with lingual frenulum classification

Speech repair strategies n (%)	Total (n=34)	Classification of lingual frenulum alteration		
		Short (n=10)	Anterior and Short (n=21)	Ankyloglossia (n=3)
Syllablic level				
Consonant cluster reduction	18 (52.9)	4 (40.0)	13 (61.9)	1 (33.3)
Metathesis	3 (8.8)	0 (0.0)	3 (14.2)	0 (0.0)
Deletion of final liquids	14 (41.1)	3 (30.0)	10 (47.6)	1 (33.3)
Deletion of intervowel liquids	4 (11.7)	1 (10.0)	3 (14.2)	0 (0.0)
Deletion of initial liquids	2 (5.8)	1 (10.0)	1 (4.7)	0 (0.0)
Deletion of final fricatives	2 (5.8)	0 (0.0)	2 (9.5)	0 (0.0)
Segmental level				
Substitution of liquids	14 (41.1)	4 (40.0)	9 (42.8)	1 (33.3)
Gliding of líquids	10 (29.4)	2 (20.0)	7 (33.3)	1 (33.3)
Fronting	9 (26.4)	2 (20.0)	7 (33.3)	0 (0.0)
Backing	4 (11.7)	0 (0.0)	4 (19.0)	0 (0.0)
Devoicing	7 (20.5)	2 (20.0)	5 (23.8)	0 (0.0)
Voicing	1 (2.9)	1 (10.0)	0 (0.0)	0 (0.0)
Assimilation	1 (2.9)	0 (0.0)	0 (0.0)	1 (33.3)
Epenthesis	1 (2.9)	0 (0.0)	1 (4.7)	0 (0.0)

4 CONSIDERAÇÕES FINAIS

Baseado nos resultados da revisão sistemática, proposta no primeiro artigo desta tese, os métodos de diagnóstico da alteração do frênulo lingual apresentaram variabilidade entre os estudos incluídos. Estes critérios utilizados não foram validados e a comparação prospectiva dos seus métodos com um padrão proposto é inexistente. O método de mensuração das alterações de fala foi realizado com diferentes instrumentos em todos os estudos, verificando-se dificuldade de comparação entre os resultados. Neste sentido, a heterogeneidade entre os métodos evidencia a carência de padronização de instrumento específico. Assim, um maior investimento no estudo de protocolo que avalia a fala nas alterações de frênulo lingual, tanto na construção quanto na tradução, adaptação e validação para diferentes países se faz necessário. Protocolos específicos favorecem a condução de uma melhor tomada de decisões clínicas de diagnóstico e opinião de especialistas da área de forma semelhante, podendo definir prática clínica baseada em evidência de qualidade. Ainda, a não delimitação de uma faixa etária, ou investigação das alterações de fala em crianças em período de aquisição de fala, podem ter sido um fator de confusão relevante para a análise dos resultados em alguns dos estudos. Portanto, a qualidade da evidência foi avaliada e foi classificada como de baixa qualidade nos estudos, e estes são insuficientes para afirmar se a presença da alteração de frênulo lingual implica em alterações de fala.

Na análise da efetividade da frenectomia associada ou não à terapia miofuncional proposta pelo estudo de ensaio clínico randomizado desta tese, os aspectos gerais e funcionais melhoraram em diferentes graus após a frenectomia e após a terapia miofuncional. A frenectomia foi eficiente para melhorar a elevação e mobilidade da língua, assim como a fala. A terapia miofuncional promoveu evolução na elevação e mobilidade da língua, além de melhora em outros aspectos da fala como a abertura de boca, posição da língua e imprecisão na fala. No entanto, quando comparadas as crianças que realizaram frenectomia associada ou não à terapia miofuncional com o grupo de crianças sem alteração de frênuo lingual, os resultados dos escores do protocolo da mobilidade de língua e função da fala e seus aspectos ainda carecem de terapia miofuncional para que os desfechos sejam semelhantes às crianças sem a alteração no frênulo lingual. Este fato demonstra a importância da

atuação conjunta entre a Fonoaudiologia e a Odontopediatria.

Ao considerar a investigação das características da fala de crianças com alteração do frênulo da língua, proposta no terceiro artigo desta tese, por meio do recorte do estudo com delineamento de ensaio clínico, as distorções foram observadas em todas as crianças da amostra, com maior frequência destas no tepe alveolar, grupos consonantais com [r], fricativa alveolar [s] e a plosiva alveolar [t]. Pode-se afirmar que a distorção ocorre quando a criança busca ajustes ou compensações para uma fala mais inteligível. No que se refere à análise de outros aspectos observados durante a produção oral, foi verificado a redução da abertura de boca, bem como outras compensações como posição da língua anteriorizada ou com laterais visíveis e movimentos mandibulares excessivos de lateralização, os quais também podem prejudicar a precisão da fala. No que diz respeito às estratégias de reparo, a mais frequente na amostra foi a redução de encontro consonantal, seguida de apagamento de líquida não-lateral final. Os resultados encontrados permitem inferir que dificuldades práxicas em função de alteração de frênulo lingual podem interferir em aquisições nos aspectos fonológicos, mais especificamente em crianças com frênulo com a inserção mais anteriorizada e com a extensão deste mais curta.

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ANEXO – Protocolo de Avaliação do Frênulo da Língua (MARCHESAN, 2010)

PROTOCOLO PARA AVALIAÇÃO DE FRÊNULO DE LÍNGUA

ANAMNESE

Nome: _____ **Sexo F ( ) M ( )**

Data do exame: __ / __ / __ **Idade:** ___ anos e ___ meses **DN:** __ / __ / __

Informante: _____ **Grau de parentesco:** _____

Estuda: ❒ sim Em que série está: ❒ não Até que série estudou:

Trabalha: ❒ sim Em que: ❒ não

Já trabalhou: ❒ não ❒ sim Em que:

Atividade física: ❒ não ❒ sim Qual:

Endereço: _____ **Nº:** _____ **Complemento:** _____

Bairro: _____ **Cidade/Estado:**)_____ **CEP:** _____

Fones: Residencial: (____) _____ Trabalho: (____) _____ Celular: (____) _____

Endereço eletrônico:_____

Nome do pai:_____ **Nome da mãe:** _____

Irmão: ❒ não ❒ sim Quantos: _____

Quem indicou para avaliação fonoaudiológica? *(Nome, especialidade e telefone)*:

_____

Qual a razão da indicação: _____

Queixa principal: _____

Queixas diversas relacionadas à:
(0) não (1) às vezes (2) sim

( ) lábios	( ) língua	( ) sucção	( ) mastigação	( ) deglutição
( ) respiração	( ) fala	( ) frênulo lingual	( ) voz	( ) audição
( ) aprendizagem	( ) estética facial	( ) postura	( ) oclusão	( ) cefaléia freqüente
( ) ruído na ATM	( ) dor na ATM	( ) dor no pescoço	( ) dor nos ombros	
( ) dificuldade ao abrir a boca	( ) dificuldade de movimentar a mandíbula para os lados			( ) Outras

Antecedentes Familiares – investigar se existem casos na família com alteração de frênulo de língua

❒ não ❒ sim Quem e qual o problema:

Problemas de Saúde

❒ não ❒ sim Quais:

Problemas respiratórios

❒ não ❒ sim Quais:

Amamentação

Peito: ❒ sim Até quando: _____ ❒ não

Mamadeira: ❒ sim Até quando: _____ ❒ não

A criança teve dificuldade de sugar o peito? ❒ não ❒ sim Se sim qual(is) dificuldade(s)?_____

Alimentação – dificuldades com a mastigação

❒ não ❒ sim Quais:

Alimentação – dificuldades com a deglutição

❒ não ❒ sim Quais:

Hábitos Orais:

❒ não ❒ sim Quais:

Apresenta alteração de fala

❒ não ❒ sim Quais:

Caso tenha alteração de fala, isto causa alguma dificuldade no relacionamento social e ou profissional?

❒ não ❒ sim Social ❒ não ❒ sim Como reage: _____
Profissional ❒ não ❒ sim Como reage:

Apresenta alteração de voz

❒ não ❒ sim Quais:

Fez cirurgia de frênulo da língua

❒ não ❒ sim Quando: _____ Quantas vezes: _____
Especialidade do profissional que operou: _____
Que tipo de cirurgia foi feita? _____
O que achou do resultado: ❒ bom ❒ médio ❒ ruim

Acrescente outras informações que considerar importantes para o caso:

_____
_____
_____
_____
_____
_____
_____
_____
_____
_____

EXAME CLÍNICO

PARTE I – PROVAS GERAIS

Mensurar utilizando paquímetro. Maior ou igual a 50,1% (0) menor ou igual a 50% **(1) Resultado =**

Medir da borda do incisivo superior, até a borda do incisivo inferior direito ou esquerdo. Utilizar os mesmos dentes para as duas medidas.	**Valor encontrado em milímetros**
Abertura máxima de boca	
Abertura máxima de boca com o ápice da língua tocando na papila incisiva	
Relação entre estas medidas, em percentagem	%

Alterações durante a elevação da língua (melhor resultado = 0 e pior = 2) Resultado =

Abrir a boca totalmente, elevar a língua dentro da boca sem tocar no palato e observar:	**NÃO**	**SIM**
1. A ponta da língua fica com formato retangular ou quadrado	(0)	(1)
2. A ponta da língua forma um "coração"	(0)	(1)

Fixação do frênulo. Somar A e B (melhor resultado = 0 e pior = 3) Resultado =

A – No assoalho da boca:	
Visível somente a partir das carúnculas sublinguais (saída dos ductos submandibulares)	(0)
Visível já a partir da crista alveolar inferior	(1)

Fixação em outro ponto: _____

B – Na face inferior da língua (face sublingual):	
Na parte média	(0)
Entre a parte média e o ápice	(1)
No ápice	(2)

Classificação clínica do frênulo (melhor resultado = 0 e pior = 2) Resultado =

Normal (0)	Gera dúvida (1)	Alterado (2)

Caso o frênulo tenha sido considerado alterado seria porque:

A fixação do frênulo é anteriorizado	O frênulo é de tamanho curto	O frênulo é curto e anteriorizado	
Anquiloglossia (fusão do frênulo no assoalho)	Outro -		Não sei

Total geral para as provas gerais: melhor resultado = 0 pior = 8

Quando a soma das provas gerais for igual ou maior que três, pode-se considerar o frênulo como alterado.

PARTE II - PROVAS FUNCIONAIS

Mobilidade da língua (melhor resultado = 0 e pior = 14). Resultado =

	executa	executa aproximado	não executa
Protrair e retrair	(0)	(1)	(2)
Tocar o lábio superior com o ápice	(0)	(1)	(2)
Tocar o lábio inferior com o ápice	(0)	(1)	(2)
Tocar a comissura labial à direita	(0)	(1)	(2)
Tocar a comissura labial à esquerda	(0)	(1)	(2)
Vibrar o ápice	(0)	(1)	(2)
Sugar no palato	(0)	(1)	(2)

Posição da língua durante o repouso (melhor resultado = 0 e pior = 4). Resultado =

Não se vê (mantém a boca fechada)	(0)
No assoalho da boca	(1)
Entre os dentes anteriormente	(2)
Entre os dentes lateralmente	(2)

Fala (melhor resultado = 0 e pior =12) Resultado =

Prova nº 1 - Fala informal
Como é seu nome? Quantos anos você tem? Você estuda/ trabalha? Fale um pouco sobre sua escola/ trabalho. Conte um fato interessante que ocorreu com você.
Prova nº 2 – Solicitar contagem de 1 a 20; em seguida, os dias da semana e, por último, os meses do ano.
Prova nº 3 – Solicitar a nomeação das figuras da prancha

Provas de fala	OMISSÃO		SUBSTITUIÇÃO		DISTORÇÃO	
	Não	Sim	Não	Sim	Não	Sim
1	(0)	(1)	(0)	(1)	(0)	(2)
2	(0)	(1)	(0)	(1)	(0)	(2)
3	(0)	(1)	(0)	(1)	(0)	(2)

Assinale quais são os sons ou grupos de sons que se apresentam com alguma alteração. Se a alteração ocorre em uma ou duas provas apenas, marque ao lado do som o número da prova onde ocorreu a alteração.

p		b		t		d		k		g		m		
n		ɳ		f		v		s		z		ʃ		
ʒ		l		ɾ́		ɾ		x		{S}		{R}		
pr	br	tr	dr	cr	gr	fr	vr	pl	bl	cl	gl	fl	vl	tl

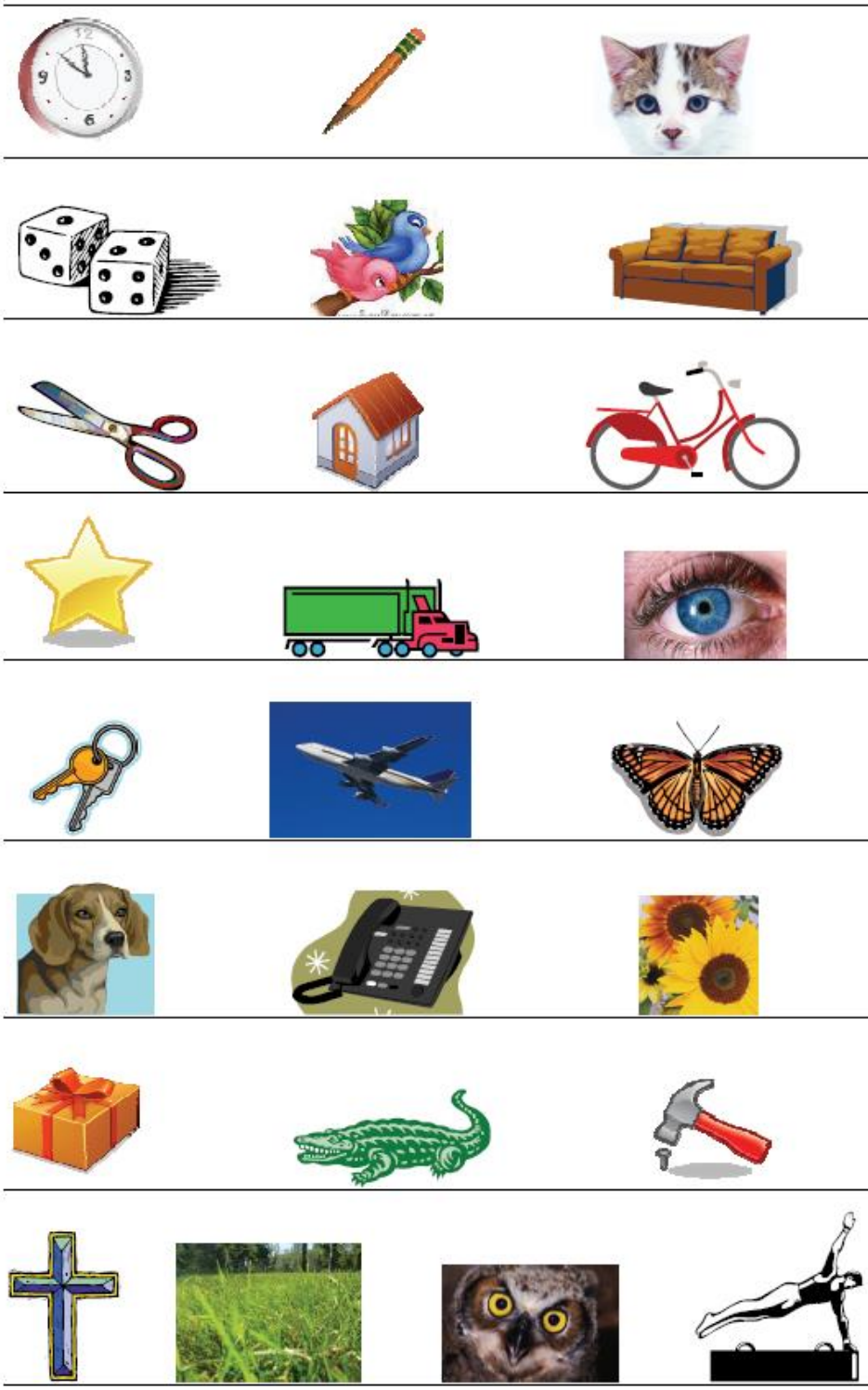
Outros aspectos a serem observados durante na fala (melhor resultado = 0 e pior =10) Resultado =

Abertura da boca: (0) adequada (1) reduzida (1) exagerada
Posição da língua: (0) adequada (1) no assoalho (2) anteriorizada (2) com laterais visíveis
Movimento mandibular: (0) sem alteração (1) desviado à direita (1) desviado à esquerda (1) anteriorizado
Velocidade: (0) adequada (1) aumentada (1) reduzida
Precisão da fala como um todo: (0) adequada (1) alterada
Voz: (0) sem alteração (1) alterada

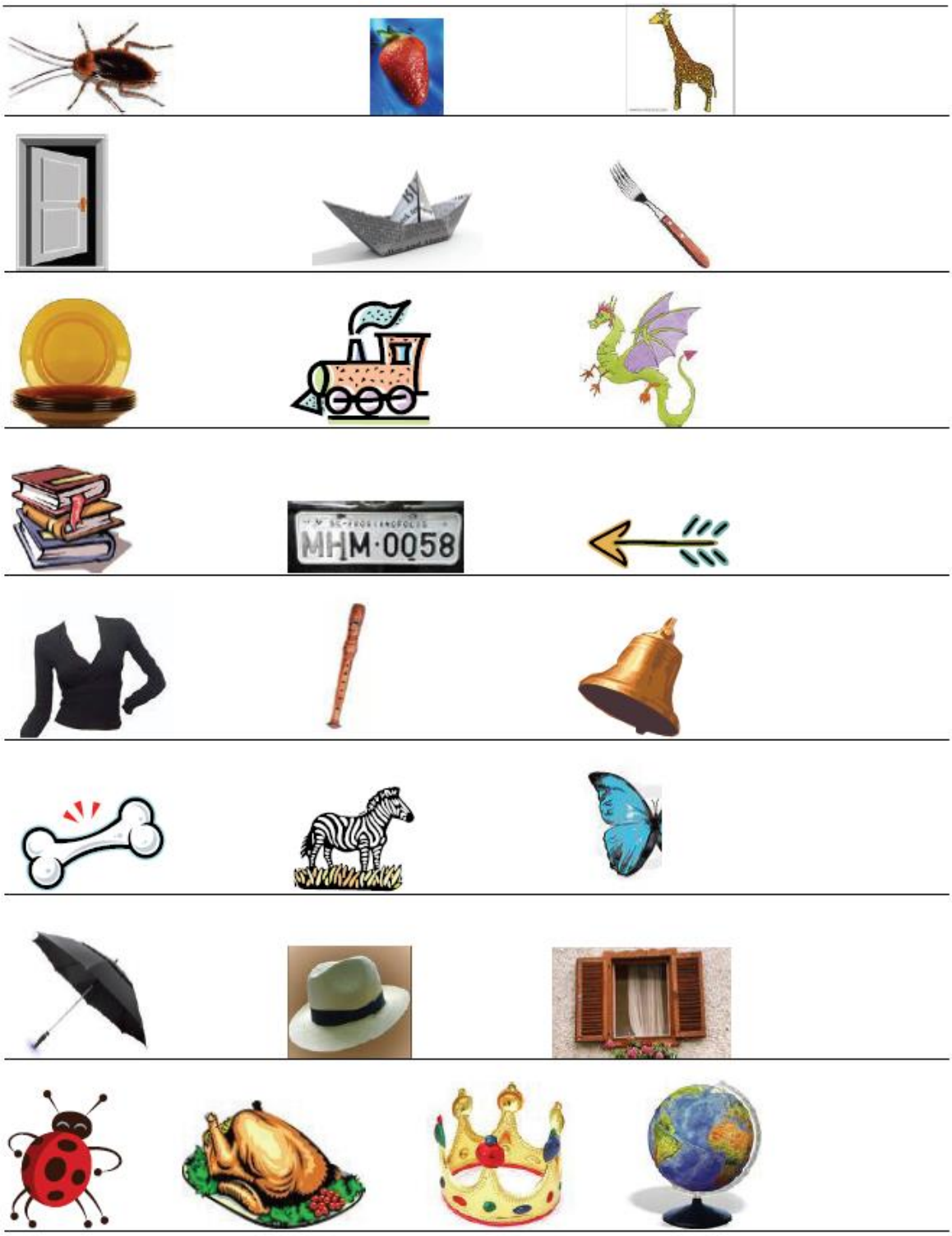
Total geral para as provas que avaliam a funcionalidade: melhor resultado = 0 e pior = 40

Quando a soma das provas funcionais for igual ou maior que 25, pode-se considerar a possível interferência do frênulo da língua.

Documentação
Sugerem-se fotos e filme das provas de: mobilidade da língua e as de fala.



Prancha com figuras para a avaliação da fala



Prancha com figuras para a avaliação da fala