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ORIGINAL ARTICLE

Relationship between short lingual frenulum and malocclusion. A multicentre study

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KEYWORDS
Ankyloglossia;
Malocclusion;
Short lingual frenulum

Abstract
Objective: Ankyloglossia is characterized by an abnormally short lingual frenulum, which impairs tongue movement. Ankyloglossia has been related to craniofacial growth disturbances and dental malocclusion. But even though there is a clear biological plausibility for this hypothesis, available evidence is scarce.
Methods: A case–control design was followed. Patients between 4 and 14 years old were routinely screened for short lingual frenulum and recruited from the pediatric Otolaryngology consultation of 3 Spanish tertiary referral hospitals. Lingual frenulum was assessed with the Marchesan system. A cohort of cases with short lingual frenulum and a cohort of healthy controls matched for sex and age were included. Both cases and controls had pictures of occlusion. Occlusion was evaluated by an expert in orthodontics, blinded for the frenulum assessment.

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Results: A total of 100 participants were included, 70 males and 30 females. The proportion of malocclusion in the short lingual frenulum group was 48%, while it was 24% in the normal frenulum group. The odds ratio of malocclusion for the short lingual frenulum patients was 2.92 (CI 95% 1.15–7.56). The difference was statistically significant ($p = .012$). This difference was significant for patients with class III occlusion ($p = .029$). There was no difference for patients with class II ($p = .317$).
Conclusions: This work supports the hypothesis that relates class III malocclusion with a short lingual frenulum.

PALABRAS CLAVE
Anquiloglosia;
Maloclusión;
Frenillo lingual corto

Relación entre frenillo lingual corto y maloclusión. Un estudio multicéntrico

Resumen
Objetivo: La anquiloglosia se caracteriza por un frenillo lingual anormalmente corto que dificulta la movilidad de la lengua. La anquiloglosia ha sido relacionada con alteraciones del desarrollo facial y maloclusión dentaria. Sin embargo, a pesar de una clara plausibilidad biológica para esta hipótesis, la evidencia disponible es escasa.
Métodos: Siguiendo un diseño de casos y controles se incluyeron pacientes entre 4-14 años atendidos en las consultas de otorrinolaringología pediátrica de 3 hospitales de tercer nivel en España. El frenillo lingual se evaluó mediante el sistema de Marchesan. Se incluyó una cohorte de casos con frenillo lingual corto, y una cohorte de controles sanos apareados por sexo y edad. A todos los participantes se les tomó fotografía dentaria en oclusión. La oclusión se evaluó mediante un odontólogo experto en ortodoncia, ciego a la evaluación del frenillo lingual.
Resultados: Se incluyeron un total de 100 participantes, 70 hombres y 30 mujeres. La proporción de maloclusión en la cohorte con frenillo lingual corto fue del 48% y del 24% en la cohorte de controles. La odds ratio de maloclusión fue 2,92 (IC 95%: 1,15-7,56). La diferencia entre grupos fue estadísticamente significativa (p = 0,012). Por subgrupos, la diferencia fue estadísticamente significativa para los pacientes con maloclusión clase III (p = 0,029), pero no para aquellos con maloclusión clase II (p = 0,317).
Conclusiones: Este trabajo apoya la hipótesis que relaciona la maloclusión de clase III con el frenillo lingual corto.

Introduction

The lingual frenulum is a vestigial embryological element. An appropriate apoptosis process separates the tongue from the primitive pharynx during embryogenesis. The ankyloglossia, also called tongue-tie, is characterized by an abnormally short lingual frenulum, which impairs tongue movement.

Despite differences between series, such condition occurs in 4–11% of newborns, with a preference for male in a proportion of 2.5:1.[1] It may be associated with genetic syndromes such as Beckwith Wiedeman, Opitz, Simosa, Kindler's, van der Woude, Ehler Danlos, or oro-faciodigital syndrome, but most times it is isolated.[2]

There is considerable controversy regarding its diagnosis, clinical significance and management.[3] Most ankyloglossia research is focused on breastfeeding, swallowing and speech difficulties. However, some recent systematic reviews have pointed out the lack of quality evidence on those very aspects.[1,4]

Some research has also related ankyloglossia to alterations of the craniofacial growth and dental malocclusion. But, even though there is a clear biologic plausibility for this hypothesis, the available evidence is scarce, and their study design did not provide high levels of evidence.

The hypothesis behind those studies are that the high tongue position, placed on hard palate, stimulates the intermaxillary synchondrosis and, therefore, is one of the main factors guiding the maxilla's growth.[5] Short lingual frenulum results in restricted tongue tip movement, which impairs the tongue function forcing a low tongue position. This low posture may cause a forward and downward pressure against the anterior body of the mandible with different consequences on the face development, mainly class III malocclusion and anterior open bite.

Most of the available evidence on this topic are case reports and case series. Only 4 studies have been conducted

on children,[6–9] but they had some important design issues, since they used anatomical classifications of the lingual frenulum, did not used controls, and they had a selection bias as the selected patients were seeking orthodontic treatment.

In this research we aim to analyze the relationship between short lingual frenulum and malocclusion in children, aiming to control potential bias and to add knowledge to this unexplored field of pediatric otolaryngology.

Patients and methods

Sample

The sample was consecutively recruited from the pediatric Otolaryngology consultation of 3 Spanish tertiary referral hospitals by 3 investigators (CCH, XMR, FBR).

Patients between 4 and 14 years old were routinely screened for short lingual frenulum. Controls were selected from children with normal lingual frenulum, matched for sex and age with cases.

Exclusion criteria were the presence of genetic syndromes, history of frenectomy or orthodontic treatments. Also, those who did not have sufficient cognitive skills to take pictures were not included. Cognitive skills were assessed by interview and by reviewing existing medical records of cognitive impairment.

Both cases and controls had pictures of the occlusion using lip retractors (Fig. 1).

Lingual frenulum evaluation

To classify a patient as having ankyloglossia there must be an anatomical and a functional criterion.

We used the Marchesan system, which covers both function and form, and was validated for children.[10] We used the clinical examination, which can be scored from 0 to 8, patients are positively diagnosed with short lingual frenulum with a score of 3 or more.

Occlusion

Occlusion was evaluated by an expert in dentofacial orthopedics and orthodontics (SMN), blinded for the frenulum assessment. Pictures were taken in a centric position, guiding the mandible to prevent pseudo-class III malocclusion.[11]

Occlusion was evaluated according to Angle's classification. Angle's classification is based on where the buccal groove of the mandibular first molar contacts the mesiobuccal cusp of the maxillary first molar (Fig. 2): on the cusp (class I); distal to the cusp by at least the width of a premolar (class II); mesial to the cusp (class III). The distal occlusal plane is a method to assess the tendency to develop malocclusion.[12]

Statistical analysis

Descriptive analysis was used to report the sample characteristics. To analyze the differences between groups we used the student *t* test. To assess the relation between the 3 types of malocclusion Anova test was used, and to assess the relationship between lingual frenulum and malocclusion Chi square test was performed. Statistical significance was determined at $p < 0.05$. All statistical data were analyzed with STATA for Macintosh v. 15.1 (StataCorp®).

Ethical considerations

The study was performed in accordance with the ethical standards laid down in the Declaration of Helsinki. The Research and Ethics Committee of the Hospital Complex of Santiago de Compostela approved the study protocol (reference 2018/198).

Results

Participants

A total of 100 participants were included, 70 males and 30 females, ratio 2.3:1. Median age was 9.36, percentile 25–75 (6.68–12.2) and the range 4.12–14.86. A description of the sample is shown in Table 1.

There were no differences between groups regarding sex or age.

Lingual frenulum

Among the case group, the mean Marchesan score was 5.48, SD 1.47. Range 3–8, percentile 25–75 (5–6).

Malocclusion

The distribution of the occlusion type between groups is shown in Table 2. The proportion of malocclusion in the

Figure 1 Occlusion pictures.

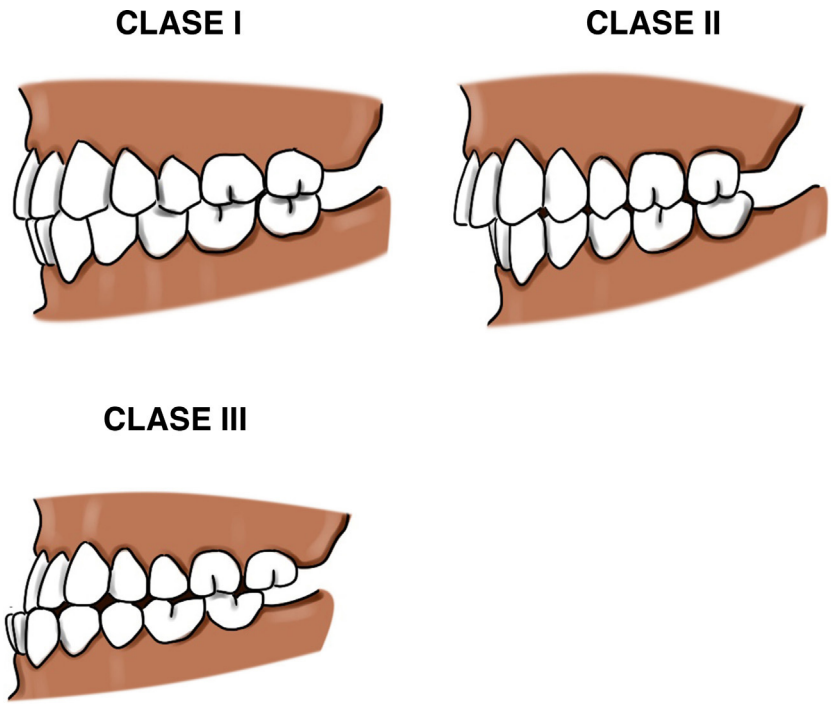

Figure 2 Occlusion.

Table 1 Sample description.

	Total	Short frenulum	Normal frenulum
Age (median, percentile 25–75) (range)	9.36 (6.68–12.2) (4.12–14.86)	9.53 (6.73–12.19) (4.12–14.86)	9.30 (6.67–12.22) (4.5–14.41)
Sex (male, female)	35, 15	35, 15	35, 15

Table 2 Occlusion type.

	Class I (*n*, %)	Class II (*n*, %)	Class III (*n*, %)
Short frenulum	26 (52%)	12 (24%)	12 (24%)
Normal frenulum	38 (76%)	8 (16%)	4 (8%)

short lingual frenulum group was 48%, while it was 24% in the normal frenulum group.

The odds ratio of malocclusion for short lingual frenulum patients was 2.92 (CI 95% 1.15–7.56). The difference was statistically significant ($p = .012$). This difference was significant for class III occlusion ($p = .029$), but there was no difference for class II ($p = .317$).

The Marchesan score increase with the occlusion classification ($p = .0011$) (Fig. 3).

Discussion

In this study we found a significant statistical association between malocclusion and short lingual frenulum. Specifically, with class III malocclusion. Higher degrees of tongue-tie were related to a higher risk of class III malocclusion. A weakness of our work is that we did not used cephalometry, since it is unethical to radiate a child without a clinical objective. Also, children were selected not from general population, but from a specialized outpatient clinic, which may introduce a selection bias. This work adds some extra evidence to this very topic. Despite solid conclusions cannot be obtained given the methodology, the risk-benefit relation support us to recommend routine examination of the lingual frenulum in patients with malocclusion, and to consider malocclusion as a symptom in patients with short lingual frenulum. In both cases treatment of the lingual frenulum should always be considered, both surgical and non-surgical.

The working hypothesis is solid. The functional matrix theory by Moss and Rankow suggests that the soft tissues guide the hard tissues.[13] Egil Harvold experimentally

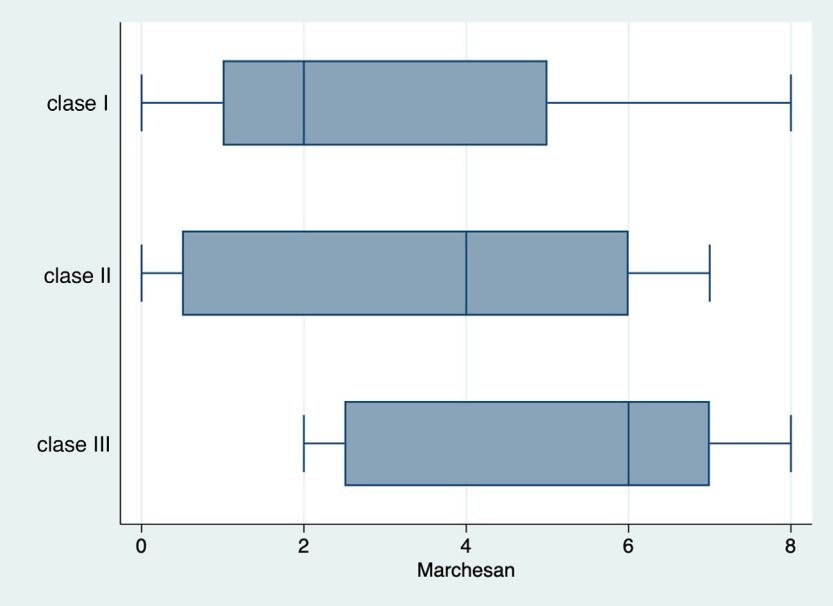

Figure 3 Marchesan score and type of occlusion.

demonstrated in primates that when tongue do not rest against the palate it causes craniofacial malformations and malocclusion.[14] To support this hypothesis, it has been shown that patients with class III malocclusion have the lowest tongue position.[15]

Our findings are in line with 2 of the 4 previous studies in children.[7,9] It must be noted that those findings have been also found in adults.[16,17]

The most prominent analysis was carried out by Vaz et al., who studied 700 children aged 9–17.[7] They described an association between short lingual frenulum with both class II and III malocclusion. However, half of their sample was selected from special schools, and they only used Kotlow classification, an anatomical classification. The second noticeable study was from Meenakshi et al.[9] They found relationship between the degree of tongue-tie to the risk of developing class III malocclusion. The flaw of this work was the selection bias, since they used patients seeking orthodontic treatment.

Finally, 2 authors did not find any kind of relationship.[6,8] García-Pola et al. studied 962 children going to dental evaluation, and did not find significant relationship between short lingual frenulum with any type of dentofacial anomaly.[6] However, their study was limited to six-year old children, and it is known that malocclusion tends to worsen with age and permanent dentition. Sepet et al. studied 80 patients aged 7–12 diagnosed with short lingual frenulum and did not find relationship between ankyloglossia and malocclusion types.[8] However, same as Meenakshi et al., sample was recruited from patients going to orthodontic treatment.

The available evidence offers opposite conclusions. However, the selection protocols and the methods used to diagnose patients differ among studies, which limit us to obtain strong scientific conclusions.

The main limitation of this work is intrinsic to its design. Transversal studies do not allow to know a temporal sequency. Therefore, despite there might be a relationship between short lingual frenulum and class III malocclusion, it cannot be known if the short lingual frenulum was prior to the malocclusion. Furthermore, the case control studies are prone to bias. Despite we tried our best to overcome those potential bias through blind assessment of the occlusion, and using age and sex matched controls, some bias could have been remained uncontrolled.

A strength of our work is that we used matched controls for age and sex, unlike the rest of authors. Yoon et al. demonstrated among 1052 children, that age is related to lingual frenulum and to the size of the jaw.[18] The size of the jaw has also a sex relationship.[19]

The examination of the lingual frenulum should be systematic among professionals dealing with pediatric patients, including the posterior tongue tie, which is often neglected.[20] There are several published methods which aim to diagnose short lingual frenulum. Recently, emphasis has shifted from definitions based merely on the anatomy of the frenulum attachment to a more functional description focus on the patient' symptoms. The working hypothesis is based on the function of the tongue, and not its shape. Diagnosis of a short lingual frenulum should always be based on a combination of form and function. Furthermore, an adequate diagnosis should be also based on its symptoms, such as motility, deglutition and speech impairment, sleep apnea,[21] and malocclusion, among others.

The Marchesan method is one of the most spread diagnostic methods.[10] It is recommended by a Cochrane review,[1] the Walsh and Tunkel review,[4] and by the work of Yoon et al.,[18] in which they concluded that Marchesan is the only independent measurement of tongue mobility that is directly associated with restrictions in tongue function.

The Kotlow technique is also very popular.[22] It measures the ''free tongue'', which is defined as ''the length of tongue from the insertion of the lingual frenulum into the base of the tongue to the tip of the tongue'', and considers a normal frenulum length to be > 16 mm. It was used by Vaz

et al.[7] and Sepet et al.[8] The problem with this method is that children may not collaborate for its precise measurement, and also measures are not age adjusted. Furthermore, it only considers the anatomic diagnosis.

Medical organizations such as the American Academy of Pediatrics and the National Institute for Health and Care Excellence now acknowledge that tongue-tie, or ankyloglossia is a significant clinical entity that should be treated as early as possible to minimize problems. Treatment includes surgical and non-surgical options, such as myofunctional therapy, or oral appliances such as the tongue pearl and tongue elevator. However, controversy arise about when a short lingual frenulum should be surgically managed, since it may regret spontaneously before 5 years-old, and if it persists there's a chance to be treat it with myofunctional therapy. Therefore, we suggest to only treat severe cases before this age. After this age, and when non-surgical methods fail, we suggest treating all cases. Myofunctional therapy have shown to improve form and function in short lingual frenulum before and after surgery,[23] therefore, management should always be accompanied by motility exercises.

Same as Vaz et al.,[7] we suggest considering malocclusion as a symptom of short lingual frenulum. Therefore, when facing a patient with malocclusion and short lingual frenulum, treatment of this condition should be included in the treatment protocol.

Regarding safety of the procedure, it is clear that the complication rate should be in accordance with the morbidity of the condition being treated. Given the low morbidity of the tongue tie, a low complication rate is mandatory. Previous reviews have reported frenotomy to be a safe surgery.[4,23,24] Also a review from the Canadian agency for drugs and technologies in health stated this surgery as a safe procedure.[25] None of the consulted authors reported severe complications. It must be mentioned that some severe complications have been reported, including Ludwig angina, severe bleeding, or airway obstruction in patients with Pierre Robin Syndrome. However, overall, frenotomy has proven to be a low-risk procedure that is likely to be beneficial with careful patient selection.

Conclusions

Our work supports the hypothesis which relates class III malocclusion with a short lingual frenulum. In this work we tried to overcome some design flaws from previous studies.

There are several factors which can influence occlusion and facial growth, such as genetics, oral breathing, atypical deglutition, among many others. Furthermore, we do not clearly understand how and why malocclusion occurs, therefore we cannot control all those factors. Larger samples and randomized clinical trials are needed to clarify these points.

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Conflict of interest

The authors declare not to have any conflict of interest.

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