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Review

Outcomes of tongue-tie release by means of tongue and frenulum assessment tools: a scoping review on non-infants

Impatto della correzione dell'anchiloglossia nei giovani e negli adulti: revisione mediante valutazioni frenulo-linguali

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SUMMARY

Objective. To evaluate outcomes of the surgical and rehabilitative procedures devoted to release the tongue-tie in non-infants when implementing the most commonly used quantitative/qualitative structured tools for tongue and frenulum assessment.

Methods. A scoping review and meta-analysis were conducted following the guidelines of the Preferred Reporting Items for Systematic Review and Meta-Analysis extension for scoping reviews.

Results. The systematic search retrieved 603 (Pubmed), 893 (Scopus), and 739 (ISI Web of Science) articles from January 2011 to December 2021. A total of 50 articles were retrieved for full-text review of which 7 were selected and included based on inclusion criteria. The majority of treatment options have been found to significantly improve the anatomical limitation of the tongue with clear benefits on descending functionality.

Conclusions. The review highlights an overall improvement in terms of clinical and functional outcomes when using validated tongue assessment tools both before and after frenulum release. This highlights the need for their rigorous implementation in research and clinical practice.

KEY WORDS: tongue-tie release, ankyloglossia, scoping review, Kotlow classification, Marchesan protocol

RIASSUNTO

Obiettivo. *Considerato il dibattito che ruota attorno alle tecniche correttive dell'anchiloglossia in soggetti giovani e adulti, l'obiettivo della presente revisione è quello di chiarire come tali tecniche impattino in soggetti con età superiore ad un anno in termini di risultati quando valutati mediante i principali strumenti quali-quantitativi di valutazione frenulo-linguale.*

Metodi. *La revisione di scopo e la meta-analisi – in accordo alle attuali linee guida – sono state eseguite a seguito di una ricerca sistematica che ha condotto a valutare 603, 893 e 739 articoli provenienti rispettivamente da Pubmed, Scopus e ISI Web of Science.*

Risultati. *50 di questi articoli sono stati consultati in estenso e 7 selezionati sulla base dei criteri di inclusione. La maggior parte dei trattamenti eseguiti ha dimostrato di migliorare le limitazioni funzionali della lingua con considerevole miglioramento delle funzioni ad essa collegate.*

Conclusioni. *La presente revisione ha mostrato un globale miglioramento in termini clinici e funzionali esplorati mediante test di valutazione frenulo-linguale sia prima che dopo il trattamento correttivo e rimarca una loro rigorosa implementazione nella pratica clinica e di ricerca.*

PAROLE CHIAVE: *frenuloplastica, anchiloglossia, revisione di scopo, classificazione di Kotlow, protocollo Marchesan*

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Introduction

Ankyloglossia – or tongue-tie - is a congenital condition characterised by an abnormally short, thick and/or tight lingual frenulum which can restrict the protrusion and elevation of the tongue [1,2], thus resulting in several functional limitations [1,2]. The incidence of ankyloglossia in the newborn population varies from 4.8% to 10% and commonly presents as difficulty breastfeeding as an infant [3]. The literature typically differentiates ankyloglossia in neonates and infants from that occurring during childhood and adolescence [1]. Beyond its important consequences in the former group (e.g., breastfeeding, breathing complications, phenomenon of asphyxiation, and food rejection) [4], in the latter typical consequences are represented by speech impairment [5-7] and functional limitations produce changes in swallowing and chewing, preventing the correct coordination of muscles. This leads to a number of problems [8,9], such as oral respiration [10], forward tongue positioning with possible malocclusions [11,12] and consequences on breathing [13] and postural control [14].

Ankyloglossia is usually treated with surgical division of the frenulum with closure (frenuloplasty) or without (frenotomy/frenulectomy) [15]. However, there are no accepted guidelines on diagnostic and treatment management of ankyloglossia [15]. In this scenario, some experiences reported that after surgical tongue-tie release different degrees of improvements are found in terms of speech, social outcomes [7,16], dysphagia, sleep-disordered breathing and obstructive sleep apnoea syndrome (OSAS) [15,17]. With respect to the large amount of evidence depicting improvement in the effectiveness of frenulectomy on feeding and decreased pain perception from the mother [2,3], many debates are still present about the impact of ankyloglossia and – vice versa – frenulectomy in non-infants on speech, feeding and sleep-related breathing disorders, so that no significant conclusions have been made to date [7,16]. This was possibly due to different treatment procedures, relations with different functions related to tongue limitations and no consistent assessment methods to evaluate the extent and/or severity of ankyloglossia in different studies [18].

Thus, the aim of the present scoping review is to evaluate the impact – related to the main functional limitations of the ankyloglossia – of surgical and rehabilitative procedures devoted to release the tongue-tie in non-infants when implementing the most commonly used quantitative/qualitative structured tools for tongue assessment and frenulum length.

Scoping review methods

The scoping review and the meta-analysis were conducted following the guidelines of the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) extension for scoping reviews (PRISMA-ScR) [19].

Eligibility criteria

A literature search was performed to identify studies evaluating outcomes of tongue-tie release procedures and implementing assessment tools in non-infants in the last 10 years. The inclusion criteria (Tab. I) of the search strategy design are categorised according to the broad Population-Concept-Context (PCC) mnemonic recommended for scoping reviews [20,21]. The scoping review focuses on applications in clinical settings and on physiological research. Thus, studies on patients > 1 year of age were included. Studies were eligible only if outcomes of morphological/functional tongue-tie and frenulum length assessment tools, together with the results of objective tests investigating disability-related aspects of the ankyloglossia (i.e. speech, breathing and feeding), were implemented before and after the tongue-tie release procedures, while studies investigating these aspects mainly using self-assessed questionnaires or sub-items of the main assessment tools were excluded.

The search was restricted to observational and interventional studies involving humans and published in English in peer-reviewed journals. Abstracts, conference proceedings and reports, retrospective studies, expert opinions, letters to the Editor, commentaries, case reports/series and reviews were excluded.

Information sources, search strategy, and study selection

A systematic search was performed in PubMed, Scopus, and ISI Web of Science electronic databases to identify primary references from January 2011 to December 2021. The following search string was used: (“ankyloglossia” OR “tongue-tie” OR “tongue” OR “lingual frenulum” OR “frenuloplasty” OR “frenectomy” OR “frenotomy” OR “tongue-tie division” OR “tongue-tie release” OR “frenulum release” OR “myofunctional therapy” OR “tongue rehabilitation” OR “speech and language therapy”) AND (“assessment” OR “lingual mobility” OR “mouth opening” OR “articulation” OR “speech” OR “feeding” OR “swallowing” OR “breathing” OR “phonation” OR “frenulum length”). The database search was followed by a review of the citations from eligible studies. The studies were selected based on their title and their abstract using the online platform Rayyan [22]. The selected studies were read thoroughly to identify those suitable for inclusion in the scoping review.

Data extraction

Two reviewers (AM and MA) independently extracted the demographic and experimental data from the selected studies. When they disagreed, they reviewed the papers togeth-

Table I. Inclusion criteria for the scoping review summarised according to the Population-Concept-Context (PCC) mnemonic, recommended for scoping reviews [20,21].

Population	Healthy and sick children > 1 years old and adults Any sex
Concept	Outcome of tongue-tie division procedures, including both surgical and rehabilitative treatments, when evaluated by means of structured tongue and frenulum length assessment tools
Context	Clinical setting, with focus on oral science, speech therapy, otorhinolaryngology, physiological research Original peer-reviewed research articles (cross-sectional and interventional study design), published in English in the last ten years

er in order to reach joint conclusions. For each study, the following information were extracted and summarised: the characteristics of the sample of subjects under evaluation, outcomes of assessment tools of frenulum length, as well as of the tongue mobility and anatomy, and possible comparators of tongue functionality; type of tongue-tie division of the investigated study group; main results of the study with focus on tongue function changes after the tongue-tie division.

Results

The literature search retrieved 603 (Pubmed), 893 (Scopus), and 739 (ISI Web of Science) articles evaluating the impact of tongue-tie release on lingual and frenulum assessment tools. A total of 50 articles were retrieved for full-text review of which 7 were selected and included based on the inclusion criteria in the last 10 years (Fig. 1). The results are summarised narratively and presented in Table II, according to the presence or absence of other complementary tests administered to evaluate the effect of tongue-tie release. Collectively, two studies reported outcomes only using a lingual frenulum protocol validated by Marchesan and co-workers [14,23], while all other studies implemented the Kotlow classification [1,13,24-26], and two studies did not administer further tests to evaluate functionalities related to tongue mobility [1,23], by means of the Quick Tongue Tie Assessment Tool®. Two studies also reported the results of the distance between the interincisal margins with the tip of the tongue positioned at the level of the superior retroincisive papilla (MOTTIP) [13,25] and of the protrusion of the tongue [13,25], while one also showed outcomes of the maximum opening of the mouth (MAB) [13]. One study performed polysomnography [13] and one evaluated speech impairment [26]. Two studies performed the scalpel frenectomy in all subjects [1,23], and 1 in sub-groups of participants [26], while laser-assisted frenuloplasty was performed in all subjects in 4 studies [13,14,24,25] or in sub-groups of participants in one [26]. Five studies also delivered rehabilitation therapy [1,13,14,24,25]. Among these investigations, two studies by Marchesan et al. and Ferrés-Amat et al. – respectively involving 10 and 101 patients – only evaluated the impact of frenuloplasty [23] and

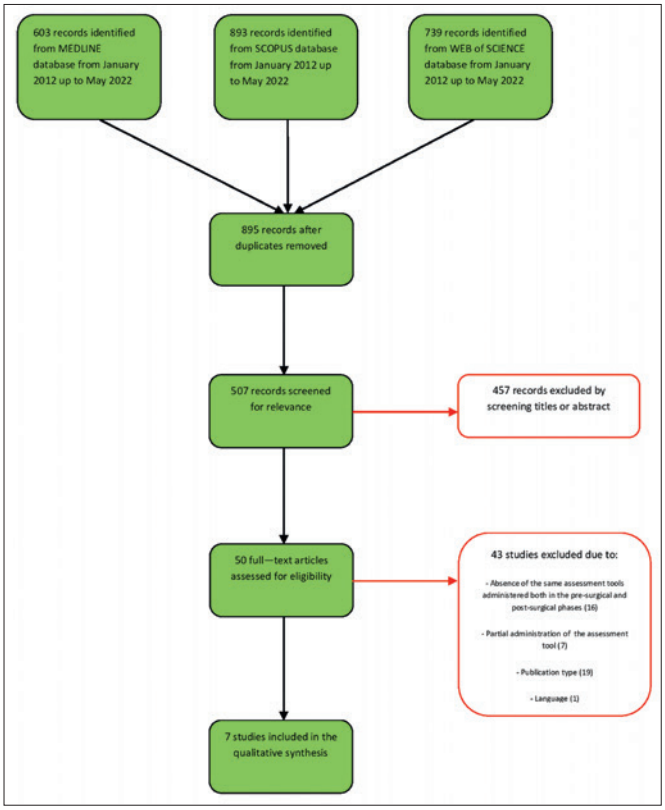

Figure 1. Selection process for studies included in the scoping review.

frenuloplasty associated with myofunctional training [1], respectively, on a specific lingual frenulum protocol validated by Marchesan et al. in 2010 [27] and on the Kotlow classification (Tab. II). Although the work by Marchesan et al. did not perform any statistical analysis on post-surgical tongue-tie release scores, the authors described improvement in tongue protrusion and tongue tip shape after 30 days [23]. Ferrés-Amat et al. found improvement (degree 1 or 2 by Kotlow classification) of tongue-tie release in 29% of patients at the first post-surgical evaluation performed at 72 hours and in 96% of patients after post-surgical rehabilitation sessions [1].
Table II also depicts the studies in which frenulum classifications were associated with other tests that are indirectly associated with tongue functionality. Tecco and co-workers

Table II. Studies included in qualitative analysis after the selection process.

Aim	Tongue/ frenulum assessment tools	Other tests	Intervention/ follow-up	Population/ pre-release tongue/frenulum assessment	Results	Reference
To describe changes in tongue mobility and orofacial functions after frenectomy	Lingual frenulum protocol by Marchesan in 2010	None	Frenectomy performed by an otolaryngologist *Follow-up: 30 days*	10 subjects (8 males, 2 females; mean age = 12.2 ± 12.4 years*)	30 days after the intervention, the best results were for tongue protrusion, while the worst were for tongue elevation. Surgery improved the altered shape of the tip of the tongue in 6 subjects, the mouth opening in 6 subjects, the speech in 4 subjects #	Marchesan et al. 2012 [23]
To assess changes in the sEMG potentials in the sub-mental, orbicularis oris, and masticatory muscles after lingual frenulectomy and OMT in subjects with ankyloglossia, characterised by Class I malocclusion.	Kotlow classification	sEMG potentials of the upper and lower fascicles of the orbicularis oris muscle, sub-mental areas, masseter, and anterior temporalis during rest position, clenching of teeth, kissing, swallowing, opening of mouth and protrusion of mandible	Laser-assisted frenectomy and orofacial myofunctional therapy in the treated group *Follow-up: T0 (before treatment), T1 (one month after the treatment) and T2 (6 months after treatment) in both treated and control group*	Treated group: 13 subjects with Class I malocclusion and ankyloglossia (4 females, 9 males; mean age = 7.0 ± 2.5 years) Control group: 11 subjects with normal occlusion and normal lingual frenulum (2 females; 9 males; mean age = 7 ± 0.8 years) **Kotlow classification Grade 3 (called "severe tongue-tie", about 4-8 mm) and Grade 4 (called "complete tongue-tie", about 0-4 mm)**	The Kotlow index passed from 3-4 to score 0 (called "normal": > 15 mm) or score 1 (called "acceptable; 12-15 mm). Treated group showed an increase in the sEMG potentials over the follow-up during clenching, swallowing (and kissing ($p < 0.05$). During the protrusion of the mandible, the masseter and anterior temporalis significantly decreased their sEMG activity, while the sub-mental area increased significantly ($p < 0.05$). During swallowing and protrusion of the mandible, the differences between the treated and the control group decreased during the follow-up ($p < 0.01$)	Tecco et al., 2015 [24]
To investigate if there is a correlation between a short lingual frenulum and the tonic-postural system before and after a laser frenectomy	Lingual frenulum Protocol by Marchesan IQ 2014	Postural evaluation by using the Spinometry® Formetric 4D	Laser frenectomy and orofacial myofunctional therapy *Follow-up: not reported*	24 patients (8 females, 16 males; mean age = 15.22 years, range = 10-26 years) **Mean lingual frenulum protocol score = 4.7 ± 1.1**	Interventions induced a significant lingual frenulum protocol score reduction ($p < 0.001$) and an improvement of shoulder opening (N.S.) during maximum mouth opening and tongue to palate with mouth open conditions	Saccomanno et al., 2020 [14]
To estimate the efficacy of lingual frenulectomy with diode-laser technology through a qualitative and quantitative evaluation	Kotlow classification MOTTIP MAB Protrusion (by Quick Tongue Tie Assessment Tool®)	Pain NRS	Laser frenectomy (Groups A-D), speech therapy (Groups A-B), sterile iodoformic gauze affixed with suture in Groups A-C. *Follow-up at T0, T1, T2, T3, and T4, with 7 days of difference amongst them. No intervention in Group E*	125 patients (59 females, 66 males; mean age = 8.37 ± 2.31 years) randomised in 5 sub-groups (A-E) **Kotlow with a length ≤ 16 mm and lack of tongue functionality**	The Kotlow score was significantly higher in T3 and T4 in Group D when compared to others ($p < 0.001$). From T1 to T4 MOTTIP had a trend of growth better in Group D than in other groups ($p < 0.001$). MAB had a lower score in Group C than in other groups from T1 to T4 ($p < 0.001$). The value of Protrusion was greater in Group E than in the other groups in T1 and T2 ($p < 0.001$)	Sfasciotti et al., 2020 [25]

continues ▶

Table II. Studies included in qualitative analysis after the selection process (*follows).*

Aim	Tongue/ frenulum assessment tools	Other tests	Intervention/ follow-up	Population/ pre-release tongue/frenulum assessment	Results	Reference
To present the protocol of action of the Fundació Hospital de Nens de Barcelona (FHNB) for the treatment of ankyloglossia in childhood and adolescence (pre-surgical, surgical technique, post-surgical)	Kotlow classification	None	Frenectomy and rhomboidal plasty Myofunctional training: begins one week before the surgical intervention so that the patients learn the exercises without pain and 24 hours after surgery *Follow-up: 72 hours, 15 days, 45 days*	101 patients (38 females, 63 males), ranking in age from 4 to 13 years old **Kotlow classification with Degree 3 (N = 11), Degree 4 (N = 74), Degree 5 (N = 16)**	In all patients the tongue is released after the lingual frenectomy and plasty. Post-surgical check-ups: degree of ankyloglossia has been improved, considering correction (degrees 1 or 2) in 29 (28%) of the patients (95% CI: 20%, 38%) Check-up after all the postsurgical orofacial rehabilitation sessions: correction of 97 (96%) of the participants (95% CI: 90%, 98%) Complication in 7 (6%) of the participants (95% CI: 2%, 13 %)	Ferrés-Amat et al. 2016 [1]
To evaluate the efficacy of lingual frenectomy to improve the length of frenulum and severity of OSAS in pediatric patients.	Kotlow classification Ruffoli classification (only for pre-treatment assessment) MAB MOTTIP Protrusion (by means of Quick Tongue Tie Assessment Tool®)	Polysomnography NRS	Laser frenectomy and Myofunctional exercises to perform at home (SG) Myofunctional and speech therapy (CG) *Follow-up: T0, T1 after 3 months*	32 female and male pediatric patients (4-13 years old) SG: 16 patients (7 females, 9 males); 8 (50%) had severe OSAS and 8 had moderate OSAS (50%) CG: 16 patients (7 females, 9 males); 3 had severe OSAS (18.8%) and 13 had moderate OSAS (81.2%) **Kotlow classification with class III-IV-V by Kotlow; Grades 2 and 3 by Ruffoli**	SG: increased in the scores: Kotlow ($p < 0.001$), MAB ($p < 0.01$), MOTTIP ($p < 0.001$) and Protrusion ($p < 0.001$). SG: 93.8% mild OSAS and 6.2% moderate OSAS at T1 CG: significant increase in all of the scores: Kotlow ($p < 0.001$), MAB ($p < 0.01$), MOTTIP ($p < 0.01$) and Protrusion ($p < 0.01$). CG: 18.75% mild OSAS, 62.5% moderate OSAS and 18.75% severe OSAS at T1. No between-groups significant differences in the Kotlow and Quick Tongue Tie Assessment Tool® quantitative variables	Fioravanti et al., 2021 [13]
To compare surgical outcomes of simple frenotomy and the 4-flap Z-frenuloplasty according to the articulation test values and tongue-tie classification in patients with ankyloglossia with articulation difficulty	Kotlow classification	PRES and REVT to assess the speech impairment of children. U-TAP to evaluate the accuracy of consonant pronunciation	Simple frenotomy or the 4-flap Z-frenuloplasty performed by the two surgeons *Follow-up: 3 months after surgery*	Out of 37 patients, 19 underwent the 4-flap Z-frenuloplasty and 18 (age 3.52 ± 0.69 years, 13 males, 6 females), the simple frenotomy (age 4.66 ± 2.05 years, 10 males, 8 females) **Kotlow classification: greater than Classification 1**	Before surgery, abnormal tongue-tie classification was observed, but after surgery, all the cases except 1 case in frenotomy and 3 cases in frenuloplasty groups were classified as "normal". When compared before and after the surgery, improved tongue-tie classification have been confirmed. No significant differences between groups ($p > 0.99$). Changes in the articulation test (consonants) were statistically significant in both groups ($p < 0.001$). Most patients improved speech articulation. No significant difference in outcomes between the two surgical groups ($p = 0.28$)	Kim et al., 2020 [26]

*NRS: numerical rating scale; MAB: maximum opening of the mouth; MOTTIP: distance between the interincisal margins with the tip of the tongue positioned at the level of the superior retroincisive papilla; PRES: Preschool Receptive-Expressive Language Scale; REVT: Receptive and Expressive Vocabulary Test; U-TAP: Urimal Test of Articulation and Phonation; SG: Study Group; CG: Control Group; sEMG: surface electromyography; protrusion: distance between the tip of the protruded tongue and the incisal margin; OMT: orofacial myofunctional therapy; N.S.: not significant; *Calculated by the authors; no statistical analysis was given in the text. In italic and in bold type are respectively given the follow-up stages and the tongue/ frenulum assessment classification before the release.*

not only found an improvement in Kotlow classification degrees (from 3-4 to 0-1 after intervention) in 13 patients treated with frenuloplasty and orofacial myofunctional therapy, but also found a significant ($p < 0.05$) increase in surface electromyography (sEMG) potentials over the follow-up during clenching and swallowing and a decrease in the sEMG activity in masseter and anterior temporalis during protrusion of the mandible [24]. Sfasciotti and colleagues randomly divided 125 patients into 5 groups according to the delivery of frenuloplasty, speech therapy and positioning of a sterile iodoformic gauze affixed with suture and found that the Kotlow and MOTTIP scores significantly improved during follow-up only in patients undergoing frenuloplasty ($p < 0.001$) [25]. Patients who were delivered the gauze positioning and did not receive speech therapy had lower MAB post-surgical scores compared to the others ($p < 0.001$) and patients who did not receive any treatments had significant ($p < 0.001$) greater values of protrusion within 2 weeks of follow-up. Fioravanti et al. implemented the same assessment tools for pre- and post-treatment stages with polysomnography in 32 patients randomly undergoing frenuloplasty and home-delivered myofunctional exercises ($n = 16$) or myofunctional and speech therapy ($n = 16$) [13]. Both groups significantly improved during follow-up along all the tongue-related scales, without significant between-group differences. The former group had a higher number of mild OSAS cases post-intervention with respect to the latter group in which a higher numbers of moderate and severe OSAS cases were found. Kim et al., in 2020, compared the surgical outcomes of simple frenotomy and the 4-flap Z-frenuloplasty according to the tongue-tie classification and speech impairment test values [26]. They found significant improvement in the within-subjects comparisons of frenulum scale and speech impairment; however, no surgical procedure demonstrated an advantage in terms of improving outcomes.

Finally, Saccomanno et al. associated the lingual frenulum protocol by Marchesan with postural evaluation using spinometry before and after laser frenectomy and orofacial myofunctional therapy in 24 patients [14]. They found that the intervention induced a significant lingual frenulum protocol score reduction ($p < 0.001$), but not significant improvement of shoulder opening during maximum mouth opening and tongue to palate with mouth open conditions.

Discussion

Ankyloglossia, also known as tongue-tie, is a congenital oral anomaly characterised by an abnormally short lingual frenulum. The lingual frenulum, according to the International Affiliation of TongueTie Professionals, is a midline remnant of the tissue between the ventral surface of the tongue and the floor of the mouth [26,28]. When it interferes with normal function, the condition is called "symptomatic ankyloglossia" [26]. The anatomy of the lingual frenulum is poorly described in literature [29,30]. Although a recent study illustrated how it develops from concurrent muscular and mucous components [31], there are no publications providing relevant additional details on the physiological morphology. Thus, it is hard to understand how pathological variables can determine functional limitations of the tongue in children, young patients and adults [32].

Due to the variety of symptoms related to different functional limitations, a multidisciplinary approach, involving otolaryngologists, pediatricians, speech pathologists and dental specialists, is advocated. Although its existence has been acknowledged for centuries [33], the clinical implications of diagnosis and management remain controversial [34]. Given the existence of several classification systems and various diagnostic criteria [35] (none of which are used as a clear reference), indications for treatment remain unclear. Indications for general surgical treatment include feeding difficulty and speech impairment [36] and speech therapy by speech pathologists as well as surgical intervention are the most commonly reported treatment modalities [26].

All these aspects are further complicated by the presence of all those conventional problems caused by limited movement of the tongue which have been frequently pinpointed for both the indication and/or follow-up of interventional programmes of tongue tie division, often in absence of an anatomical landmark of the frenulum and tongue position [37-39]. In this vision, the strict inclusion criteria applied in the present scoping review highlighted that – among all the classifications (mostly qualitative) used in the literature to evaluate the indication and resolution rate of tongue-tie division [6,16,40,41] – the following quantitative classifications were most widely used for both inclusion and follow-up strategies: Kotlow classification (normal ≥ 16 mm), Marchesan classification, the MOTTIP (normal ≥ 23 mm), MAB (normal MY ≥ 35 mm) and the lingual protrusion. In different studies, additional classifications were used together [13,25] (Tab. II).

Changes in Kotlow classification

Collectively, studies implementing the Kotlow classification – which stages the ankyloglossia as more severe in relation to the distance of the tip of the tongue–lingual insertion of the frenulum [42] – demonstrated that frenectomy associated with rehabilitation induced a decrease in the severity of this classification, passing from stage 3-4 to 0 or 1 with respect to a matched group of patients not undergoing any type of treatment [24]. Furthermore, this reduction –

which appeared after 21 days of follow-up – was mostly similar to patients who also underwent rehabilitation in association with surgery, but better than those who received a sterile iodoformic gauze at the end of the intervention [25]. Interestingly, after 28 days of follow-up no differences were found when comparing patients only undergoing surgery or rehabilitation. This result was also confirmed by another study by Fioravanti and colleagues demonstrating that both types of intervention induced a significant reduction in the Kotlow classification in two groups of OSAS patients and that no between-group differences were found during follow-up [13]. When comparing the effect of simple frenotomy or the 4-flap Z-frenuloplasty on two groups of patients with tongue-tie Kotlow classification > 1 and speech impairment, Kim and colleagues demonstrated that both techniques significantly improved frenulum length and, consequently, speech ability in both groups of participants with no between-groups effect [26]. Finally, Kotlow classification was found to be significantly improved in a cohort study by Ferrés-Amat et al. in which a large sample of young participants – in the absence of a control group – underwent frenectomy with rhomboidal plasty associated with myofunctional training [1].

Changes in Marchesan protocol

The Marchesan protocol – which associates qualitative anatomical and functional parameters of the tongue with a quantitative ratio of the measurements of the distance from superior to inferior incisive in with open mouth wide with or without the tongue tip touching the incise papilla [27,43] – was found to be improved in a study by the same author involving 10 participants undergoing frenectomy; it was demonstrated that the surgical procedure also induced benefits in terms of protrusion, shape of the tongue tip, mouth opening and speech [23]. Using the same multiparametric diagnostic protocol, Saccomanno and co-workers demonstrated – in the absence of a control group – that laser frenectomy and rehabilitation treatment led to significant reduction in the lingual frenulum score and changes (even if not significant) in postural parameters [14].

Changes in complementary assessment tools

Finally, three tests were used by the group of Sfasciotti et al. [25] and Fioravanti et al. [13] with the aim to complete the Kotlow classification with the additional information included in the Marchesan protocol: i.e. MOTTIP, MAB and lingual protrusion. These tests, previously used in other studies [44,45], were demonstrated to improve, especially in the first weeks of follow-up, in patients undergoing surgical tongue-tie without sterile iodoformic gauze positioning at the end of the intervention with or without speech therapy with respect to all other groups [25] and to be equally benefitted by surgical/rehabilitation or only rehabilitation [13].

Consequences on other dysfunctions

Given the notions speculating limitations in tongue movements are involved in different impairments of speech [5-7], swallowing and chewing, alterations of the correct coordination of muscles [8,9], oral respiration [10], forward tongue positioning with possible malocclusions [11,12], OSAS [13] and postural consequences [14], some authors have evaluated some of these descending consequences with regards to the tongue-tie division. Underpinnings of these alterations are thought to be related to adaptation of muscles when tongue function is restricted by a tongue-tie [24]. Since the tongue cannot function as it is supposed to, other muscles have to help. This leads to a whole cascade of compensations and adaptations throughout the stomatognathic apparatus, involving the reciprocal relationships between intrinsic and extrinsic tongue muscles [24]. The former behaves as a scaffolding by which the intrinsic muscles can be moved around in the oral cavity, while the latter are continuously modifying their dimension and contour [24,46]. Conversely, when a tongue-tie is surgically treated, the patient has no muscle memory of how to use his/her tongue without the restriction there; since the brain takes time to rewire itself, rehabilitation can aid in this functional re-education [24]. In this regard, a rehabilitation protocol – together with frenulectomy – was held by Tecco et al. to be pivotal in re-patterning the muscles, ensuring full range of motion of the tongue, and making sure the tissues do not re-attach after surgery [47]. Over 6-months of follow-up, this is associated with significant changes in the sEMG potentials of the masticatory and sub-mental muscles in some tasks, particularly during maximal voluntary clenching, kissing, and swallowing [24]. In line, also an improvement – even if not significant – in term of shoulder opening during mouth opening and tongue to palate with mouth open when studied with spinometry was demonstrated by Saccomanno et al. [14], possibly reflecting restoration of the above-mentioned neuromuscular cascades induced by the tongue-tie along the tonic-postural system [14,48]. According to the literature regarding involvement of tongue-tie in OSAS development [17,49], in the study by Fioravanti and co-workers patients undergoing laser frenectomy and myofunctional exercises had more improvement compared to those undergoing only myofunctional exercises when staging their OSAS after 3 months of follow-up, although no between-group differences in tongue and frenulum assessment were found [13]. This aspect is relevant since it highlights the importance of anatomical and functional restoration when tongue-tie is involved in the development of OSAS. In-

deed, in these cases the altered lingual posture determines a different direction of growth of the jaw bones which can lead to orofacial dysmorphia with a reduction in the influx of incoming air with consequent respiratory problems [5,50]. Therefore, it decreases the size of the upper airway support and progressively increases the risk of upper airway collapse during sleep inducing OSAS [5,49], for which the study by Fioravanti et al. demonstrated that successful treatment should involve anatomical release supported by a myofunctional therapy, beyond the impact on tongue and frenulum assessment tools [13]. Finally, following evidence showing a benefit of tongue-tie release on speech impairment [51], Kim et al. demonstrated that simple frenotomy and the 4-flap Z-frenuloplasty equally impacted speech impairment tests [26]. In parallel with a similar effect on the post-operative Kotlow classification improvement, the study reinforced the idea that limited tongue tip mobility is an underpinning factor contributing to speech impairment [51] and that its simple anatomical release may be useful in reversing the alteration.

Final remarks and future perspectives

When taking into account the "lens" through which the present scoping review selected the original studies, it has to be noted that some relevant studies were excluded due to the lack of a rigorous evaluation of the tongue and the frenulum before and after the tongue-tie release (see for example [37-39]). This aspect assumes relevance i) when in other studies a certain degree of dyscrasia was demonstrated between tongue/frenulum assessment tools and functional tests (see for example the studies by Saccomanno et al. [14] and Kim et al. [26]) and ii) when – on the other hand – the recognised assessment tools have to be considered the reference landmark for follow-up, also of disorders related to the tongue-tie (see for example Tecco et al. [24]). Furthermore, the studies included – besides rigorously following the tongue-tie release with the assessment tools –suffer in some cases from limitations in terms of small sample size [23] and/or lack of a control group [1,14,23], size differences between the study and control groups (see for example [13,26]) and different timing of the first follow-up (from 24 hour in Ferres-Amat et al. [1] to 3 months in Kim et al. [26] and Fioravanti et al. [13]). All these aspects – together with the different treatment options proposed – make interpretation of the impact of tongue-tie division on tongue and frenulum assessment equivocal and complex. However, if on one hand the present work showed that the majority of treatment options have been found to significantly improve – regardless of the assessment tools and tongue-tie release procedures implemented – the anatomical limitation of the tongue with clear benefits on descending functionality, on the other it highlighted the need of rigorous implementation in research and clinical practice of validated tongue assessment tools both before and after frenulum release. This would have the purpose of better correlating improvements with benefits on functional limitations, standardising results across different research groups and facilitating the transfer of clinical knowledge between those involved in the diagnostic process of tongue-tie and its release.

For future perspectives, the results of the present scoping review stress the importance of implementation of oral and extra-oral functional tests which could clinically integrate anatomical assessment of the tongue and frenulum and the utility of a multidisciplinary approach that can merge surgical and rehabilitative approaches. Following these directions and the lessons learned by the bias of the selected study protocols, more clear and suitable assessment tools devoted to thorough assessment of the quantitative and qualitative consequences of tongue-tie and its release on both oral and extra-oral dysfunctions are needed.

Conflict of interest statement

The authors declare no conflict of interest.

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Author contributions

AM, MAr, MA: conceptualization; MAr, AM, ADB, MA: methodology; MAr, MA, AM, ADB, FG: formal analysis; AM, MAr, MA, DJ: data curation; AM, MAr, BM, IM, ADB, MA, BM: writing-original draft preparation; MAr, AM, ADB, MA, AC, IM: writing-review and editing; MA, DJ, AM, AC, BM, FG: supervision. All Authors have read and agreed to the published version of the manuscript.

Ethical consideration

N/A.

Informed consent

N/A.

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