Research Article

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Roberta Lopes de Castro Martinelli (*Hospital Santa Therezinha, Brazil*)

Irene Queiroz Marchesan (*CEFAC, Brasil*)

Jose Roberto Lauris (*Universidade de Sao Paulo*)

Heitor Marques Honorio (*Universidade de Sao Paulo*)

Reinaldo Jordão Gusmão (*Universidade de Campinas*)

Giedre Berretin-Felix (*Universidade de Sao Paulo*)

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VALIDATION OF THE LINGUAL FRENULUM PROTOCOL FOR INFANTS

ROBERTA LOPES DE CASTRO MARTINELLI, PH.D., SLP

IRENE QUEIROZ MARCHESAN, PH.D., SLP

JOSÉ ROBERTO LAURIS, PH.D., HEITOR MARQUES HONÓRIO, PH.D.,

REINALDO JORDÃO GUSMÃO, PH.D., GIÉDRE BERRETIN-FELIX, PH.D.

ABSTRACT

Introduction: In Brazil, the law 13.002/2014 states that all newborns in the country must undergo lingual frenulum assessment before hospital discharge. **Objective:** To validate the Lingual Frenulum Protocol for Infants. **Material and Methods:** The study used data from 100 infants assessed at 30 days of life at Santa Therezinha Hospital, in Brotas, SP, Brazil. The LFPI was administered to the 30-day-old infants and all assessments were registered in patient records and videotaped. Live exams, assessments and re-assessments using images of the infants were performed by two examiners experienced in Orofacial Myofunctional Disorders. The study included healthy full-term infants of both genders, who were being breastfed exclusively. Prematurity, perinatal complications, craniofacial anomalies, neurological disorders, and visible genetics syndromes were the exclusion criteria. The validation process included the analysis of content validity, criterion validity, construct validity, reliability, sensitivity, specificity, positive predictive value, and negative predictive value. **Results:** Concerning content validity, the percentage of agreement was 100%. The criterion validity was measured by comparing the common items of the LFPI and BTAT. The agreement percentage for the analyzed items was 100%. The construct validity was analyzed by comparing the partial and total scores from the assessment of infants with and without lingual frenulum alteration at 30 and 75 days of life. Results demonstrated similarity of scores between the control group and the group that underwent frenotomy. The results from the inter-rater and intra-rater agreement analysis indicated perfect agreement. The values of sensitivity, specificity, positive predictive value, and negative predictive value were 100%. **Conclusion:** The study concluded that the LFPI was a valid and reliable assessment tool, that ensures accuracy when diagnosing lingual frenulum alterations within the investigated parameters.

KEYWORDS: ankyloglossia, breastfeeding, clinical protocols, validation studies, surgical procedures

INTRODUCTION

Although the human tongue is one of the most important structures of the body, there is poor understanding of its complex muscle anatomy. As a result, the assessment and treatment of tongue disorders lags behind that of other structures of the body. Recent studies on the anatomy and histology of the tongue have demonstrated that the lingual frenulum does not stretch and does not undergo spontaneous rupture (Martinelli, Marchesan, Gusmão,

Rodrigues, & Berretin-Felix, 2014). Another study found that the lingual frenulum remains unchanged over time (Martinelli, Marchesan, & Berretin-Felix, 2014). Moreover, recent studies using ultrasound have demonstrated the importance of the tongue, in terms of orofacial function, as it plays an essential role for milk extraction during breastfeeding (Sakalidis, Williams, Garbin, Hepwoth, Hartmann, Paech & Geddes, 2013; Elad, Kozlovsky, Blum, Laine,

Po, Botzer, Dollberg, Zelicovich, & Bem-Sira, 2014; Burton, Deng, McDonald, & Fewtrell, 2013; McClellan, Kent, Hepworth, Hartmann, & Geddes, 2015).

As the human tongue performs unique movements, which are very important for the performance of all orofacial functions, any movement restriction may interfere with speech, chewing, swallowing, and sucking. Furthermore, the anatomic variations of the lingual frenulum may interfere with tongue movements that may justify the importance of using validated protocols for a lingual frenulum assessment.

Protocols are important for effective assessment, diagnosis and treatment. The assessment protocols provide parameters for studying the case and defining the treatment plan. Protocols also provide standardized assessment that benefits comparative findings and assists epidemiological studies (Genaro, Berretin-Felix, Rehder, & Marchesan, 2009; Graziani, Fukushima, & Genaro, 2015). The Lingual Frenulum Protocol for Infants – LFPI (Martinelli, Marchesan, & Berretin-Felix, 2012; Martinelli, Marchesan, Rodrigues, & Berretin-Felix, 2012; Martinelli, Marchesan, & Berretin-Felix, 2013) assists with the diagnosis of movement restriction of the tongue due to frenum alterations. The protocol is scored and consists of a structural and functional evaluation, a nutritive and non-nutritive suction evaluation, and a clinical history. In Brazil, the law 13.002 passed in 2014 (Brazil, 2014) stating that all newborns in the country must undergo a lingual frenulum assessment before hospital discharge. The anatomical-functional evaluation, popularly known as the “Neonatal Screening Test,” is the first part of the LFPI administered to all newborns (Martinelli, Marchesan, Gusmão, & Berretin-Felix, 2014). Scores indicate whether there is any lingual frenulum alteration or not. In case of doubt, re-assessment is recommended at 30 days of life. The re-assessment consists of the administration of the full protocol – anatomical-functional evaluation, nutritive and non-nutritive suction evaluation, and clinical history (Martinelli, Marchesan, & Berretin-Felix, 2012; Martinelli, Marchesan, Rodrigues, & Berretin-Felix, 2012; Martinelli, Marchesan, & Berretin-

Felix, 2013; Martinelli, Marchesan, Gusmão, & Berretin-Felix, 2014). The aim of this study was to validate the full Lingual Frenulum Protocol for Infants (LFPI).

MATERIAL AND METHODS

The current study used data from infants assessed at 30 days of life at Santa Therezinha Hospital, in Brotas, SP, Brazil. The LFPI was administered to 30-day-old infants and all assessments were registered in patient records and videotaped. The study was approved by the Ethics Committee of the Faculty of Odontology of Bauru – University of Sao Paulo under the number CAAE 38143814.8.0000.5417.

Based upon the analysis of the total score of the protocol and the minimum difference between groups, at 5% level of significance and 80% strength of study design, a standard deviation of 6 was considered to calculate the sample size. 64 infants were required for the t-Test, and an additional 25% were needed for the Mann-Whitney test. Sample loss was also considered; thus, 100 infants were found to be the optimal sample size to validate the LFPI. Of the 100 infants, 46 were females and 54 were males.

Each infant was assessed at 30 days of life by Examiner 1 (E1), a Speech Language Pathologist, with breastfeeding and lingual frenulum assessment expertise. Sharp images and data from each assessment were collected. Of the 100 infants, 21 (16 males and 5 females) were referred for frenotomy. Of the 21 infants, 19 (14 males and 5 females) underwent the frenotomy, which was performed at 45 days of life by an Otorhinolaryngologist. Of the 79 infants without lingual frenulum alteration, 19 infants (14 males and 5 females) were randomly selected to be part of the control group. Thirty days after undergoing frenotomy, the 19 infants and the control group were re-assessed by E1 using the LFPI. All assessments and re-assessments were videotaped and registered in the patient's records.

Subsequently, another experienced Speech Language Pathologist, Examiner 2 (E2), analyzed the images and data provided by E1. The calibration between E1 and E2 was performed using a live exam, analysis of the data collected during the LFPI administration, and clinical case discussion.

The study included healthy full-term infants of both genders, who were exclusively breastfed. Prematurity, perinatal complications, craniofacial anomalies, neurological disorders, and visible genetics syndromes were the exclusion criteria.

The Lingual Frenulum Protocol for Infants (LFPI)

The LFPI consists of a clinical history and an anatomical functional evaluation, (nutritive and non-nutritive) (APPENDIX 1). The scores of the LFPI correspond to a 0-3 scale: 0 is indicative of free tongue movement; 1 is indicative of the presence of any alteration, not necessarily caused by the lingual frenulum; 2 is indicative of restricted tongue movement; and 3 is indicative of the presence of a lingual frenulum alteration that clearly restricts tongue movement.

The sum of the scores from the anatomical-functional evaluation and the nutritive and non-nutritive sucking evaluation, are sufficient for the final diagnosis. Although the clinical history is important, it is not essential for an effective diagnosis. The anatomical-functional assessment was designed to provide an accurate diagnosis.

The sum of the scores from the anatomical-functional evaluation, nutritive and non-nutritive sucking evaluation, equal to or greater than 9, was indicative of the interference of the lingual frenulum with the tongue movements. Data from the assessment and re-assessment were considered for the process of validation of the LFPI. The significance level adopted was 5% ($p < .05$).

Content Validity

The content validity was determined by the judgment of three experts, who individually had over 5 years of lingual frenulum assessment

and research experience. The 3 experts assessed the content of the full protocol (clinical history, anatomical-functional evaluation, nutritive and non-nutritive sucking evaluation, questionnaire and images) and rated the clarity of each item of the LFPI. The examiners received verbal and written guidance as to the completion of the protocol. The examiners rated each item on a scale of 1-4: 1= no clarity; 2= unclear; 3= clear; and 4= very clear, in order to perform content validation, by applying the equation of the Content Validity Index – CVI (Alexandre & Coluci, 2011). If the examiners marked options 1 or 2, the items had to be reformulated (Wynd, Schmidt, & Schaefer, 2003).

Criterion Validity

The criterion validity was measured by comparing the LFPI and the Bristol Tongue Assessment Tool, BTAT (Ingram, Johnson, Copeland, Churchill, Taylor, & Emond, 2015). Although the BTAT has not been fully validated, it has fulfilled some of the steps required for complete validation. Therefore, the criterion validity was measured by comparing the common items of the LFPI and the BTAT: a) tongue tip appearance during elevation; b) lingual frenulum attachment to the floor of the mouth and c) tongue position during crying. The Spearman's correlation coefficient was used to determine the criterion validity considering the LFPI and the BTAT.

Construct Validity

The construct validity was performed by comparing the scores of the LFPI of the control group to the infants who underwent surgery. The assessments were independently performed by the same two Speech Language Pathologists, experienced in Orofacial Myofunctional Disorders (E1 and E2). E1 performed the assessment of each infant at 30 days of life. Each assessment performed by E1 was photographed and video recorded. Subsequently, E2 analyzed the films and photos and filled the protocol. The infants who had to have frenulum alteration were referred for surgery. Each infant underwent surgery at 45 days of life. At 75 days of life the control group and the infants who underwent to frenotomy were re-assessed by E1 using the

LFPI. The re-assessments were photographed and video recorded. All images of the re-assessment performed by E1 were analyzed by E2. Post-operative data of the infants who underwent frenotomy and the data of the infants from the control group were compared, as demonstrated in Table 1. The Wilcoxon Test and the Mann-Whitney were used for the analysis of the construct validity.

Reliability

Inter-rater agreement analysis was used to indicate reliability. E1 administered the protocol

to 100 infants across 30 days. E2 performed the assessment using the photos and videos collected by E1 during the administration of the protocol. The results of the assessments were compared. Intra-rater agreement analysis was performed by comparing data from the assessment and re-assessment of 20% of the sample conducted by E2, using the same protocol. 15 to 30 days after the first assessment, the re-assessment of the data was performed in order to avoid the interference of memory.

Table 1 - Score comparison of the clinical history, anatomical-functional evaluation, nutritive (NS) and non-nutritive sucking (NNS) evaluation and total scores from the clinical examination and full protocol considering assessment of infants with and without lingual frenulum alteration performed by E2 at 30 and 75 days (Mann-Whitney test).

Subjects = 38	Assessment at 30 days						
	Without lingual frenulum alteration			With lingual frenulum alteration			p-value
	25 %	Med	75 %	25 %	Med	75 %	
Clinical history	0	1	2	5	5	6	.001*
Anatomical-Functional	0	1	2	6	8	9	.001*
NNS e NS	0	0	0	3	4	4	.001*
Clinical examination	0	1	2	10	11	13	.001*
Full Protocol	1	2	4	16	17	19	.001*

*significant difference $p < .005$.

Subjects = 38	Assessment at 75 days						
	Without lingual frenulum alteration			With lingual frenulum alteration			p-value
	25 %	Med	75 %	25 %	Med	75 %	
Clinical history	0	0	1	0	0	1	.402
Anatomical-Functional	0	1	2	0	2	2	.418
NNS e NS	0	0	0	0	0	0	1.000
Clinical examination	0	1	2	0	2	2	.452
Full Protocol	0	2	3	0	2	3	.624

*significant difference $p < .005$.

The Intraclass Correlation Coefficient (ICC) was used for reproducibility analysis, quantitative aspects. The scale suggested by Menz, Latt, Tiedemann, Mun San Kwan and Lord, (2004), was used to interpret the ICC values.

The error of the method calculation was used for inter/intra-rater agreement analysis. The casual error occurs due to the difficulty or in the accuracy of either identifying or defining certain points. Casual errors of great magnitude imply difficulty in reproducing the measurements. Systematic error occurs when a given measurement is continuously under or super-estimated. Systematic errors of great magnitude distort the results in a certain direction. The casual and systematic errors were considered for the analysis. The values suggested by Houston (1983) were used to interpret the data. As for the casual errors, the smaller the measurement errors, the greater the reliability of the data. Concerning the systematic errors, the greater the percentage ($p > .05$), the smaller the difference between measurements.

Sensitivity, Specificity, Positive Predictive Value and Negative Predictive Value

The analysis was performed using the dynamic images from the assessment, comparing the correspondent items in both protocols – LFPI and BTAT. Tongue tip appearance, attachment to the alveolar ridge on the bottom of the mouth and the elevation of the tongue were compared and the scores for the items were analyzed. According to the BTAT, score 8 is indicative of the absence of alteration, while zero is indicative of a severe reduction of tongue function. When the sum of the items of the anatomical-functional evaluation of the LFPI is zero, it is indicative of a total absence of alteration, and 12 is indicative of a severe alteration.

The sensitivity (S) was calculated by considering the number of true positives: the number of infants who were diagnosed with altered lingual frenulum according to both protocols, divided by the total number of infants diagnosed using the BTAT. Specificity (SPC) was calculated by considering the number of true negatives: the number of infants who did

not have lingual frenulum alterations according to both protocols, divided by the number of infants who did not have lingual frenulum alteration according to the BTAT. The positive predictive value (PPV) was calculated by dividing the number of true positives by the total number of infants who had lingual frenulum alterations according to the LFPI. The negative predictive value (NPV) was calculated by dividing the number of true negatives by the total number of infants who did not have lingual frenulum alterations according to the LFPI.

RESULTS

Content Validity

The examiners analyzed the content of the LFPI and rated the clarity of each item of the protocol. 79% of the items were rated “very clear” and 21% “clear”, thus demonstrating 100% agreement.

Criterion Validity

The criterion validity was measured by comparing the common items of the LFPI and the BTAT. Using the images provided by E1 from the administration of the LFPI and the BTAT, E2 assessed 20% of the 100 infants in the study. The agreement and disagreement percentages between protocols were calculated. The agreement percentage for the analyzed items was 100%. The analysis of the concurrent validity between protocols demonstrated negative correlation, since the protocols have inverse scales. The Spearman's correlation coefficient value was -0.997 demonstrating strong correlation among the assessed items of 20% of the sample ($p < .001$).

Construct Validity

The construct validity was analyzed by comparing the partial scores from the assessment of infants with and without lingual frenulum alteration at 30 and 75 days of life as well as by comparing the total scores from the assessment of the infants with and without lingual frenulum alteration at 30 and 75 days of life. Post-operative data of the infants who underwent frenotomy and the data of the infants from the control group were compared.

Results from the assessments demonstrated similar scores between the control group and the group that underwent frenotomy. Moreover, similar total scores concerning the clinical examination and the full protocol were observed (Table 1).

Inter-rater and intra-rater agreement analyzes

Of the 100 infants assessed, E1 and E2 found that 79 had normal lingual frenulum and 21 had lingual frenulum alteration (inter-rate agreement). The ICC was found to be 0.99 for the anatomical-functional evaluation and 0.98 for the nutritive and non-nutritive sucking. The ICC was found to be 0.99 for the clinical examination and the complete protocol. The consistency of the results demonstrated that there were conditions for the establishment of excellent repeatability: the same measuring instrument (LFPI), the same observers, under the same conditions, repetition over a short period of time, and the same objectives. The analysis of the error of the method had values ($p > 0.05$), thus demonstrating the absence of disagreement between the results reported by A2 in two distinct moments. Therefore, the results were indicative of very good agreement.

Analysis of sensitivity, specificity, positive predictive value and negative predictive value

The sum of the scores from the BTAT and the sum of the scores from the LFPI, were considered to calculate the values of S, SPC, PPV, and NPV. 75% was the cutoff point considered for the LFPI while 25% was the cutoff point for the BTAT. The values of S, SPC, PPV, and NPV were 100%. Lingual frenulum alteration occurrence was 21%, (16% males and 5% females).

DISCUSSION

The current study was motivated by the fact that the literature does not report the existence of fully validated tools for the assessment of infants' lingual frenulum. Although there are assessment protocols for infants cited in the literature, BTAT (Ingram, Johnson, Copeland,

Churchill, Taylor, & Emond, 2015) and Hazelbaker (Amir, James, & Donath, 2006), the protocols are partially validated. All requirements for the validation of the LFPI were met.

For content validity, the items regarding clarity were observed (Alexandre & Coluci, 2011). The examiners rated the clarity of each item of the LFPI. The CVI of the items of the LFPI demonstrated 100% agreement. The 100% agreement is above the literature requirements for a validated assessment tool (Alexandre & Coluci, 2011; Wynd, Schmidt, & Schaefer, 2003; Hermida & Araújo, 2006; Siqueira, 2008).

Concerning the criterion validity, the comparison of the results from the administration of the LFPI and the BTAT demonstrated that the percentage of agreement between protocols for the analyzed items was 100%, showing strong correlation. The literature reports that the comparison of similar items from distinct assessment tools tend to show positive results (Folha, Valera, & Felício, 2015).

Douglass (1993) discusses the importance of comparing an assessment tool to be validated with a gold standard validated tool. Although BTAT has not been fully validated, it has fulfilled some of the steps required for complete validation. Therefore, the criterion validity was measured by comparing the common items of the LFPI and the BTAT. The analysis of the concurrent validity between protocols demonstrated negative correlation since the protocols have inverse scales.

The construct validity was performed by comparing the partial scores from the assessment of infants with and without lingual frenulum alteration at 30 and 75 days of life as well as by comparing the total scores from the assessment of the infants with and without lingual frenulum alteration at 30 and 75 days of life. The post-operative images and reports were analyzed, and preoperative and post-operative clinical history and clinical examination were compared. Post-operative data from the infants with lingual frenulum alteration, who underwent frenotomy, and data of the infants without lingual frenulum alteration

were compared to verify possible changes. Results from post-operative assessment demonstrated agreement with the literature concerning the importance of frenotomy for infants with lingual frenulum alteration: improvements in breastfeeding patterns such as better latching, improvement of sucking (greater number of suctions), pauses (shorter pauses between groups of sucking), improvement in sucking, swallowing, and breathing coordination, less nipple pain, and breastfeeding maintenance (Knox, 2010; Buryk, Bloom, & Shope, 2011; Constantine, Williams, & Sutcliffe, 2011; Berry, Griffiths, & Westcott, 2012; Martinelli, Marchesan, Gusmão, Honório, & Berretin-Felix, 2015; Emond, Ingram, Johnson, Blair, Whitelaw, Copeland, & Sutcliffe, 2014).

Regarding inter/intra-rater agreement, independent assessments of the 100 infants by two examiners (E1 and E2) and the re-assessment of 20% of the sample by E2 demonstrated the reliability of the LFPI. Training and calibration between examiners are essential requirements for reliability and repeatability of the assessment tool. The results demonstrated excellent repeatability of the LFPI. The reliability results were found to be better than the results obtained by the Hazelbaker Assessment Tool for Lingual Frenulum Functions – HATLFF (Amir, James, & Donath, 2006) and the BTAT (Ingram, Johnson, Copeland, Churchill, Taylor, & Emond, 2015).

The scientific rigor of an assessment tool is greater when, besides validity and reliability analyses, S, SPC, PPV, and NPV are added (Menz, Latt, Tiedemann, Mun San Kwan, & Lord, 2004). The research demonstrated good S, SPC, PPV, and NPV for the cutoff point adopted. Those values indicate that the LFPI is an effective tool to diagnose subjects with and without lingual frenulum alteration. Lingual frenulum assessment protocols with S, SPC, PPV and NPV analyses were not found in literature.

The literature cites several definitions, classifications and assessment tools for lingual frenulum (Messner, Lalakea, Aby, MacMahon, & Bair, 2000; Ballard, Auer, & Khoury, 2002; Suter, Bornstein, 2009). The present study

considered the anatomical-functional aspects, sucking and swallowing during breastfeeding, and breastfeeding history for the lingual frenulum alteration diagnosis. Lingual frenulum alteration occurrence in this study was 21%. Researches on lingual frenulum alteration from other countries have reported 0.1% to 12.8% of occurrence (Messner, Lalakea, Aby, MacMahon, & Bair, 2000; Ballard, Auer, & Khoury, 2002; Suter & Bornstein, 2009). The criteria for assessment established by this study may explain the high occurrence of lingual frenulum alteration found in the study. Moreover, different definitions of lingual frenulum alterations provided by the literature and different lingual frenulum assessment tools might explain the difference of occurrence reported (Knox, 2010; Berg, 1990; Wallace & Clarke, 2006).

The LFPI was validated to identify the lingual frenulum alteration in infants considering content validity, criterion validity, construct validity, reliability, sensitivity, specificity, positive predictive value and negative predictive value. Since the absence of agreement on the criteria used for evaluation and anatomical classification of the lingual frenulum may be the cause of the variation in the reported occurrence rates of ankyloglossia, specific protocols may lead to more standardized assessment, planned therapeutic actions, clinical procedure documentation, evidence-based clinical practice, and may contribute to future epidemiological studies on the occurrence of ankyloglossia.

CONCLUSION

The study concluded that the LFPI is found to be a valid and reliable assessment tool, ensuring accuracy to diagnose lingual frenulum alterations within the investigated parameters. The LFPI is an effective tool that can be used to differentiate the anatomic variations of lingual frenulum for an assertive diagnosis and better treatment plan. It may be administered by other health professionals, provided that they are qualified and trained to administer the protocol.

CONTACT AUTHOR:

Roberta Lopes de Castro Martinelli, SLP, Ph.D. Hospital Santa Therezinha, Avenida Rui Barbosa, 703, Centro, Brotas, SP, Brazil CEP17380-000.

Business telephone number: +551138715745

Fax number: +551138680818

e-mail - robertalcm@gmail.com

Other Affiliations:

CEFAC, São Paulo, SP, Brasil and
Faculdade de Odontologia de Bauru da
Universidade de São Paulo, Bauru, SP, Brasil

Irene Queiroz Marchesan, SLP, Ph.D.
CEFAC, São Paulo, SP, Brasil

José Roberto Lauris, Ph.D.
Faculdade de Odontologia de Bauru da
Universidade de São Paulo, Bauru, SP, Brasil

Heitor Marques Honório, Ph.D.
Faculdade de Odontologia de Bauru da
Universidade de São Paulo, Bauru, SP, Brasil

Reinaldo Jordão Gusmão, Ph.D.
Faculdade de Ciências Médicas da
Universidade de Campinas, Campinas, SP,
Brasil.

Giédre Berretin-Felix, Ph.D.
Faculdade de Odontologia de Bauru da
Universidade de São Paulo, Bauru, SP, Brasil

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