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ORIGINAL ARTICLE

Ankyloglossia: a morphofunctional investigation in children

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OBJECTIVES: To provide diagnostic criteria for ankyloglossia in children by anatomical measurements; to investigate the correlation between severity of ankyloglossia and a series of morphofunctional findings; to evaluate the potential mismatch between a clinical suspect of ankyloglossia and the authentic anatomical diagnosis.
DESIGN: Two different techniques of anatomical measurements and a clinical evaluation of a series of morphofunctional findings were performed.
SUBJECTS AND METHODS: In 200 children referred for evaluation and treatment of tongue-tie, the length of the frenulum and the interincisal distance were measured in maximum opening of the mouth and with the tip of the tongue touching the palatal papilla. Occlusion, type of bite, tongue resting position, swallowing mechanism, oral floor mobility, frenulum insertion modality and speech were investigated. Any correlation between these morphofunctional findings and anatomical measures was investigated.
RESULTS: Children with a frenulum length more than 2 cm and an interincisal distance of more than 2.3 cm were normal. In both measurements, significant correlations among mean values and other variables were observed. Moreover, three levels – mild, moderate and severe – of ankyloglossia were assessed.
CONCLUSIONS: Length of frenulum and interincisal distance allow an assessment of severity of ankyloglossia in children. Ankyloglossia was not associated with infantile swallowing.
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Keywords: ankyloglossia; tongue-tie; children

Introduction

Ankyloglossia – from the Greek words 'agkilos' for crooked and 'glossa' for tongue – or 'tongue-tie' represents a congenital oral anomaly characterized by an abnormally short lingual frenulum that may affect the mobility of the tongue. The lingual frenulum recedes as a natural process of the child's growth and development that occurs during the first 6 months to 6 years of life. Ankyloglossia occurs when the lingual frenulum persists as an anatomical abnormality. The severity of ankyloglossia varies from absence of clinical significance to a completely fixed tongue to the floor of the mouth. The clinical consequences of ankyloglossia include difficulties during infant feeding and swallowing, speech problems, orthodontic and orthopedic anomalies, and social/mechanical problems (Wright, 1995; Messner and Lalakea, 2000). A unified method of classifying the severity of ankyloglossia is lacking. Few studies have proposed standards of classification for ankyloglossia by employing anatomical and/or functional criteria, but they measure different things (Fletcher and Meldrum, 1968; Fletcher and Daly, 1974; Hazelbaker, 1993; Kotlow, 1999; Ballard *et al*, 2002; Garcia Pola *et al*, 2002). The management of tongue-tie also varied (Wilder and Gelesko, 1997; Kotlow, 1999; Sanchez-Ruiz *et al*, 1999; Messner and Lalakea, 2000; Garcia-Pola *et al*, 2002; Lalakea and Messner, 2003). A variety of methods for the repair of tongue-tie are used, including the incision of the frenulum ('clipping' or frenotomy), the frenectomy and the frenuloplasty (Kotlow, 1999; Lalakea and Messner, 2003). Complications of surgery may further limit tongue movement (Lalakea and Messner, 2003).

The aim of the study was to provide diagnostic criteria for ankyloglossia in children by means of anatomical measurements; to investigate the correlation between severity of ankyloglossia and a series of morphofunctional findings.

Subjects and methods

Subjects

The study was performed in a group of 200 children aged 6–12 years (100 males and 100 females; mean age 9.3 years; 1.3 s.d.), attending the Department of Pedodontology of Santa Chiara's Hospital in Pisa (Italy) for evaluation and treatment of tongue-tie. The subjects

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were referred by their pediatricians or family doctors. None of the children had undergone orthodontic treatment prior the study or had medical problems. All the patients were examined by the same two specialists in orthodontics (GM, GMR).

Measurement techniques
We performed two different methods of anatomical measurements to classify the ankyloglossia. Both types of measurement were carried out at maximum opening of the mouth and with the tip of the tongue touching the palatal papilla. The first (technique A) measured directly the length of the frenulum by recording the distance between the insertion of the lingual frenulum into oral floor and the tongue (Figure 1). In the second (technique B), an indirect evaluation of the length of frenulum was performed by measuring the distance between the incisal margins of the upper central and the lower homolateral central incisor (Figure 2). All measurements were taken using a Boley gauge with results expressed in centimeters and read to the nearest millimeter.

Morphological and functional findings
As reported in Table 1, we considered for each patient gender, occlusion according to Angle's (1899) classification, type of bite (cross-bite, deep-bite, anterior open-bite, posterior open-bite), habitual resting position of the tongue (normal, low or interposed), mechanism of swallowing (infantile or mature), mobility of the oral floor (present or absent), modality of frenulum insertion according to Mukai *et al* (1991), and mobility of the tongue. Moreover, for each patient a speech evaluation was carried out by three different speech therapists, blinded to the clinical status of the examined child. For this purpose, the pronunciation of 21 different phonemes was evaluated. In more detail, each child had to read a list of well-defined words for each of the 21 considered phoneme. For all children examined, the number of words and the words chosen for each considered phoneme remained always the same.

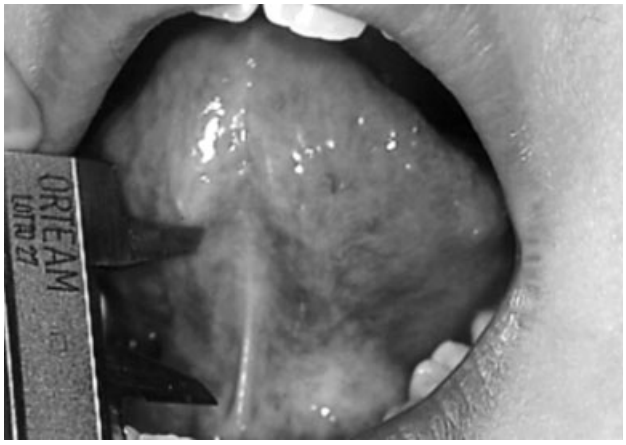

Figure 1 Technique A – The length of the frenulum was measured by recording the distance between the insertion of the lingual frenulum into oral floor and the tongue

Figure 2 Technique B – The length of frenulum was indirectly evaluated by measuring the distance between the incisal border of the upper central incisor and the incisal border of the lower homolateral central incisor

Statistics
The distribution of the observed values with the two performed techniques was based on quartile values. Student's *t*-test to compare mean values between two level variables and the analysis of variance to compare means among classes of multi-level variables were used. Pearson's correlation coefficient was used to test the relationship between continuous variables. SAS system was used to perform statistical computations. Data are expressed as mean value ± s.d. Significance was considered at the 0.05 α level.

Results

For each morphofunctional finding investigated, the numbers of the patients and the mean values of measurement obtained from the techniques A and B were recorded, and the presence of significant differences among the mean values were investigated (Table 1). We did not find any age-related difference in the mismatch between prevalence of authentic ankyloglossia and suspected problems with the tongue-tie which could justify a role of the age in affecting our data. For both methods of measurement, significant differences among the mean values were observed when occlusion, type of bite, resting position of the tongue, modality of frenulum insertion, mobility of the tongue and speech were considered (Table 1). The mobility of the oral floor showed significant difference only between the mean values obtained from the technique B (Table 1). In fact, the mean values obtained from the technique A were not influenced by this morphofunctional finding as they originated from direct measures of the length of frenulum. Concerning modality of swallowing and gender of the patients, no significant difference was shown in the mean values (Table 1). Twenty-three children (11 males and 12 females) did not show any anomalies for any of the investigated morphological and functional findings, considered healthy subjects, and the

Table 1 Mean values obtained by measurements of the length of frenulum (Technique A) and measurement of the interincisal distance (Technique B) and correlation with the morphofunctional findings

		Technique A		*Technique B*	
Morphofunctional findings	*Patients*	*Mean ± s.d.*	P-*value**	*Mean ± s.d.*	P-*value**
Gender			NS		NS
Male	100	1.85 ± 0.10		2.16 ± 1.20	
Female	100	1.81 ± 0.96		2.14 ± 1.82	
Modality of occlusion			$P < 0.001$		$P < 0.001$
Class 1	89	1.89 ± 0.44		2.20 ± 0.62	
Class 2	76	1.91 ± 0.44		2.19 ± 0.70	
Class 3	35	1.49 ± 0.58		1.84 ± 0.85	
Type of bite			$P < 0.042$		$P < 0.024$
Normal	77	1.88 ± 0.43		2.17 ± 0.65	
Anterior open-bite	66	1.86 ± 0.46		2.24 ± 0.67	
Deep-bite	50	1.67 ± 0.60		1.93 ± 0.80	
Cross-bite	3	2.33 ± 0.11		3.03 ± 0.58	
Posterior open-bite	4	1.87 ± 0.96		2.35 ± 0.49	
Resting position of the tongue			$P < 0.004$		$P < 0.024$
Normal	151	1.90 ± 0.44		2.20 ± 0.62	
Low	41	1.56 ± 0.58		1.90 ± 0.94	
Interposed	8	1.92 ± 0.49		2.47 ± 0.57	
Mechanism of swallowing			NS		NS
Mature	110	1.86 ± 0.99		2.22 ± 1.21	
Infantile	90	1.70 ± 0.81		2.06 ± 1.15	
Mobility of the oral floor			NS		$P < 0.001$
Present	149	1.86 ± 0.91		2.70 ± 0.64	
Absent	51	1.76 ± 0.81		1.57 ± 0.77	
Modality of frenulum insertion			$P < 0.001$		$P < 0.001$
Grade F1	97	1.96 ± 0.40		2.30 ± 0.59	
Grade F2	96	1.77 ± 0.51		2.07 ± 0.72	
Grade F3	7	0.91 ± 0.30		0.87 ± 0.54	
Mobility of the tongue			$P < 0.001$		$P < 0.001$
Movement accurated and rapidly performed	135	2.06 ± 0.32		2.40 ± 0.54	
Movement inaccurately performed	39	1.55 ± 0.43		1.94 ± 0.71	
Movement incompletely performed or absent	26	1.06 ± 0.27		1.81 ± 0.48	
Speech			$P < 0.001$		$P < 0.009$
Normal	147	1.91 ± 0.46		2.10 ± 0.70	
Incorrect	53	1.46 ± 0.51		1.75 ± 0.69	

Mean values expressed in centimetres.
s.d., standard deviation; NS, not significant.
*As result of analysis of variance.
Significance: $P < 0.05$ α level. Speech Evaluation
Phonemes investigated: /p, t, k, b, d, g, f, v, s, S, z, ts, dz, tS, dZ, m, n, J, l, L, r/.
Each pronunciation was assessed categorically as normal or incorrect (substituted, omitted, distorted or nasalized).

mean values were 2.04 cm ± 0.32 s.d. and 2.33 cm ± 0.53 s.d., respectively with technique A and technique B. The enrolled children were distributed in quartiles based on range values 0.7–1.5, 1.6–1.9, 2.0–2.2, 2.3–2.9 in technique A, and 0.3–1.6, 1.7–2.2, 2.3–2.7, 2.8–4.0 in technique B. This allowed three levels of ankyloglossia (mild, moderate and severe) for each technique of measurement (Table 2). Children with lower values (2.0 cm for A; 2.3 for B) were defined as having various grades of ankyloglossia (Table 3). The distribution of the patients in each grade with their morphological findings are shown in Table 3.

Discussion

Although the results suggested that the two classifications might be considered as similar, that obtained by the interincisal distance was preferred due to the following: The measurement of the interincisal distance was simpler and better tolerated. The values obtained from this method of measurement were representative of mobility of the oral floor.

Ankyloglossia is reported to be more common in males than in females. Studies carried out in newborns and young infants documented a prevalence male-to-female ranging from 1.6 to three times (Friend *et al*, 1990; Messner *et al*, 2000; Ballard *et al*, 2002). In our study, we found slight difference female-to-male in children with ankyloglossia (see Table 3). This could be a sampling bias of our patients being not a random sample and our data might simply reflect clinical presentation by gender-related variables. Moreover, in our selected group of children by anatomical measurements, we found a large number of subjects who did not

Table 2 Classification of levels of ankyloglossia

	Technique A	*Technique B*
Level of normality	≥2 cm	≥2.3 cm
Level of ankyloglossia		
Mild	1.6–1.9 cm	1.7–2.2 cm
Moderate	0.8–1.5 cm	0.4–1.6 cm
Severe	≤0.7 cm	≤0.3 cm

have ankyloglossia. There are only a few studies reporting contrasting results concerning our ankyloglossia and occlusion anomalies. For instance, Tuerk and Lubit (1959) reported the presence of open-bite and Angle's class 3 malocclusion in patients with ankyloglossia and infantile swallowing pattern. In line with this, Mukai *et al* (1993) demonstrated that 84% of the examined patients with ankyloglossia had Angle's class 3 malocclusion. On the contrary, Mazzocchi and Clini (1992) and Garcia Pola *et al* (2002) could not find any relation between the short length of the frenulum and the occurrence of either dental, or orthodontic anomalies. In the present study, we found that 61.5% of the enrolled subjects possessed bite anomalies which were related with the anatomical measures used for ankyloglossia. In particular, the patients with deep-bite possessed the lowest mean value which was statistically significant compared with other patients. In contrast, the subjects with posterior or anterior open-bite or cross-bite showed mean values similar or even longer compared with children with normal bite (Table 1). By Angle's classification of occlusion the majority (55.5%) of the enrolled children possessed malocclusions. Both measurements were significantly the lowest in patients of Class 3. A protrusive chin may be considered as a result of either maxillary hypodevelopment or mandibular hyperdevelopment because of low tongue posture and ankyloglossia (Couly, 1989; Defabianis, 2000). In our study, the patients belonging to class 3 possessed a low tongue posture. Taken together, with previous observations the type of bite, occlusion and resting position of tongue are useful clinical predictors for the diagnosis of severity of ankyloglossia.

The current literature reports swallowing abnormalities associated with ankyloglossia, although a complete agreement is difficult to reach (Wright, 1995; Sanchez-Ruiz *et al*, 1999). The disagreement could be due to the maturation of the mechanism of swallowing. In babies swallowing is infantile type, by 2 and 4 years of age it

Table 3 Patients in each level of ankyloglossia and normality and their distribution for each morphofunctional finding

Values of measurement expressed in centimeters	*Technique A*				*Technique B*			
	0.7	*0.8–1.5*	*1.6–1.9*	*≥2*	*0.3*	*0.4–1.6*	*1.7–2.2*	*≥2.3*
Number of patients	6	49	56	89	3	50	53	94
Gender								
Male	4	23	24	49	1	25	26	48
Female	2	26	32	40	2	25	27	46
Modality of occlusion								
Class 1	2	15	29	43	0	17	26	46
Class 2	0	18	22	36	2	17	20	37
Class 3	4	16	5	10	1	16	7	11
Type of bite								
Normal	1	14	25	37	2	15	24	36
Anterior open-bite	2	16	19	29	0	16	17	33
Deep-bite	3	19	9	19	1	19	11	19
Cross-bite	0	0	0	3	0	0	0	3
Posterior open-bite	0	0	3	1	0	0	1	3
Resting position of the tongue								
Normal	2	29	49	71	1	31	47	72
Low	4	18	5	14	2	18	5	16
Interposed	0	2	2	4	0	1	1	6
Mechanism of swallowing								
Mature	2	23	35	50	2	22	28	58
Infantile	4	26	21	39	1	28	25	36
Mobility of the oral floor								
Present	5	33	40	71	1	27	41	80
Absent	1	16	16	18	2	23	12	14
Modality of frenulum insertion								
Grade F1	0	15	33	49	1	12	29	55
Grade F2	3	30	23	40	1	33	23	39
Grade F3	3	4	0	0	1	5	1	0
Mobility of the tongue								
Movement accurated and rapidly performed	0	6	46	83	0	11	41	83
Movement inaccurately performed	1	23	9	6	0	17	11	11
Movement incompletely performed or absent	5	20	1	0	3	22	1	0
Speech								
Normal	2	29	43	73	3	25	45	74
Incorrect	4	20	13	16	0	25	8	20

turns into a mature pattern (Peng *et al*, 2004). The persistence of the infantile swallowing pattern might be due to the inability to elevate the tongue (see Wright, 1995, for a review). In our study, we could not confirm this observation because patients having the infantile swallowing pattern did not differ in the frenulum length and the interincisal distance from those who developed the mature swallowing pattern. Nonetheless, patients with infantile swallowing showed anomalies of bite (i.e. anterior open-bite and deep-bite) which are commonly related to ankyloglossia, although we could not find a significant correlation between infantile swallowing and ankyloglossia. Concerning the effect of tongue-tie on speech, there is general agreement that this complaint neither prevents nor delays the speech onset and plays only a minor role in the etiology of speech disorders. Nonetheless, a decreased tongue mobility slightly impairs speech articulation with an emphasis on selective phonemes. In the present study, we could demonstrate a significant impairment of speech in patients carrying ankyloglossia confirming previous studies. In normal children, Fletcher and Meldrum (1968) documented errors in speech articulation in those subjects carrying 'limited lingual freedom', when compared with subjects with 'greater lingual freedom'. Significant association between short lingual frenulum and lingual dysglossia, such as rhotacism, was referred (Garcia Pola *et al*, 2002). In children with ankyloglossia, Messner and Lalakea (2002) found errors in articulating formal speech which they attributed to a decreased tongue mobility. However, they ruled out the occurrence of ankyloglossia as a determining factor because the interincisal distance was not significantly different between patients with abnormal compared with normal speech. In this study the authors did not use a standardized speech test which was evaluated by not blinded speech pathologists, calling for further investigations. In the present study, we could demonstrate that patients with abnormal speech showed mean values of measurement significantly lower than patients with normal speech, in a high number of children undergoing the same standardized speech test evaluated by blind speech pathologists. In our children, we found a relationship between the presence of speech anomalies and a decreased mobility of the tongue, but only for those subjects whose frenulum length resulted in moderate or severe levels of ankyloglossia.

In our study, the modality of frenulum insertion resulted to be significantly related with the grade of severity of ankyloglossia. In fact, the distribution of the patients in the four levels of both classifications varied according the modality of frenulum insertion. While the majority of the patients with grade F1 showed normal values of measurement, the greater part of patients with grade F2 had mild or moderate ankyloglossia. Finally, none of the children with grade F3 showed normal values of measurement. This last observation might lead to believe the presence of grade F3 of frenulum insertion as a diagnostic index of ankyloglossia. However, as only seven children with grade F3 were present in our study, we preferred to consider the presence of grade F3 of frenulum insertion more as an index of complaint rather than a diagnostic index of ankyloglossia.

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