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ORIGINAL ARTICLE

Instrumented Assessment of Oral Motor Function in Healthy Subjects and People with Systemic Sclerosis

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Abstract The aim of the present study was to provide quantitative data of oral function in healthy subjects (HSs), validity of measurements and estimation of measurement bias, as well as quantify oral impairment in persons with scleroderma (SSc). 151 HSs and 12 subjects with SSc were recruited and assessed using instrumented tools, measuring maximal mouth opening; lip strength; and tongue strength, protrusion, retraction, and endurance. Twenty HSs were also retested 3–5 weeks later in order to assess the test–retest reliability of the measurements. Intraclass correlation coefficients proved to be satisfactory (>0.8) for both inter-rater and test–retest reliabilities of all measurements except for tongue retraction. In the HS group, maximal mouth opening and tongue and lips strength values were larger ($P < 0.05$) for males than females, while no significant differences were found for other variables. Older subjects had statistically significantly lower tongue retraction values and tongue endurance values than younger subjects. The SSc group showed a statistically significant decrease ($P < 0.05$) in almost all the measurements. Assessment procedures proved to be valid and reliable. Gender and height were predictors of mouth opening, lip and tongue strength, while age correlates with tongue retraction and endurance. Measurements highlighted the strong impact of SSc on oral functions and in particular on tongue protrusion, tongue strength, and endurance.

Keywords Oral motor function · Mouth · Tongue · Lips · Deglutition · Scleroderma

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Introduction

Oral impairments in persons with neurological and multisystemic disorders are common and frequently assessed in the clinical setting. Clinical appraisal of subjects with oral disorders requires a multivariate assessment in order to collect data on body structure, body function, and participation [1, 2]. A clinical and instrumental assessment is critical to identify abnormal oral physiology, allowing for a better understanding of the recovery process and possible prescription of therapy [3]. Clark et al. [4] and Solomon et al. [5] recommended the use of objective measures as more valid and reliable than subjective measures for the assessment of oral functions. The importance of objective quantitative assessment is highlighted by several studies in which devices have been proposed to assess tongue and lips physiology [6–12] and oral dysfunction such as swallowing disorders and dysarthria.

The tongue is composed of a complicated arrangement of extrinsic and intrinsic muscles and is arguably the most important oral structure for speech articulation and swallowing. The sounds of speech traditionally are classified by the general position of the tongue as it shapes the upper airway to filter sound. During oral and pharyngeal stages of swallowing, the tongue moves in several dimensions in the oral cavity for bolus preparation, to propel the bolus to the oropharynx, and it is involved in pharyngeal wall movements [13]. Cantilever beams [3, 14], air-filled bulbs [10–12, 15], and sensor sheets [16] have been used to evaluate maximum tongue pressure in normal subjects or to compare dysphagic subjects with non-dysphagic subjects. Most of these studies found an age effect on maximal force production in healthy subjects, while there is some controversy on the effect of gender. A reduction of force

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output has been documented in dysphagic and dysarthric subjects [17, 18].

The lips and lower face muscles also play an important role in the initiation of deglutition [9] and provide cues to the emotional or pragmatic intent of the message while simultaneously forming the sound of the message itself [9, 19]. The lips comprise circumferential muscles, the orbicularis oris superior and inferior; other muscles contribute to mouth closure and aperture, like mentalis, risorium, and buccinators. Mouth closure is important for swallowing so that the bolus stays contained in the mouth during oral phases of swallowing. For speech labial approximation, lip elevation and retraction, protrusion, and rounding occur for certain phonemes [19]. Devices have been developed to assess interlabial pressure during speech [20]; perioral stiffness [6] and force [7]; force and control of orofacial structures [8, 9, 21]; lip strength (Lstren), including a strain gauge dynamometer [22], an ultraminiature transducer [3], and an oral myometer [23]. Moreover, strength measures have been obtained using the Iowa Oral Performance Instrument [15].

Mouth opening and closure is important for the oral phase of swallowing, oral hygiene, dental treatment, for the positioning of denture teeth and the articulation of many consonants. With respect to maximal mouth opening (MMO), several researchers measured the distance between the incisal edge of the upper and lower first incisors using the Willis Bite Gauge [24, 25], showing that MMO is influenced by gender, age, and anthropometric measurements [26].

Several studies showed the impact of mobility reduction, fatigue, and weakness of oral structures leading to swallowing disorders [27, 28]. Reddy et al. studied dysphagia measuring lip closure pressure, lip interface shear force, tongue thrust, and swallow pressure and found "significant differences in each of these parameters measured in normal and dysphagic patients" [3], while Stierwalt et al. found a reduced tongue strength but similar tongue endurance in individuals with dysphagia compared with healthy subjects [29].

Conflicting results have been reported on the relationship between oral impairment and dysarthria. Solomon in a review pointed out that the relationship between weakness and fatigue of oral structures and speech mobility reduction is still unclear and that preliminary evidence indicates that speech function is rather robust [30]. Further, a review of the literature on dysarthria in subjects with neurological conditions found no difference for tongue strength and pressure control between the control group and dysarthric subjects after traumatic brain injury [18].

Systemic sclerosis (SSc), also known as scleroderma, is a multisystemic disorder characterized by fibrosis, vascular obliteration, and capillary vascular changes that involve the skin and internal organs [31]; women are four times more likely to develop this condition. The face and the mouth are frequently involved in systemic sclerosis: the main stomatologic manifestations include limited mouth opening, xerostomia, skin atrophy, trigeminal neuralgia [32]. Structural abnormalities lead to damages to oropharyngoesophageal mucous membrane, mastication muscles, and salivary glands causing changes of voice and mouth functions, (i.e., speaking, chewing, and swallowing). Eighty percent of subjects with SSc have orofacial manifestations [33]. Vitali et al. [26] assessed subjects' perception of his/her oral disorders, in 84 subjects with SSc, in several domains such as structure impairments, swallowing, and quality of life and found that 36 % of subjects showed moderate to severe oropharyngolaryngeal involvement. Swallowing disorders and impairment of mouth (e.g., decrease in mobility and strength) were the most commonly reported problems, further 55 % of subjects reported oral-related decreased quality of life. Unfortunately, few quantitative studies on mouth impairment are available making reliable and objective description of tissues damage and functional disorders not easy. This scarcity of reliable information leads to difficulties in comparing subject's perception of oral disorders with instrumental measurements and in assessing the impact of treatments on oral structures.

The overall aim of the present study was to expand the previous findings on oral disorders in SSc subjects [31–33] and to quantitatively measure oral impairment and treatment outcome after rehabilitation intervention also in subjects with severe limitation in mouth opening.

The specific aims of the present study were to (1) provide normative data on oral function, (2) study validity of measurements and estimation of measurement bias of healthy subjects, and (3) compare healthy subjects with subjects with SSc to provide an initial overview of oral impairments in this pathology and to assess discriminant validity.

Materials and Methods

Sampling

A sample of 151 Healthy Subjects (81 females, 70 males) without temporomandibular or oropharyngeal disorders was tested, and 20 of these were retested 3–5 weeks later in order to assess the test–retest reliability of the measurements. Mean age (standard deviation) was 47.8 (17.2) years for Healthy Subjects (HSs). The sample was stratified by age, the age groups being 20–40; 41–60; and 61–81 (Table 1). Mean reported heights for HSs were 1.64 (0.06) m and 1.75 (0.06) m, respectively, for females and males.

Table 1 Frequency and height of subjects in each age group

	Age group (years)	HS male ($n = 70$)	HS female ($n = 81$)	SSc female ($n = 12$)
Number of subjects	20–40	26	30	1
	41–60	23	30	2
	61–81	21	21	9
Height (m)	20–40	1.78 (0.07)	1.65 (0.07)	1.65 (–)
	41–60	1.75 (0.06)	1.63 (0.05)	1.56 (–)
	61–81	1.70 (0.05)	1.62 (0.05)	1.61 (0.06)

Heights are reported as mean and (Standard Deviation)

HS healthy subjects, *SSc* subjects with scleroderma

A sample of 12 females, with known SSc having a mean age of 63.1 (12.7) years and mean reported height of 1.60 (0.06) m, with the same inclusion–exclusion criteria was assessed.

The study was approved by the local ethical committee. All subjects received information regarding the study and were included after signing the informed consent forms.

Instrumental Assessment

On the bases of preceding studies that assessed oral impairments, we developed a battery of tests using existing and new devices adapted to assess oral disorders for subjects with SSc (Fig. 1). Tests and parameters tested are described below (Table 2):

Test 1 MMO was measured in millimeters, using calipers, as the distance between the incisal edge of the upper and lower first incisors [24, 25].

Test 2 According to Sjögren et al. [34], Lstren was measured by a device consisting of a button (diameter 3 cm, as used in myofunctional therapy) connected to an elastic band of known stiffness [N/m]. The button was placed between lips and incisal teeth. The elastic band was pulled by the rater's hand using a custom-made device (Fig. 1a). The device moved on a graduate track to allow a precise assessment of the lengthening of the elastic band until the button slipped out from between the lips. The final measure of the elastic band was multiplied by its stiffness to calculate Lstren (in Newtons).

Test 3 As suggested by Reilmann et al. [35], tongue protrusion strength (Tstren) in the sagittal plane was measured in Newtons by asking the subject to push as hard as possible against a CITEC[1] digital dynamometer (Fig. 2).

Test 4 Anterior tongue retraction (Tretr) was assessed using calipers by asking the subject to move his or her tongue backward while keeping it close to the floor of the mouth (millimeters). The distance between the incisal edge and the tip of the tongue was measured.

Test 5 Tongue protrusion (Tprot) was measured in millimeters using a RUPAC dial indicator,[2] an instrument measuring distances with a contact point attached to a spindle and gears that moves a pointer on the dial (Fig. 1b). We asked the subjects to push the rod connected to the strain gauge dial, as far as possible as reported elsewhere [36] (Fig. 1b).

Test 6 Similarly, to Goozée et al. [18], tongue endurance (Tend) was measured using a dial indicator (Fig. 1b), by asking the subject to move his or her tongue back and forth as many times as possible for 60 s, pushing the spindle connected to the dial indicator from the starting position (the lips) to final position.

The mean value across two repetitions of the Test 5 was calculated to set the target position, thus normalizing the variable to each person's own protrusions measure. An acoustic feedback was provided each time the spindle reached the target. Distance covered by the tongue was calculated as maximum Tprot $\times$ number of movements (millimeters) to provide an endurance-related variable. We chose this procedure because it was more efficient in discriminating between HSs and SSc subjects than simply counting the number of repetitions.

Each subject was assessed by a speech therapist. Standardized sentences were used to explain the tests to the subjects. The order of assessments was quasi-randomized to take fatigue into account. Tests were divided into blocks of more (Lstren, Tstren, Tend) and less (MMO, Tretr, Tprot) demanding tests. The more demanding and less demanding blocks were randomized.

A latex protection was applied to the instruments and they were sterilized before and after each assessment. Before the start of data collection, a preliminary study on 10 healthy subjects [mean age 43.7 (19.6) years] was carried out in order to refine the methodology and assess the stability of measurements during a single assessment session. Eight measurements were taken for each instrument for each subject. An intraclass correlation coefficient (ICC) was used to calculate the agreement between

[1] http://www.citec.nu/frm/uk.htm.

[2] http://www.rupac.com/.

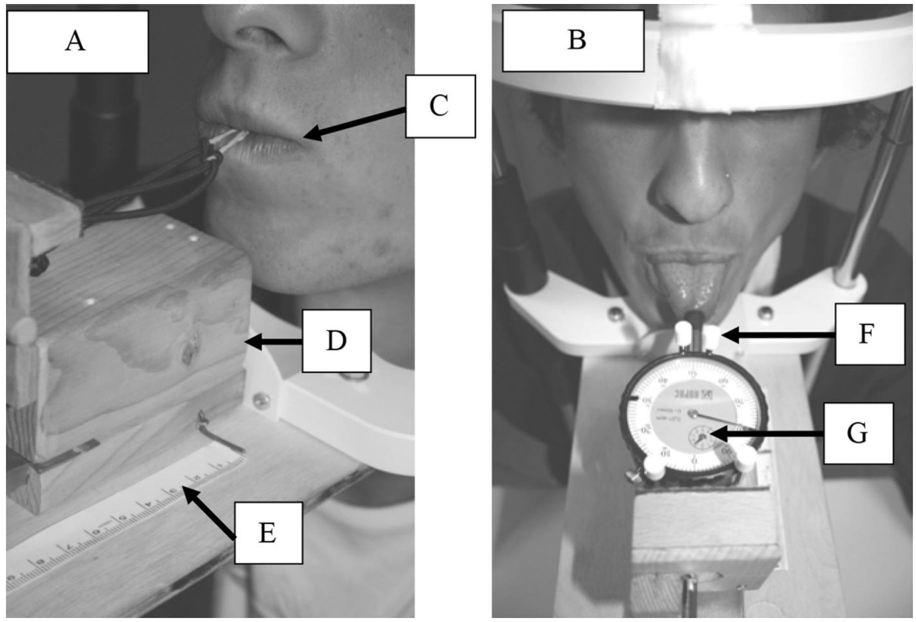

Fig. 1 (**A**) Measurement of lip strength. After button placement (*C*), the wooden block (*D*), which is moved on a track, was pulled by the rater until the button slipped out from between the lips. The length of the elastic band was measured by the marker showed in (*E*). (**B**) Measurement of tongue protrusion. The subject was required to move the tongue as far as possible by pushing the rod (*F*) connected to the strain gauge dial (*G*)

Table 2 Instrumental tests performed during the assessment procedure

Test number	Test	Abbreviations	Unit of measurement
1	Maximal mouth opening	MMO	Millimeters
2	Lip strength	Lstren	Newtons
3	Tongue protrusion strength	Tstren	Newtons
4	Anterior tongue retraction	Tretr	Millimeters
5	Tongue protrusion	Tprot	Millimeters
6	Tongue endurance	Tend	Millimeters

Fig. 2 Body posture during the assessment of tongue protrusion strength using the dynamometer. The same posture was requested for the assessment of lip strength, tongue protrusion, and tongue endurance

measurements. Results showed high ICCs (above 0.8) even with only two measurements, thus two repetitions for each test were taken in the final study.

Posture

In order to improve the reliability of the measurements, the subject's posture was standardized for all assessments except for test 1 and test 4 (Fig. 2). Movements of the trunk and shoulders were controlled by the backrest of the chair and by the desk, the subject's forehead was placed on a support in order to keep the head vertical and avoid head and neck movements, especially in the sagittal plane. Jaw motion was unrestrained because jaw movements increase tongue strength and mobility [3, 37]. An inclinometer placed on the head controlled unwanted head movements by providing an acoustic signal if the head was rotated in the sagittal plane.

Data Analysis

Descriptive statistics were used to describe the study population in terms of demographics and disease characteristics.

Median and 5th–95th percentiles were calculated in order to provide normative ranges. The association

Table 3 Standard error of measurement and intraclass correlation coefficients for inter-rater and test–retest reliabilities

	Inter-rater				Test–retest			
	ICC	CI−	CI+	SEM	ICC	CI−	CI+	SEM
MMO	0.97	0.93	0.98	0.16 [mm]	0.83	0.56	0.93	0.57 [mm]
Lstren	0.84	0.6	0.94	2.49 [N]	0.87	0.67	0.95	1.89 [N]
Tstren	0.97	0.94	0.99	1.75 [N]	0.97	0.92	0.99	1.93 [N]
Tretr	0.99	0.99	1.00	0.04 [mm]	0.77	0.42	0.91	0.67 [mm]
Tprot	0.93	0.82	0.97	0.55 [mm]	0.90	0.75	0.96	0.67 [mm]
Tend	0.92	0.79	0.97	96.51 [mm]	0.86	0.65	0.95	114.05 [mm]

ICC Intraclass correlation coefficient, *CI* ±95 % confidence interval, *SEM* standard error of measurement, *MMO* maximal mouth opening, *Lstren* lip strength, *T stren* tongue protrusion strength, *T retr* anterior tongue retraction, *T protr* tongue protrusion, *T end* tongue endurance

Table 4 Medians, 5th–95th percentiles and *P* values for male, female HS, and subjects with Scleroderma

	HSs ($n = 151$) Median (5th–95th percentiles)	Males ($n = 70$) Median (5th–95th percentiles)	Females ($n = 81$) Median (5th–95th percentiles)	P1 HS males versus HS females	SSc ($n = 12$) Median (5th–95th percentiles)	P2 Hs female versus SSc
MMO (mm)	43.0 (32.5–54.0)	46.2 (31.5–56.0)	41.0 (33.0–49.5)	>.0450	37.8 (19.0–44.0)	0.0013
Lstren (N)	7.6 (2.7–12.0)	9.0 (3.5–12.7)	6.5 (2.7–9.8)	>.0001	5.3 (0–11.0)	0.1307
Tstren (N)	12.0 (6.0–22.0)	15.0 (6.0–23.0)	11.0 (5.5–19.0)	>.0001	6.5 (2.0–17.5)	0.0197
Tretr (mm)	29.0 (17.0–42.5)	28.8 (12.0–43.5)	29.5 (19.0–40.0)	0.9244	24.8 (11.0 –38.5)	0.098
Tprot (mm)	49.8 (30.4–66.1)	48.9 (32.6–65.3)	49.9 (28.6–66.3)	0.7499	27.2 (2.2 –56.6)	>.0001
Tend (mm)	317.6 (130.5–529.8)	333.4 (131.4–536.1)	311.9 (116.3–528.6)	0.1557	177.2 (7.2–855.4)	>.0001

HSs Healthy subjects, *SSc* subjects with scleroderma, *P1 P* values (GLM) between males and females, *P2 P* values (GLM) between healthy females and SSc

between instrumental measurements and subjects' height and age was analyzed using Pearson's correlation coefficient.

General Linear Model (GLM) was used to assess the extent to which the two repeated measurements differentiated between males and females in the healthy subjects group and between subjects with SSc and healthy females. Before using GLM, we checked the relationship between dependent variables and aging and height. Visual inspection and univariate analysis (Pearson's correlation coefficients, *r*) showed that age was associated with MMO, Tretr, and Tend. Thus, age was entered in the model when calculating differences between male and females for those variables. Height was linearly and weakly ($r < 0.33$) correlated with Tstren, Lstren, and MMO. When building the GLM model for Tstren and Lstren, height was no longer significantly associated with those dependent variables. Thus, we corrected MMO both for age and height. We also checked presence of outliers and distributions of residuals. We found no outliers and residuals were normally distributed with zero mean and a common variance.

7 (4.6 %) HSs showed temporomandibular joint disorders (malocclusion), 4 (2.6 %) HSs showed abnormally short lingual frenulum (ankyloglossia), while none of the SSc subjects showed malocclusion or ankyloglossia. Given the small number of subjects with these disorders we did not take these factors into account in the statistical analysis.

In order to assess the inter-rater reliability, healthy persons were consecutively assessed by two independent raters; to assess reliability at test–retest, two consecutive assessments were collected by the same rater. The ICC coefficient was calculated for each variable along with the standard error of measurements (SEM). The SEM of a test referred to the variability of test scores that would have been obtained from a single person had that person been tested multiple times.

Results

Inter-rater and test–retest reliabilities for the assessed variables proved to be satisfactory (Table 3) in terms of ICC and SEM values except for Tretr.

The MMO values for males and females, and between age group and height are reported in Tables 4, 5 and 6; the GLM (Table 4) revealed significant differences between

Table 5 Normative data: medians and (5th–95th percentiles) of each variable according to HSs' age and height in females

Females							
Age (year)	Height (m)	MMO (mm)	Lstren (N)	Tstren (N)	Tretr (mm)	Tprot (mm)	Tend (mm)
20–40 $n = 30$	≤1.63	41.3 (32.5–51.0)	4.8 (1.6–8.6)	8.0 (5.0–19.0)	33.0 (21.0–37.0)	48.8 (22.8–66.4)	2961.1 (1343.4–5706.1)
	>1.63	45.5 (33.5–54.0)	6.5 (2.0–12.0)	12.5 (6.5–19.0)	34.0 (22.0–41.5)	48.8 (30.4–66.6)	3418.1 (1163.0–4861.4)
41–60 $n = 30$	≤1.63	38.0 (22.5–43.5)	8.5 (2.7–10.6)	11.0 (6.5–19.0)	30.0 (23.5–39.5)	53.4 (36.5–67.8)	3160.6 (762.0–4760.7)
	>1.63	40.5 (35.0–52.0)	7.0 (3.6–9.8)	11.5 (6.0–17.5)	26.5 (21.0–41.5)	45.9 (8.5–69.2)	2842.2 (1512.4–6848.6)
61–81 $n = 21$	≤1.63	44.0 (35.5–47.5)	6.3 (2.4–8.6)	8.5 (4.0–22.0)	23.8 (17.0–45.5)	47.6 (25.0–60.7)	2515.9 (1647.9–4546.9)
	>1.63	38.3 (27.0–47.5)	6.2 (2.7–9.2)	10.0 (5.5–20.0)	27.5 (8.5–44.0)	52.6 (28.0–65.0)	3199.7 (1003.9–5879.8)

Table 6 Normative data: medians and (5th–95th percentiles) of each variable according to HSs' age and height in males

Males							
Age (y)	Height (m)	MMO (mm)	Lstren (N)	Tstren (N)	Tretr (mm)	Tprot (mm)	Tend (mm)
20–40	≤1.75	48.0 (35.0–59.6)	8.6 (3.7–11.3)	14.5 (6.5–23.5)	30.3 (26.5–43.5)	46.1 (32.9–71.2)	3819.5 (1313.8–6160.5)
n = 26	>1.75	48.5 (36.5–56.0)	8.9 (51.8–13.1)	14.5 (1.0–39.5)	34 (22.5–44.0)	47.3 (28.2–64.4)	3839.9 (928.2–6382.2)
41–60	≤1.75	49.3 (33.5–53.5)	9.4 (7.2–12.7)	13.5 (5.5–18.5)	27.8 (12.0–36.0)	49.9 (28.4–60.1)	2869.9 (1366.5–4752.5)
n = 23	>1.75	42.5 (31.0–58.0)	10.7 (9.1–12.9)	16.0 (7.5–20)	27.0 (6.5–42.5)	56.0 (46.2–62.9)	4027.6 (2166.5–5089.6)
61–81	≤1.75	42.5 (28.0–51.0)	8.2 (4.4–9.3)	18.0 (13.5–19.5)	25.8 (24.0–31.0)	40.3 (33.7–56.1)	2556.3 (1954.8–3991.5)
n = 21	>1.75	45.8 (19.5–62.0)	6.7 (0.4–12.1)	13.8 (6.0–24.5)	27.3 (0.0–42.5)	54.0 (36.3–68.7)	2644.8 (1135.2–4277.8)

genders with males showing larger values, also a significant correlation was found for height (Table 7) and MMO. Further, SSc showed statistically significant decrease in MMO with respect to healthy females (Table 4).

A statistically significant gender difference in Lstren was found (Table 4): Lstren was 2.5 N (38 %) higher in males than in females. A correlation between Lstren and height was observed (Table 7). The HS group did not show significantly stronger lips than the SSc group (Table 4) with subjects with SSc showing only a 15 % reduction compared to healthy females.

A significant difference in Tstren was found between males and females: the median Tstren was more than 35 % higher in males than in females (Table 4). Tstren also increased with height but not with age (Table 7). The group comparison showed a significant difference between SSc and healthy subjects with the SSc group being about 23 % weaker than healthy females (Table 4).

The medians of Tretr were similar in males and females (Table 4). Analysis revealed that older subjects had statistically significantly lower Tretr values than younger subjects, however, no correlation with height was observed. Lastly, Tretr did not show statistically significant differences between healthy females and subjects with SSc (Table 4).

No gender-related differences were found for Tprot; moreover, no association between Tprot and height and age was observed (Table 7). The comparison between the SSc group and the HS group revealed a statistically significant difference: Tprot was about 42 % lower in the SSc group compared to healthy females.

Table 7 Pearson's correlation coefficients between instrumented variables and height and age in Healthy Subjects

	Height	Age
MMO	0.29*	−0.14
Lstren	0.33*	0.03
Tstren	0.25*	−0.02
Tretr	0.06	−0.40*
Tprot	−0.05	0.11
Tend	0.10	−0.22*

* $P < 0.05$

Tend correlated with age but no differences were found between males and females. A statistically significant difference was seen between HS and SSc (Table 4): participants with SSc showed a sharp reduction (92 %) of Tend compared to healthy females (177.2 vs. 2515.9 mm).

Discussion

The primary aim of the study was to provide normative data, validity of measurements and estimation of measurement bias of various measurements important for evaluation of mouth function in healthy subjects and subjects with SSc. The secondary aim was to understand how subjects with SSc differ from normative data.

Assessment of Healthy Subjects

In general, the measurement devices showed good test–retest reliability and validity in healthy subjects and some parameters were successful in identifying gender and age differences similar to those already established in the literature.

Maximal Mouth Opening (MMO)

Mouth opening plays an important role in bolus acceptance, consonant articulation, and oral hygiene [2]. Measurement of mouth opening showed good inter-class correlation coefficient values with low SEM values, confirming the reliability of the device in terms of both inter-rater and test-rest reliabilities. In accordance with some studies, the average MMO was 43 mm [25, 40], while other studies report higher values: 47.1 mm in Nepalese subjects [24], 50.77 mm in a French population [38], and 49.10 mm in a Chinese (Taiwanese) population [39]. This variability may reflect the well-documented relationship between MMO and height [38, 40]; indeed, our data showed a positive correlation between height and MMO. Our findings corroborate the existing literature on the difference between genders [38, 39] with a median MMO of 46.2 mm for males and 41.0 mm for females. These data are similar to those obtained by several authors in different populations: 43.0 mm for males and 41.0 mm for females in an Irish population [25] and 45.3 mm for males and 41.5 mm for females in a Jordanian population [40].

Lip Strength (Lstren)

Decreased Lstren may cause difficulties with feeding, eating, speech, facial expressions, and saliva control [41]. Inter-rater and test–retest reliabilities showed good ICC values but high SEM values, and this implies that when assessing an individual subject, a moderate error of measurement must be expected. High SEMs are due to the high standard deviation values, reflecting differences in dentofacial morphology, class occlusion, and oral habits among the subjects [23, 42]. One way to reduce variance, thus improving SEM values, is to improve the assessment procedures: more careful positioning of the device and a more standardized procedure of applying forces may increase the reliability of measurements. In agreement with other studies, we found that males were stronger than females [21, 42, 43], while no linear correlation was observed between Lstren and age [21].

Tongue Protrusion Strength (Tstren)

Tstren assessment is important in the rehabilitation process because of its role in a wide range of activities including speech and bolus manipulation [21]. Inter-rater and test–retest reliabilities proved to be satisfactory, with a SEM value of around 1/7 of the median strength score; thus, small differences in measurements were observed when a subject was tested twice, or by two raters.

No correlation was found between Tstren and age, while males were stronger than females, suggesting that a qualitative and objective assessment of the tongue can reveal differences between genders. Our results are in contrast with those by Clark et al. [21]. They found that Tstren did not differ between males and females, while age group comparisons showed lower strength in the oldest subjects. No other studies were found on Tstren since most of the published reports addressed tongue elevation strength. Although both measurements refer to tongue strength, comparisons between our findings and these studies should be made with caution because of the involvement of different tongue movements. These studies showed controversial results for age, since some investigators have found that tongue strength can be negatively correlated [16, 44, 46], or not correlated [11, 42] with age, depending upon the methods used to measure, experimental setting and sample involved. Similar controversial results were found for gender: Nicosia et al. [10] and Vitorino [45] reported no significant difference between males and females, while Crow et al. [46], Stierwalt et al. [29], and Trawitzki et al. [47] showed decreased tongue strength in females.

Anterior Tongue Retraction (Tretr)

Anterior Tretr provides an important function in velar plosive consonant production, yawning, and the posterior motion of the tongue during swallowing. Unfortunately, we did not find any studies addressing Tretr.

Tretr proved to be satisfactory only for inter-rater reliability.

Conversely, test–retest reliability for this variable was low in terms of ICC and SEM (Table 3). Tretr is rarely voluntarily performed and needs high tongue competence; this leads to difficulty in carrying out the same movement twice, resulting in less repeatable movements. Similar to Tprot, no gender-related difference or height correlation was found. In contrast, Tretr deteriorates with age: males in their seventies showed a 14 % mean reduction of Tretr with respect to thirty-year old subjects.

Tongue Protrusion (Tprot)

Tprot is essential to have good intraoral mobility and range and for a number of complex orofacial behaviors such as licking and mouth clearance [48]. Tprot proved to be reliable since excellent ICC and SEM values were found for inter-rater and test–retest reliabilities. The validity of the measurement is debatable, however, since no correlation between Tprot and age, height or gender was found. This may be due to the fact that Tprot is also a function of tongue competence: precise tongue movement and muscle synergies are necessary in order to achieve maximal protrusion; further studies are necessary to assess the impact of these cofactors on Tprot.

Tongue Endurance (Tend)

Tongue endurance is often seen in clinical assessments, and influences quality of feeding and speaking [49]. Several papers have defined tongue endurance as “the length of time 50 % of maximum pressure can be sustained” [29, 44–46]; in the present study, tongue endurance was assessed as maximum repetition in a given period even if holding the time constant may have incorporated a fatigue component for some, and not for others. Maximum repetition number to measure endurance has been used in other papers [18, 50] and appears closely related to activities such as speaking and swallowing involving repetitive movement of the tongue. Both inter-rater and test–retest reliabilities of measurements with the dial indicator were good. In agreement with findings reported in the literature, Tend was associated with age since older people showed worse performance [49] but not with height or gender [29, 45, 46].

Impact of SSc on Oral Structure and Function

At present, there are few quantitative studies on SSc oral disorders, and those report mostly on mouth opening [51–53]. In a previous descriptive study of ours [26] using self-administered tests, subjects with SSc reported a high prevalence of oral disorders; in the present study, we implemented an objective assessment to quantitatively measure these reported disorders [26]. The sample consisted of women with SSc since in this pathology females represent more than 75 % of the whole population. The instrumented evaluation discriminated between HS and SSc clearly shows the impact of this pathology on oral functions probably due to increased stiffness, vascular changes, and bone resorption [52–54]. We found a 42 % reduction of Tprot with respect to healthy females highlighting the impact of tissue damage on tongue muscles. SSc-related disorders also led to a 23 % reduction of tongue strength and to a 92 % reduction of tongue endurance compared to healthy female. These impairments along with the reduction of tongue mobility may influence quality of speaking and difficulty in the oral preparation of food, especially in chewing hard food and in moving food inside the mouth as reported elsewhere [55, 56].

Besides tongue impairments, a trend toward reduction of Lstren was found. This is an important finding since resorption of the lips, increased rigidity, and weakness are problems frequently reported by subjects with SSc [26, 54]. These impairments may lead to a change in facial expression and appearance [55] and to difficulty in forming the lips to take the food from dining utensils. Finally, confronted with healthy subjects, the subjects with SSc showed a sharp reduction in oral mobility, with 30 % of them having less than 30 mm, which is considered a severe limitation [56] of mouth opening, suggesting that some distresses in activity of daily living reported during clinical evaluation [57] and in self-administered questionnaires [26, 55] may be due to the difficulties in opening of the mouth.

In conclusion, the assessment procedures proved to be valid and reliable. During the oral assessment, subject’s age, height, and gender have to be taken into consideration. Age correlates with mouth opening, Tretr, and endurance; height predicts mouth opening, Lstren, and tongue strength, while gender differentiates between mouth opening, Lstren, and tongue strength.

The comparison between SSc performances and normative data confirmed that the assessment has good discriminant validity and showed important reduction of oral function in SSc.

Limitation

In the future, assessment of sensitivity to change and correlations between clinical and instrumental variables are necessary before the use of these instruments in clinical practice. Measurement of anterior and posterior tongue elevation strength, tongue lateralization, and cheek compression might also prove to be useful in describing oral function.

This is a first attempt to provide quantitative data on oral impairments in persons with SSc. The assessment of a larger group of SSc subjects is needed to further validate the present findings.

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Conflict of interest The authors declare that they have no conflict of interest.

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