



Maximum amplitude of mouth opening in systemic sclerosis

Amplitude máxima da abertura de boca na esclerose sistêmica

Máxima amplitud de abertura de la boca en la esclerosis sistémica

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Abstract

*Systemic sclerosis is a chronic multisystemic disease that is part of the group of systemic autoimmune disorders, rare, progressive, of unknown etiology. It reaches vital organs and perioral tissues, causing hypertonia of the speech organs, face with the appearance of a mask, difficulty in chewing, swallowing disorder and limitation of mouth opening. **Objective:** To measure the maximum amplitude of mouth opening of subjects with systemic sclerosis. **Methods:** A not randomized and controlled, exploratory clinical study. 20 subjects participated (10 of the study group with Systemic Sclerosis and 10 of the control group), of both genders, from the Rheumatology Service of a University Hospital. For the measurement of maximum mouth opening, it was used a digital caliper. It was checked the maximum interincisal distance in the*

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Authors' contributions: SEZMB Substantial contribution to the conception and design of the scientific work, the literature review, the wording participation, critical review of the work and final approval of content to be published. LFA participated in the data collection, methodology, critical review of the work and final approval of the content to be published. CSA Anny Karoline Andrade da AKAS and Luiz Barros Filho LBF participated in the data collection and literature survey. JCM Substantial contributions to the conception and realization of research and critical review of the work - final approval of content to be published. CPHARC critical review of the work - final approval of content to be published.

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Received: 18/09/2015

Accepted: 04/12/2015



front view. In this study, we chose the normalized index with values of 45 mm. The T-Independent test (p of 5%) was applied. **Results:** Most of the sample consisted of female subjects (90%), aged between 23 and 64 years old. The variation in maximum mouth opening was between 24,5 mm and 46,7 mm (an average of 36,7 mm \pm 7,17), while, in the control group, the average was 46,5 mm. **Conclusion:** Significant evidences about the decrease of the maximum amplitude of mouth opening in the studied group when compared with the control group, highlighting the need to more studies in the area.

Keywords: Speech, Language and Hearing Sciences; Rheumatology; Systemic Sclerosis; Measurements; Mouth.

Resumo

Esclerose Sistêmica é uma doença multissistêmica crônica que faz parte do grupo de distúrbios autoimunes sistêmicos, progressiva, rara, de etiologia desconhecida. Atinge órgãos nobres e os tecidos periorais, causando hipertonia dos órgãos fonoarticulatórios, face com aparência de máscara, dificuldade na mastigação, desordem na deglutição e limitação na abertura da boca. **Objetivo:** Mensurar a amplitude máxima de abertura de boca de sujeitos com Esclerose Sistêmica. **Métodos:** Estudo clínico exploratório, não randomizado e controlado. Participaram 20 sujeitos (10 do grupo de estudo com esclerose sistêmica e 10 do grupo controle), de ambos os gêneros, do serviço de Reumatologia de um Hospital Universitário. Para a mensuração da abertura máxima de boca foi utilizado paquímetro digital. Verificou-se a distância interincisiva máxima na visão frontal. Neste estudo, optou-se pelo índice de normalidade com valores de 45 milímetros. Aplicou-se o teste T-Independente (p de 5%). **Resultados:** A maioria da amostra foi composta pelo gênero feminino (90%), com idades entre 23 e 64 anos. No grupo de estudo a variação na abertura máxima de boca ocorreu entre 24,5 e 46,7mm (média: 36,7mm \pm 7,17) enquanto no grupo controle a média foi de 46,5mm. **Conclusão:** Evidências significativas de diminuição da amplitude máxima de abertura de boca no grupo de estudo quando comparado ao controle, evidenciando a necessidade de mais estudos na área.

Palavras-chave: Fonoaudiologia; Reumatologia; Esclerose Sistêmica; Medidas; Boca.

Resumen

Esclerosis Sistémica es una enfermedad crónica multissistémica que hace parte del grupo de trastornos autoinmunes sistémicos, progresivo, raro, de etiología desconocida. Llega a órganos vitales y a los tejidos periorales, causando hipertonia en los órganos fonoarticulatórios, (ta) cara con apariencia de máscara, dificultad en la masticación, desorden en la deglución y limitación en la apertura bucal. **Objetivo:** Medir la amplitud máxima de la apertura bucal de sujetos con Esclerosis Sistémica. **Métodos:** Ensayo clínico exploratorio, no aleatorizado y controlado. Participaron 20 sujetos (10 en el grupo de estudio con esclerosis sistémica y 10 en el grupo control), de ambos sexos, del servicio de Reumatología de un Hospital Universitario. Para la medición de la apertura bucal máxima se utilizó calibrador digital. Se ha encontrado la distancia interincisal máxima en vista frontal. En este estudio, se optó por índice de normalidad con valores de 45 mm. Se aplicó a la prueba T-independiente (p de 5%). **Resultados:** La mayor parte de la muestra fue compuesta por mujeres (90%), con edades comprendidas entre 23 y 64 años. En el grupo de estudio, la variación en la apertura máxima boca fue entre 24,5 y 46,7mm (media: 36,7 \pm 7,17mm), en el grupo control la media fue de 46,5mm. **Conclusión:** Evidencias significativas de la disminución de la amplitud máxima de la apertura bucal en el grupo de estudio en comparación con los controles, lo que sugiere la necesidad de más estudios en el área.

Palabras claves: Fonoaudiología; Reumatología; Esclerosis Sistémica; Medidas; Boca.

Introduction

The Systemic Sclerosis (SS) is a chronic multisystemic disease that is part of the autoimmune systemic disorders group¹. Progressive, rare and of unknown etiology¹. It reaches vital organs and perioral tissues, causing limitation in mouth opening², hypertonia of the speech organs, face with the appearance of a mask¹, difficulty in chewing and swallowing². It is estimated an incidence of three to 19 individuals per million habitants in general population³. It is more common in women (3:1), between 35 and 64 years old, slightly more common in women during productive phase³, being the average age of diagnosis of 50 years. For reasons not clearly defined, collagen deposition occurs in the body⁴, which affects the connective tissue producing changes in the vascular system and has as main characteristic the fibrosis affecting the skin and viscera^{5,6}.

Clinical manifestations of the SS include the Raynaud's phenomenon (RyP)⁷, which is characterized by reversible episodes of vasospasm of extremities, associated with typical color changes that occur after exposure to cold or stress situations, usually occurring in hands and feet and in some more severe cases, in nose, ears or tongues; tenosynovitis, arthralgia progressing to polyarthritis, cutaneous sclerosis, telangiectasia in the face and periunguals, skin calcification, cardiomyopathies, pulmonary disease, renal crisis, gastrointestinal involvement and esophageal abnormalities (with dysphagia and gastroesophageal reflux), which amount to 90% in these patients⁸.

As for orofacial manifestations, there are few reports in the literature^{2,9}, being cited stiffness, skin atrophy and face with the appearance of a mask¹⁰; perioral furrow wrinkling; hardening and loss of elasticity of the oral mucosa, tongue and soft palate stiffening, shortened frenulum of the tongue⁶, pathological resorption of the alveolar ridge of the head of the condyle of the jaw⁸; change in chewing and disorder in swallowing^{2,9}, pneumophonarticulatory incoordination and microstomia, which develops due to collagen deposition in the perioral tissues, causing progressive limitation of mouth opening^{4,10,11,12}, xerostomia and neuralgia of the trigeminal^{13,14}.

In the clinical practice of Speech Language Pathology and Audiology, the evaluation of facial morphologic aspects is justified due to the involve-

ment of orofacial structures in oral functions, being relevant the measurement of anthropometric measures in this region¹⁵, since its presence in the national literature is scarce¹⁶.

The measurement of orofacial structures in Speech Language Pathology and Audiology assessment contributes both to the establishment of therapeutic behavior, and for monitoring the Speech Therapy results¹⁷. Specially, in the analysis of the active interincisal maximum distance, since orofacial miofunctional and cervical disorders can limit the mouth opening, according to the literature.^{18,19}

Limitations in opening the mouth, especially in rheumatic diseases, are consequences of disorders in the temporomandibular joint, which may be a temporary or progressive and degenerative manifestation, showing that early diagnosis, treatment and monitoring can minimize its consequences and incapacities²⁰.

Thus, the interest for this research arose because of the scarcity of national and international Speech Language Pathology and Audiology studies of patients with Systemic Sclerosis especially regarding the limitation in mouth opening, since that, for these individuals, limiting jaw movements can interfere in oral hygiene, speech, nutrition, oropharyngeal inspection and dental treatment and can affect the quality of life of patients^{2,8,21}.

Therefore, the aim of this study was to evaluate the range of maximum mouth opening of individuals with SS.

Methods

Observational exploratory clinical study, cross-sectional, quantitative analytical, non-randomized and controlled, held at the Rheumatology clinic of the University Hospital (HU) and in the Clinical School of Speech Language Pathology and Audiology of the Universidade Federal de Sergipe [Federal University of Sergipe (UFS)] approved by the CAAE 0326.0.107.000-11, following the Resolution 466/2012 of the National Health Council of the Ministry of Health. The study was conducted from March 2012 to November 2014.

The sample was consisted by convenience and was divided into two groups:

- **Study Group** (SG): ten subjects of both genders, aged from 23 to 64 years old, all from the Rheumatology service of the HU/UFS and with

previous medical diagnosis of SS given by a rheumatologist.

- **Control Group (CG):** ten subjects, matched by age and gender with the SG, all companions of patients coming from the HU/UFS Rheumatology service.

The eligibility criteria for the SG were: being an adult or elder, having rheumatologic medical diagnosis of SS and present cognitive capacity that would allow participation in the stages of the study and, of the CG, were: being an adult or elder, healthy and without complaints of pain in the temporomandibular joint and limitation of mandibular movements. The exclusion criteria, for both groups, were the absence of the lower and upper central and lateral incisors, to refuse or give up any of the stages of the research.

SG subjects sought Speech Language Pathology and Audiology care after the referral of a rheumatologist and, when accepting to participate in the study, they signed the Terms of Free and Clarified Consent, as well as the Terms of Authorization of Image Use.

The survey participants were asked to integrate study and, when they accepted, they signed the Terms of Free and Clarified Consent and the Terms of Authorization of Image Use.

The subjects presented disease duration between two and 20 years old (average of: 10.9 years old), being possible the group to be considered heterogeneous, regarding the period of evolution of the condition.

Later, an interview was conducted orally, to investigate whether the subject had the eligibility criteria for SG or CG, and collection of a history of the disease (for the SG), of information about the general health status, presence or absence of Speech Language Pathology and Audiology complaints and other inferences that could affect the life of the subject.

Then, the procedure was carried out to collect the maximum amplitude of mouth opening. Therefore, we asked each participant to remain seated with their feet properly on the floor, with their back straight and head oriented according to the Frankfurt parallel plane to the horizontal plane and the sagittal plane median perpendicular to the horizontal plane. Next, we proceeded with the pachymetry, requesting the maximum mouth opening and, with the aid of a digital caliper (6" Western^R PRO) with resolution and reproduc-

ibility of 0,01 millimeter (mm) (according to the manufacturer's specifications), was measured the distance between the incisal faces of the upper central incisor and lower right, for three times, with the average of obtained values being obtained and the result transcribed in mm.

The maximum opening was measured using a digital caliper. The verification was carried out vertically. The overbite was measured previously and added to the previously obtained aperture value, reaching the maximum interincisal distance. We opted for the normal index for maximal mouth opening of 45 mm, since authors^{22,23} stressed that normal indexes for maximum aperture range from 45 mm to 60 mm for the adult. It is considered, the opening inferior to 40 mm, in adults as a warning to potential muscle or joint problems²².

To increase the reliability of the measures, the measurements were performed by a single examiner, expert and experienced in orofacial motricity, accompanied by a speech therapist and three students of the Speech Language Pathology and Audiology Course/Fellows of the PIBIC Program/COPEs of UFS and FAPITEC / SE. During this procedure, it was always used the same caliper.

The results were tabulated in Excel spreadsheet software (Microsoft[®] Office package) to descriptive analysis. Thus, they calculated the numeric variables as average and standard deviation. After the distribution of normal data was verified, since the sample was small by the Kolmogorov-Smirnov Test and, since the results indicated normality, we applied the T-independent test, considering p value of 5%.

Results

The sample of the SG was composed by 90% female and 10% male, aged between 23 and 64 years old (average: 48.6 years and standard deviation \pm 14.76). The variation of the maximum amplitude of mouth opening varied from 24,5 mm and 46,7 mm (as can be seen in Table 1).

In the control group, 90% of the subjects were female and 10% male, aged between 23 and 62 years old (average: 47.6 years and standard deviation \pm 13.66). There was no statistically significant difference when comparing the age groups ($p = 0.99$). In this group, the variation of the amplitude of mouth opening varied from 45.2 mm to 53.7 mm (see Table 1).

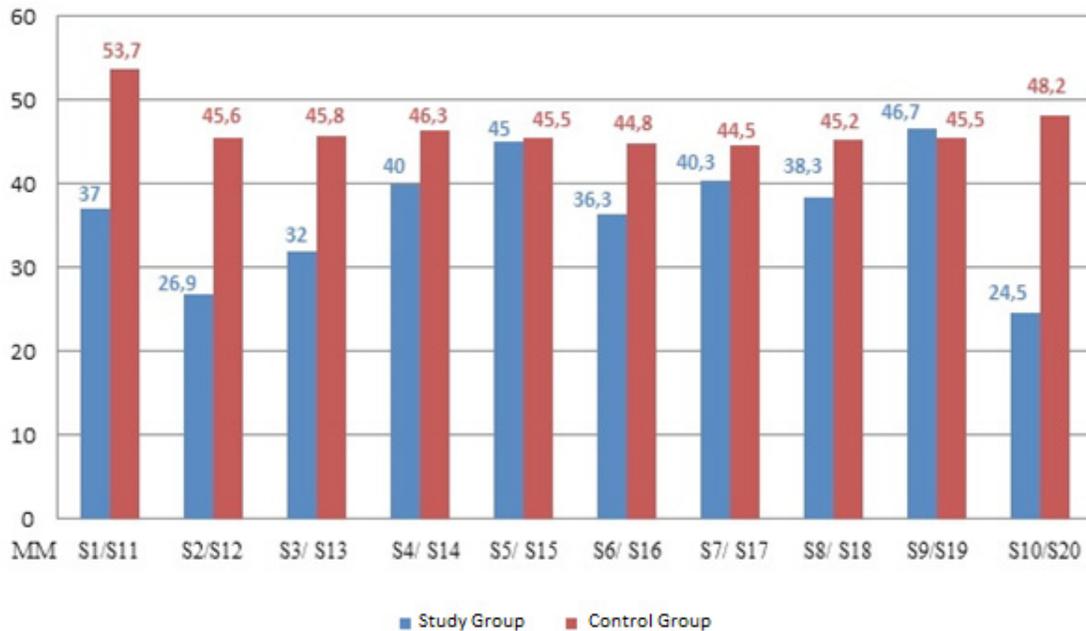
Table 1. Distribution of the study samples regarding gender and age.

STUDY GROUP			CONTROL GROUP		
SUBJECT	GENDER	AGE	SUBJECT	GENDER	AGE
S1	M	23	S11	M	23
S2	F	42	S12	F	43
S3	F	45	S13	F	45
S4	F	51	S14	F	50
S5	F	60	S15	F	59
S6	F	62	S16	F	60
S7	F	64	S17	F	62
S8	F	57	S18	F	53
S9	F	57	S19	F	55
S10	F	25	S20	F	26
AVERAGE:		48,6			47,6
STANDARD DEVIATION:		14,76			13,66

Subtitle: S= Subject, M = Male and F = Female. T-Independent Test. p value of 5%

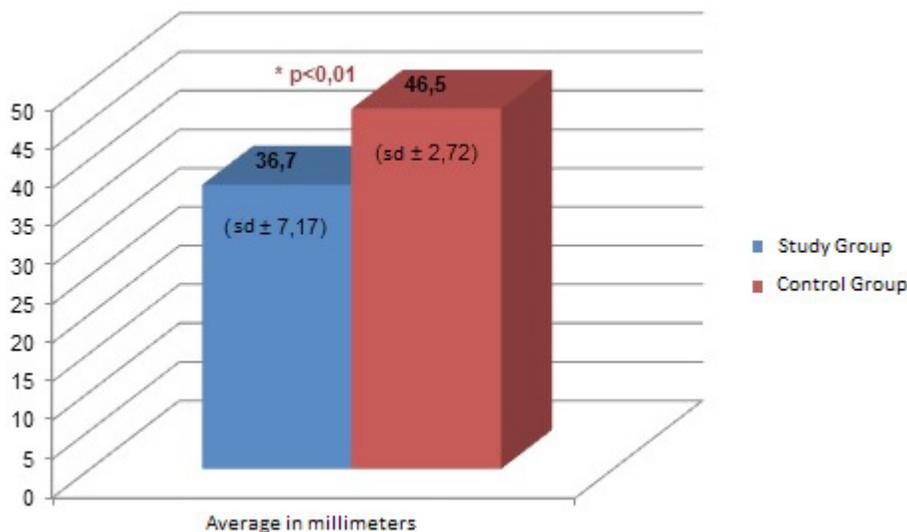
Chart 1 shows the individual values of the maximum amplitude of mouth opening in millimeters of the subjects of the study and control groups, paired by gender and age.

In chart 2, it can be seen the maximum average amplitude of mouth opening between the two groups, showing statistically significant differences.



Subtitle: MM= millimeter e S = subject

Chart 1. Individual values of the maximum amplitude of mouth opening in millimeters of the subjects of the study and control groups, paired by gender and age.



Subtitles: p = p value and sd = standard deviation.

Chart 2- Comparing the values obtained between the control and study groups of the averages of maximum amplitude of mouth opening and standard deviations from the T-independent Test.

DISCUSSION

The Speech Language Pathology and Audiology performance in SS, is a scarce and incipient condition^{2,13,12} and, for being a rheumatic disease, the partnership with Rheumatology becomes essential. Thus, this study sought to investigate the mouth opening, in order to describe the findings because of its importance to the oral functions^{2,4,8,11,12,13,14}.

Although the study sample is small and because it is a rare disease,¹ when faced with small populations derived from rare diseases, as in this work, these samples provide significantly effective evidence and, most of them, are useful analysis²⁴. But, we must be judicious when we use conclusions coming from the statements and inferences of these studies, because of the reduced number of participants.

Some international^{25,26} and national authors^{2,8,13,14} also revealed studies focused on small groups of individuals when it comes to this condition due to its rarity.

Regarding gender, the prevalence was female, corroborating the findings in literature^{3,27}, a ratio of three women affected for every man^{3,13}. Furthermore, the SS is cited as uncommon in men, setting a worse prognosis²⁷, especially under 30 years old³, this data can be seen in Table 1 (subject 1).

The diagnosis usually occurs between 35 and 64 years³, and, in this sample, the subjects (S) had between 23 and 64 years old and two subjects (S1 and S10) were diagnosed at early ages (being S1 with 21 and S10 with 17 - as in table 1), although most of the subjects are in the age range recommended by the literature. The results obtained corroborate with the study¹¹, involving thirty patients with SS, where the majority were female (87%), but with an average age higher than this study (58,6 years).

During the clinical evaluation, it was observed a reduction of the maximum amplitude of the SG mouth opening (average of: 36,7 mm), since this was confirmed by the control group, that showed higher average, approximately with ten millimeters. This limitation is a common finding in cases of SS and possibly is related to the thickening of the skin by chronic accumulation of collagen, characteristic of the disease^{2,11,12,13,14,25,2,28,29}. It can be observed that 80% of the SG showed a maximum mouth opening with less than 40 mm, data regarded as an alert to possible muscle or joint problems^{22,23}. In this population, this restriction is observed by microstomia that occurs due to the collagen deposition in perioral tissues^{28,29}, leading the individual to complain about the difficulty in oral hygiene, speech, nutrition, oropharyngeal inspection and dental treatment and can affect the quality of life

of the patient²¹. This has been confirmed during an interview, by reports of difficulties in performing maneuvers of oral hygiene, both for scleroderma claw in the hands^{8,29}, making it difficult handling the tooth brush to brush the teeth, by the difficulty in opening the mouth, as well as difficulty during chewing^{2,9,13,14,28,29}.

Diverging from the results of the CG, which presented maximum amplitude of mouth opening of 46,5 mm, demonstrating the normal index, only S16 and S17 obtained values below, but close to the normality^{22,23}, equivalent to 20% of the sample of this group. Even when found that the difference in amounts considered normal^{22,23}, it is suggested that more studies should be conducted, with larger populations, for better understanding about these findings.

Studies verifying the implications of this reduction in the SS are cited in most of national and international works, by dentists and/or rheumatologists^{2,4,7,8,9,10,28}, given that this also motivated us to perform this work.

Conclusion

The results showed significant evidence of decreased maximum amplitude of mouth opening in the study group when compared with the control, suggesting the need for evaluation and Speech Language Pathology and Audiology conduct in SS and more studies in the area.

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