Stomatognathic system characteristics in systemic sclerosis: a case report

Características do sistema estomatognático na esclerose sistêmica: relato de caso

Características del sistema estomatognático en la esclerosis sistémica: reporte de un caso

*Sílvia Elaine Zuim de Moraes Baldrighi**
*Anny Karoline Andrade Silva**
*Luiz Barros Filho**
*Leylane Fonseca Almeida**
*José Caetano Macieira**
*Gerlane Karla Bezerra Oliveira Nascimento**
*Carla Patrícia Hernandez Alves Ribeiro César**

Abstract

Systemic sclerosis is a rare disease of unknown etiology and pathogenesis, characterized by an autoimmune process. It often affects the stomatognathic system. ***Objective:*** *To compare clinical and electromyographic evaluation of a subject with systemic sclerosis (S1) and one without rheumatic disease (S2).* ***Methods:*** *Clinical, descriptive exploratory study consisted of two female subjects with 26 years old. The clinical evaluation was performed in the Orofacial Motricity and Physiology Laboratory of the Federal University of Sergipe. We partially used the MBGR protocol. The recording of the electrical activity in the hemifaces by surface electromyography, focusing on chewing, was performed at rest and at maximum intercuspation.* ***Results:*** *The clinical evaluation showed restraint in the subject's mouth opening with S1 rheumatic disease (24,5mm) compared to 49,0mm in S2. It was observed a normal*

*Universidade Federal de Sergipe – UFS – Aracaju –SE – Brazil.

Authors' contributions: SEZMB conception and design of scientific work, literature review, writing participation, critical review of the work and final approval of the content to be published. AKAS and LBF data collection, design and drawing of the scientific work, literature review, writing, critical review and final approval of the content to be published. CPHARC, GKBON and LFA critical review of the work and final approval of the content to be published. JCM design and conduction of the research and critical review, final approval of the content to be published.

Part of this study was presented at the IX Encontro Brasileiro de Motricidade Orofacial (in the form of a banner) in June 2016, in the city of Bauru / SP – 9th Brazilian Orofacil Motricity Encounter.

Correspondence address: Sílvia Elaine Zuim de Moraes Baldrighi - silviazbaldrighi@uol.com.br
Recebido: 08/10/2016
Aprovado: 04/12/2016

posture of parted lips, bony palate with increased depth, reduced width and dental absence. Change in mobility and tone of the speech organs. The masticatory pattern was unilateral, with decreased speed and atypical contraction of the mentual. Swallowing with sharp contraction of the mentual and the presence of residue after swallowing in S1. These changes were not observed in S2. Regarding the electromyographic register, S1 showed a higher electrical activity to accomplish chewing when compared to S2. ***Conclusion:*** *Significant alterations in the clinical evaluation and electric chewing activity related to the stomatognathic system in the subject with systemic sclerosis.*

Keywords: *Speech, Language and Hearing Sciences; Reumatology; Clinical diagnosis; Chewing.*

Resumo

A Esclerose sistêmica é uma afecção rara de etiologia e patogênese desconhecida, caracterizada por um processo autoimune. Frequentemente atinge o sistema estomatognático. ***Objetivo:*** *Comparar avaliação clínica e eletromiográfica de um sujeito com esclerose sistêmica (S1) e outro sem doença reumática (S2).* ***Método:*** *Estudo exploratório clínico, descritivo, composto por dois sujeitos de 26 anos de idade, do gênero feminino. A avaliação clínica foi realizada nos Laboratórios de Motricidade Orofacial e de Fisiologia da Universidade Federal de Sergipe. Utilizou-se parcialmente o Protocolo MBGR. O registro da atividade elétrica nas hemifaces por meio da eletromiografia de superfície, com enfoque na mastigação, foi realizado em repouso e na máxima intercuspidação.* ***Resultados:*** *A avaliação clínica revelou restrição na abertura de boca do sujeito com doença reumática de S1 (24,5mm) comparada com 49,0mm de S2. Observou-se postura habitual de lábios entreabertos, palato ósseo com profundidade aumentada, largura reduzida e ausências dentárias. Alteração na mobilidade e tonicidade dos órgãos fonoarticulatórios. O padrão mastigatório foi unilateral, velocidade diminuída e contração atípica de mentual. Deglutição com acentuada contração de mentual e presença de resíduos após a deglutição em S1. Essas alterações não foram observadas em S2. Quanto ao registro eletromiográfico evidenciou-se em S1 maior atividade elétrica para realizar a mastigação, quando comparado com S2.* ***Conclusão:*** *Evidenciaram-se alterações significativas na avaliação clínica e na atividade elétrica da mastigação relacionadas ao sistema estomatognático no sujeito com esclerose sistêmica.*

Palavras-chave: *Fonoaudiologia; Reumatologia; Diagnóstico clínico; Mastigação*

Resumen

Esclerosis sistémica es una enfermedad rara de etiología y patogénesis desconocida, caracterizada por proceso autoinmune. Con frecuencia afecta el sistema estomatognático. ***Objetivo:*** *Comparar evaluación clínica y electromiográfica de un sujeto con esclerosis sistémica (S1) y uno sin enfermedad reumática (S2).* ***Metodo:*** *Estudo de exploración clínica, descriptivo, compuesto por dos sujetos de 26 años de edad, sexo femenino. La evaluación clínica se realizó en el Laboratorio de Motricidad Orofacial y Fisiología de la Universidad Federal de Sergipe. Utilizó parcialmente protocolo MBGR. El registro de la actividad eléctrica en hemifaces por electromiografía de superficie, centrándose en la masticación, se realizó en reposo y con la máxima intercuspidación.* ***Resultados:*** *Mostró moderación en la apertura de la boca del sujeto con enfermedad reumática S1 (24,5mm) en comparación con 49,0mm de S2. Se observó postura normal de labios entreabiertos, paladar óseo con un aumento de la profundidad, reducción de anchura y ausencia dental. Cambio en la movilidad y el tono de los órganos del habla. El estándar masticatorio era unilateral, disminución de la velocidad y contracción atípico de mentual. La ingestión con fuerte contracción mentual y la presencia de residuo después de tragar en S1. Estos cambios no se observaron en S2. En cuanto el registro electromiográfico S1 mostró una mayor actividad eléctrica para llevar a cabo la masticación, en comparación con S2.* ***Conclusion:*** *Se presentaron cambios significativos en la evaluación clínica y la actividad eléctrica de masticar relacionada con el sistema estomatognático en el sujeto con esclerosis sistémica.*

Palabras clave: *Fonoaudiología; Reumatología; Diagnóstico clínico; Masticación*

Introduction

Systemic sclerosis (SS) is a chronic multisystemic disease that is part of the group of progressive autoimmune disorders of unknown etiology[1,2], rare, with an incidence of two to ten new cases per million inhabitants per year[2]. It is characterized by excessive deposition of collagen in the connective tissue, in addition to thickening of the skin and early compromise of internal organs[2,3]. It usually affects women, in a ratio of 3: 1 when compared to men. It usually begins between the third and fifth decade of life and the average age, at the time of diagnosis, is generally at 50 years[2].

Regarding the impacts related to the stomatognathic system (SS), research is scarce in the literature, but some researchers have revealed alterations involving orofacial motricity, such as restriction of mouth opening[3,4,5,6], face with a mask appearance[7], hypertonia of phonoarticulatory organs, abnormal lip position, inefficient chewing and swallowing disorder[8,9].

The studies in orofacial motricity have recommended the clinical evaluation, being important the introduction of objective evaluation, with the use of surface electromyography (SEMG). SEMG is a non-invasive and easy to perform method. Nowadays, professionals of several health areas use this resource in the diagnosis and orientation of the therapeutic conducts. Regarding the functioning of the stomatognathic system, the use of SEMG is one of the objectives to aid in the clinical diagnosis[10] and, when associated with other clinical methods, allows a better understanding of the involvement of the mandibular muscles, contributing to the treatment of this system[11,12,13].

Due to the scarcity of data on the work of Speech, Language and Hearing Sciences in the field of orofacial motricity in SS cases, and the concern to deepen the knowledge about this condition, the objective of this study was to compare clinical evaluation and electrical activity during chewing between one case with SS and another without rheumatic disease.

Methods

This is a clinical exploratory, descriptive, consistent and fulfilling the ethical principles of autonomy, non-maleficence, beneficence and justice of the resolution 466/12 of the National Health Council study. Approved by the Research Ethics Committee of the Federal University of Sergipe (UFS) under the number CAAE 36835814.1.0000.5546. It was carried out in the Grupo de Estudo de Motricidade Orofacial - Group of Study of Orofacial Motricity (GEMO) – and in the Laboratory of Physiology of UFS in partnership with the Rheumatology ambulatory of the University Hospital of UFS.

During the data collection, the subjects who accepted to participate in this study were informed about the research procedures through information letters and then signed the Free and Informed Consent Term, as well as the Authorization Term for the Use of Image / Filming. The evaluations took place on separate days for each subject from September to December of 2015.

Two female, being one (S1) of 26 years old (study), with previous clinical diagnosis of diffuse systemic sclerosis and the other subject (S2), with 26 years old (control), without rheumatic disease, participated in this study.

The inclusion criteria for S1 were diagnosis confirmed by a rheumatologist of SS, to be an adult and present cognitive ability that allowed the participation of the subject in all stages of the study. For S2, regarding the inclusion criteria, it was necessary to be a female adult without rheumatic disease and without complaints of changes related to the stomatognathic system.

The exclusion criteria, for both subjects, were: undergoing Speech, Language and Hearing Sciences treatment, restriction in diet to the food selected for electromyographic evaluation, being on medications that may interfere in the Central Nervous System and refusing to participate in the study during any of its steps.

The process of Speech, Language and Hearing Sciences and electromyographic diagnosis was performed in different stages. Firstly, the initial interview was previously elaborated on personal records, previous history of the disease, medications used, general state impairments, Speech, Language and Hearing Sciences complaints and other inferences about the subject (S1). And for S2, an initial interview was also conducted to verify if the subject met the inclusion / exclusion criteria of the research. The instrument was applied by the examiners (orally) and the data recorded on their own spredsheet. Some previous and current

medical examinations were consulted for a better understanding of the clinical picture of S1.

Description of S1- female, 26 years old, self-declared white, from the city of Maceió, Alagoas, married, without children, university student with diagnosis of Systemic Sclerosis (SS) and denied a family history on the disease.

Description of S2 - female, 26 years old, self-declared white, native of the state of Sergipe, single, without diagnosis of rheumatic disease.

Chart 1 shows the clinical history data of S1 up to the moment of her entry in the University Hospital / UFS for clinical evaluation and, in Chart 2, the clinical and Speech, Language and Hearing Sciences complaints brought by S1 at the time of the anamnesis:

Chart 1. Characterization of S1 (Subject 1) according to the time, disease evolution and beginning of the Speech, Language and Hearing Sciences intervention

Subject	Gender	Current Age	Age at the beginning of the disease	Evolution period and medical diagnosis	Speech, Language and Hearing Sciences Intervention
S1	Female	26 years old	Between 14 and 15 years old	Around 2 years	Eleven years after the diagnosis

Chart 2. Clinical and Speech, Language and Hearing Sciences manifestations reported by S1 during anamnesis. Complaints present at the beginning of symptoms

Subject	Clinical Manifestations	Speech, Language and Hearing Sciences Signs and symptoms
S1	• Thick and hardened skin • Stiffening of the hands, sclerodermic claws. • White spots on hands and elbows • Wounds on the skin and fingertips • Weight loss • Thin face, especially lips and nose.	• Frequent choking (For all consistencies) • Cough • Respiratory tiredness • Difficulty in mouth opening

After the initial interview, the Speech, Language and Hearing Sciences clinical evaluation started, focusing on the aspects of orofacial motricity constituted by observation and analysis of the stomatognathic system.

During the clinical evaluation, the MBGR protocol[14] was partially used. The results of the evaluation of S1 and S2 from the MBGR protocol are described in a comparative way as follows: anatomophosphological aspects and posture / position condition in Chart 3, mobility and muscle tonus in Chart 4 and oral functions in Chart 5. It should be noted that some items of the protocol were not applied, justified by the adaptation of the essential components of this instrument to be used in the condition investigated.

Chart 3. Aspect and postural condition / position of the phonoarticulatory organs of Subject 1 (S1) compared to subject 2 (S2)

		S1	S2
Anatomophysiological aspects and postures	Usual lip position	Parted lips	Closed
	Mucosa of the tongue	Marked by teeth on R and L sides	Normal
	Hard palate	Increased depth and reduced width	Normal Depth and Width
Teeth and occlusion	Dental failure	Present	Absent
	Dental and gingival conservation	Regular	Good
	Bacterial plaque	Present	Absent
	Occlusion	Altered	Apparently Normal

Subtitle: R= Right, L= Left

Chart 4. Mobility and muscle tonus of phonoarticulatory organs of S1 (Subject 1) compared to S2 (Subject 2)

		S1	S2
Mobility of Lips	Snap	Not performed	Normal
	Closed Forward	Not performed	Normal
Mobility of tongue	Forward	Tried to perform	Normal
	Snap	Tried to perform	Normal
Mobility of cheeks	Inflate the right/left side	Not performed	Normal
Mandibular movement	Mouth opening	24,5 mm	49,0mm
Muscle Tonus	Superior Lip	Increased	Normal
	Inferior Lip	Increased	Normal
	Mentual	Increased	Normal
	Tongue	Increased	Normal
	Cheek: right and left	Increased	Normal
Palpation	Palpation pain	Absent	Absent

Subtitle: mm = millimeters

Chart 5. Results of the oral functions of S1 (Subject 1) and S2 (Subject 2)

Oral Functions		S1	S2
Breathing	Type	Average/superior	Average/superior
	Mode	Nasal	Nasal
Chewing	Incision	Anterior	Anterior
	Chewing Pattern	Unilateral Left	Bilateral alternate
	Lip Closure	Unsystematic	Systematic
	Speed	Decreased	Adequate
	Atypical Muscle Contractions	Mentual	Absent
Liquid and solid Swallowing	Tongue posture	Not seen	Not seen
	Inferior Lip Posture	Contact with the superior (unsystematic)	Contact with the superior
	Mentual Muscle contraction	Accented	Absent
	Coordination	Adequate	Adequate
	Solid food residue after swallowing	Present	Absent
	Compensatory movements	Anteriorization of the head	Absent

The materials used during the evaluation, both for the clinical examination and for evaluation of the oral functions were: disposable gloves, spatulas and disposable cups, Mikatos[R] stethoscope (adult Ref.162), focus flashlight, digital caliper (6" Western[R] PRO) with resolution and reproducibility of 0.01millimeters (mm) (according to manufacturer's specifications). For better observation of oral functions, they were recorded using the Sony DSC-W730 camera, and the video samples were analyzed later. Liquid (water) and solid (bread) consistencies were used to evaluate the oral functions of chewing and swallowing, and cervical auscultation was performed. During the evaluation of the maximum width of the mouth opening, each participant was asked to remain seated, with their feet properly supported on the floor with erect spine and the head oriented with the Frankfurt parallel plane to the horizontal plane and the median sagittal plane perpendicular to the horizontal plane. Then, the patient proceeded with pachymetry, requesting a maximum mouth opening and, with the aid of the digital caliper, the distance between the incisal faces of the upper and lower right central incisors was measured, and the result was transcribed in millimeters[15].

Regarding speech, no change was observed in the phonoarticulatory production of the participants. Only S1 presented limitation of mandibular movements during speech, without impairment to intelligibility.

After the clinical evaluation, the subjects were submitted to the electromyographic evaluation through the electromyograph, which is an apparatus capable of recording the action potentials that occur through the voluntary activation of the muscle or the response to electrical stimulation. In Speech, Language and Hearing Sciences the use of SEMG is recent and it aims to aid in the diagnosis and therapy of oral motor, swallowing, chewing and speech disorders[16].

The device used was the biofeedback, J & J Engineering, of specification I-330-C2 + Hardware Guide, with four channels, which evaluates some parameters such as heart rate, temperature and the electromyography of any muscle. In this study, it was chosen to evaluate the masseter muscle. For this purpose, the 2223BRQ/3M[R] electrode was used for short or long term procedures. According to the manufacturer, this brings the desired accessibility to excellent performances of adhesiveness and integrity in the skin of the patient.

Electrical activity was recorded, individually, with subjects sitting in a chair with their backs resting on the backrest, eyes opened, parallel feet resting on the floor and arms resting on the lower limbs. Afterwards, the skin was cleaned for placing the electrodes with cotton soaked in 70 ° alcohol, to improve the conduction of the action potentials and to reduce the impedance of the system, and, only then, the electrodes were placed on the skin that covers the masseter muscles[16].

The electrodes were applied at the moment of palpation, after noticing greater muscle contraction (ideal point for electrode placement), since, at this point, the neuromuscular junction site is the best area for the nerve impulses to be captured during maximal intercuspation[17]. The placement of the electrodes followed the longitudinal direction of the muscle bundles; to avoid possible interferences of the musculature, these were fixed bilaterally, at the point of greatest muscular contraction, and arranged longitudinally to the muscular fibers. After this step, the reference electrode was placed, or the ground, to avoid interferences in the signal capture[16,17].

The analysis of the electrical activity was performed in the left and right hemifaces during rest (ten seconds), at the maximum habitual intercuspation (three seconds) and in chewing the food (recording the time necessary to chew the whole food)[17] . These records can be visualized in Chart 6 - record of the gross electrical signal result in microvolts (μV), Root Mean Square (RMS), positive value, and in Chart 7 - the analysis can be observed after the normalization of the signal.

Chart 6. Values obtained from the gross electric signal record, measured in microvolts (μV), of S1 (Subject 1) and S2 (Subject 2) with raisins and bread

	Side	Type of Food	Masseter	Tightening 1	Tightening 2	Tightening 3	Chewing
S1	Left	Raisin	24,63993263	56,77857208	59,75972748	60,80763245	29,62975502
	Right	Raisin	9,997159958	38,36635971	34,03851318	35,76332474	14,85827351
	Left	Bread	24,51255989	61,79139328	62,66057968	50,84383392	31,57767105
	Right	Bread	9,490726471	33,25403595	35,4628334	34,69395065	13,96433353
S2	Left	Raisin	1,392133713	101,5520859	104,5873566	90,56510162	9,637273788
	Right	Raisin	1,483022094	138,4146271	143,0903778	134,9872131	12,30749702
	Left	Bread	1,60427928	115,2615433	113,2061234	88,80243683	14,36058235
	Right	Bread	1,521294	130,8016	133,097	173,0283	19,3931

Chart 7. Root Mean Square calculation after normalization of the electromyographic signal of S1 (Subject 1) and S2 (Subject 2)

	Bread		Raisin	
	S1 %	S2 %	S1 %	S2 %
Right Masseter	40,5	13,31	41,2	8,87
Left Masseter	54,05	13,58	50,12	9,75

Some foods can be used to evaluate electrical activity; such as 25g of bread, a quarter of an apple, a stuffed type biscuit, or a teaspoon of roasted peeled peanuts[17]. For the present study, the use of bread was chosen because it is a food with greater consumption in the general population, being thus more easily accepted. The amount of bread used in this research was 25g[17]. In addition to three raisins that were chewed at one time, as proposed by the literature[16].

For the normalization of the electromyographic signal, the RMS calculation was done by the quadrature of each data point, sum of the squares, dividing the sum by the number of observations, and taking the square root. This technique is used for the rectification of RAW, in which all numbers will be converted to positive values, instead of positive and negative values[18]. For normalization, two decimal places were rounded.

Discussion

Progressive systemic sclerosis is a chronic multisystemic disease that is part of the group of systemic autoimmune disorders; progressive, rare, of unknown etiology[1]. It affects both the skin and the noble organs, reaching the cardiovascular system, lungs, gastrointestinal tract and kidneys[19]. It is characterized clinically by vascular involvement, fibrotic alterations of the skin and internal organs[1].

As to the stomatognathic system, there are few reports in the literature, especially in Speech, Language and Hearing Sciences studies[6,8], but it is observed in these individuals difficulties mainly in the oral functions of chewing and swallowing, restriction of mouth opening[20] and alteration in muscle tonus and posture of phonoarticulatory organs[6,8,9,21].

Speech, Language and Hearing Sciences actuation in SS is a scarce and incipient condition[6,8], as it can be observed in Chart 1, since S1 only started the Speech, Language and Hearing Sciences treatment eleven years, after the medical diagnosis of SS given by the rheumatologist. Therefore, it is reinforced that the partnership with Rheumatology becomes essential in this condition[8,9].

This study aimed to compare the clinical evaluation and electric chewing activity of a subject with SS with another, without rheumatic disease and without complaints of changes in the stomatognathic system, in order to better describe and understand these findings, as well as to enable future interdisciplinary actions.

Although the sample studied is only a case report, the rarity[22] of this disease, as well as its extensive clinical heterogeneity, make the findings of this study relevant, because when faced with small

populations derived from rare diseases, these samples offer significantly efficacious evidence and, for the most part, are useful analyzes[23]. However, when we use conclusions from these statements, we must be careful and cautious, especially in case studies.

The incidence of SS is of two to ten new cases per million inhabitants per year[2,23] and studies show that this varies according to the geographic location and ethnicity[24]. The prevalence is higher in women at a ratio of 3: 1, usually at ages of 35-64 years old. It is rare in children and in men under 30 years of age[25].

It is estimated that less than 10% of patients develop the disease before the age of twenty years old[26,27]. As we can see in Chart 1, S1 began to notice the first signs and symptoms between the ages of fourteen and fifteen, making her case even rarer, corroborating the literature[26,27].

Regarding the initial signs and symptoms of the disease (Chart 2), some authors[7,22] have cited that in the face, skin thickening leads to orofacial manifestations including stiffness, skin atrophy, loss of expression with diminished lips and nose tapering[7]. These typical changes occur in the perioral region, causing a reduced oral opening and a "mausekopf" face (mouse face), with loss of facial wrinkles[7,20,22]. It was observed that, at the beginning of the S1 disease, the patient presented thick and hard skin, a thin face, mainly related to lips and nose, which confirms the findings of the consulted literature[7,22].

The patient was diagnosed with diffuse SS (DSS). In this classification, individuals present hand edema, gradual skin thickening or arthritis, which are usually the first manifestations of the disease[7]. Sclerotic changes are also observed in the fingers and hands where contracture flexures produce shortened, claw-like, fingers[22]. Changes in skin pigmentation may occur both during the inflammatory phase of the disease and in the phase of progressive fibrosis. Depending on the patient's natural skin tone, this can become hypo or hyperpigmented (appearance of "salt and pepper"). Areas of depigmentation arise particularly in the scalp, chest and upper back and on areas of pressure, such as the back of the hands or pre-tibial regions[7]. These changes were observed only in S1. As mentioned previously, S1 presents DSS, which, in addition to affecting hands, feet and internal organs, can also affect the face (phonoarticulate organs) as occurred and, consequently, caused the limitation of the mouth opening[3] , as seen in Chart 3. These complications can be justified by the exaggerated accumulation of collagen in several organs that lead to fibrosis and cause functional disorders in the involved organs[28].

In this condition, pulmonary disease can occur in about 80% of patients. Pulmonary involvement is second in frequency, soon after esophageal involvement and is currently the main determinant of the risk of death in this pathology. Patients with pulmonary disease have, therefore, a poorer prognosis. Interstitial lung disease is initially manifested by dry cough and fatigue[7]. S1 also reported respiratory tiredness (Chart 2) and was diagnosed with pulmonary fibrosis, evidencing this finding mentioned in the literature[7].

Regarding the Speech, Language and Hearing Sciences examination, when the studied subjects were compared, S1 presented alterations (Chart 3) related to the mobility, tonicity and habitual posture of the phonoarticulatory organs, while S2 did not present them.

The tonicity, mobility and posture of the phonoarticulatory organs become significant during oral functions, that is, they should not be observed only in isolation[29]. Although S1 was able to perform some phonoarticulatory organs mobility activities, there was evidence of muscular stiffness of the cheeks and mentual, important not only at rest, but also during the chewing function. It was also observed absence of lip seal in S1. Such characteristics are not expected in healthy subjects, as observed in S2. This change in the habitual posture of the lips in S1 can be explained by the alteration in the performance of the function of each muscle, most probably due to the thickening of the skin and the loss of the common elasticity in the cases of SS[7].

Changes in the aspects related to the mobility, tonicity of the phonoarticulatory organs and in the usual resting posture of the lips were cited in the literature[6,8,9,21], in patients with SS. Possibly, these alterations occur due to the decrease in the function of the musculature of the face, indicating an increase in stiffness, which may be contributing in reducing the amplitude of the mandibular movements and may also cause impairment in stomatognathic functions. The explanation for this is possibly related to the chronic deposition of collagen, which makes the skin progressively thick, with loss of elasticity, limiting facial movements, and may lead to orofacial manifestations as observed in the

case studied; among them, there is the restriction in mouth opening frequently mentioned in the literature, in the case of individuals with SS [6,7,8,9,20,22].

Regarding the dental situation, the findings related to the alterations found during the clinical evaluation process, revealed, in S1, absence of dental elements and presence of bacterial plaque[7]. These changes can be explained by poor hygiene, bacterial plaque accumulation and limitation of mouth opening[9]. The difficulty of handling the toothbrush occurs due to sclerotic changes in the fingers and hands[22], and it was observed that the condition of the gums was not complete. It is believed that this alteration occurs due to the resorption of the teeth caused by the chronic deposition of collagen[22], as well as due to the accumulation of bacterial plaque mentioned above.

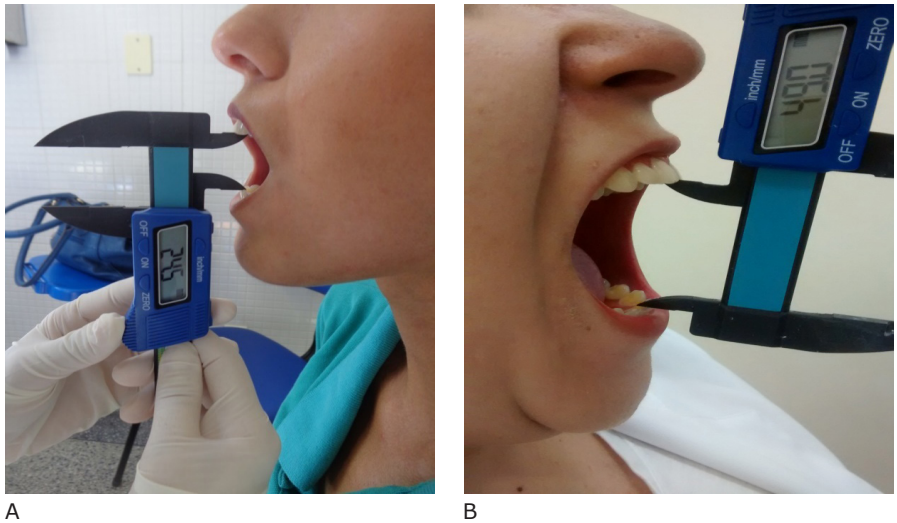
Normality indexes for maximum mouth opening[15] vary between 45 and 60mm for the adults[15], considering the opening of less than 40mm in the adult as an alert for possible muscular or joint problems. In SS, the restriction in mouth opening mobility was cited in some national and international articles developed by physicians or dentists, focused on SS[4,5,20,22] and by Speech, Language and Hearing Sciences pathologists[6,8,9].

Regarding mouth opening, S1 presented restriction, evidencing 24.5 mm; when compared to S2, which presented 49.0mm (Figures 1A and 1B). This limitation is common in SS cases and possibly related to thickening of the skin by the chronic accumulation of collagen, characteristic of the disease[4,5,6,7,9,20].

In the oral functions (Chart 5), it was observed at the time of the evaluation that breathing, both in S1 and S2, is within the normal (nasal) mode [29,30], except for the lack of lip sealing S1, but at the time of the evaluation, the patient used the nose to breathe, a condition that probably occurs due to stiffness in the orofacial musculature, as well as by thickening of the skin, which hinders the closure of the lips. But this condition does not prevent the nose from exercising its function[7].

According to the literature[30], bilateral chewing alternated with occluded lips is indicated as a mature chewing pattern and acts on the development, growth and stability of the dental arches due to balanced action of the orofacial muscles and the pressure exerted on the teeth. For proper chewing, favorable anatomical conditions are required to promote uniform distribution of masticatory forces in the tissues supporting the teeth.

The chewing function was altered in S1 when compared to S2, which exhibited a bilateral masticatory pattern considered normal [30]. When asked to eat bread, during chewing, S1 presented anterior incision, inefficient grinding, atypical muscle contractions, decreased speed and unilateral chewing pattern to the left side. Studies[8,9,21] revealed that these alterations may be related to the limitation of



A B

Figure 1. Restriction of mouth opening of the Subject 1 (24,5mm) (A) compared to the normal value of Subject 2 (49,0mm) (B)

mandibular movements, as well as to the impaired dental situation, common in this condition, such as the presence of periodontal disease and absence of teeth[22]. This fact can also occur due to the stiffening of the skin, characteristic of the disease, and due to the chronic deposition of collagen, which can lead to the alteration in the movements and structures involved in this act[7,22].

During swallowing, for both liquid and solid, partial lip closure was observed without the presence of the tongue between the teeth, posture of the lower lip in contact with the upper one, with adequate containment of the food, but with a marked contraction of the mentual muscle, of the cervical musculature and with anteriorized head position. At the moment of the evaluation, during the presentation of the solid and liquid consistencies, no change was observed in the coordination between swallowing and breathing, presence of residuals after swallowing and rigidity of the larynx during the performance of this function, since this can also occur due to the stiffening of the musculature[7]. Although the patient complained of swallowing and coughing (Chart 2), these signs were not observed during the clinical evaluation. In SS, low dysphagia[7,9] can occur, since the involvement of the digestive tract is the most frequent visceral manifestation of SS, affecting 70 to 90% of patients.

The main objective of the work with orofacial motricity in Speech, Language and Hearing Sciences is to balance the stomatognathic system, rehabilitating and adapting the functions of breathing, suction, swallowing, chewing and speaking, as well as posture and deleterious habits[11]. Nowadays, technological development allows the use of precision measuring instruments and clinical use, among which SEMG[11,13]. This examination analyzes, in an objective and meticulous way, the muscular activity that allows to complement the evaluation of the stomatognathic system in the Speech, Language and Hearing Sciences clinic[11,12].

Regarding the comparison of the electric signal, after normalization of the signal, it was concluded that S1 presented electrical potential greater than S2.

Analyzing Chart 2, during the chewing of raisins, S2, regarding the right masseter (RM), an activity equivalent to 8.87% was presented and, regarding the left masseter (LM), the activity was equivalent to 9.75% of the maximum muscle power. For chewing bread, the RM presented 13.3% and LM: 13.58% of activity when compared to the maximum muscle power in this subject.

The values found in the electrical analysis of S1 revealed, for chewing raisins, that RM presented 41.2%, and LM 50.12%, while for bread RM presented 40.5%, and ME 54.05% of electric activity of the maximum power.

Therefore, S1 presented greater electrical potential when compared to S2. Since this is a single individual research, and due to not having obtained another study that analyzed the electrical activity in individuals with SS, this fact makes it difficult to discuss the comparison among the studied subjects, but it is believed that S1 required a higher electric charge of the musculature to perform the chewing function, probably due to the increased stiffness of the musculature found in SS[7,22], necessitating the use of a greater grouping of muscular fibers when compared to the healthy subject, inferring greater effort to perform chewing.

Conclusion

This work revealed significant changes in the stomatognathic system related to postural aspects, mobility and muscle tonus of the phonoarticulatory organs, restriction of mouth opening, chewing, swallowing and electrical activity of the masseter muscle during chewing of the subject with systemic sclerosis, when compared to the subject without rheumatic disease. Therefore, it demonstrates how essential a speech therapist in the interdisciplinary team is, as well as the need for more studies with larger samples to deepen the knowledge and improve the behavior in these cases.

References

1. Imboden JB, Helmann DB, Stone JH. Esclerodermia. "In" Hummers LK, Wigley FM. Current: Diagnóstico e Tratamento Reumatológico. 3ªed. Porto Alegre: Editora Artmed. 2014. p.228-37.

2. Mayes MD, Lacey JV Jr., Beebe-Dimmer J, Gillespie BW, Cooper B, Laing TJ, et al. Prevalence, incidence, survival, and disease characteristics of systemic sclerosis in large US population. Arthritis Rheum.Aug.2003; 48(8): 2246-55.

3. Matarese G, Isola G, AliBrandi A,Gullo AL, Bagnato G, Cordasco G, et al. Oclusal and MRI characterizations in systemic sclerosis patients: A prospective study from Southern Italian cohort. Joint Bone Spine.Jan, 2016; 83(1): 57-62.

4. Vincent C, Agard C, Barberot S, N'Guyen JM, Planchon B, Durant C. et al.. Orofacial manifestations of systemic sclerosis: A study of 30 consecutive patients. Rev. Stomatol. Chir. Maxillofac.2010; 111(3):128-34.

5. Yuen HK, Marlow NM, Reed SG, Mahoney S, Summerlin LM, Leite R, et al. Effect of orofacial exercises on oral in adults with systemic sclerosis. Disabil.Rehabil. Sep. 2012; 34(1): 84-9.

6. Baldrighi SEZM, Almeida LF, Alves CS, Silva AKA, Barros-Filho L, Macieira JC, et al. Amplitude Maáxima da abertura de boca na esclerose sistêmica. Distúrb. Comum. Marc 2016; 28(1): 162-70.

7. Magro PCFM. Esclerodermia: Revisão de literatura e caracterização da população observada na consulta de Reumatologia do Centro Hospitalar Cova da Beira. [Dissertação de mestrado em Medicina]. Portugal: Universidade da Beira do Interior Faculdade de Ciências da Saúde, 2009.

8. Baldrighi SEZM, Almeida LF, Lima MC, César CPHAR, Macieira JC. Impacto da intervenção fonoaudiológica na Esclerose Sistêmica. Distúrb. Comum. Set. 2014; 26(3): 596-605.

9. Almeida LF, Lima MC, Macieira JC, César CPHAR, Baldrighi SEZM. Intervenção Fonoaudiológica na esclerose sistêmica: relato de casos. Rev.CEFAC. 2016, 18(1): 273-85.

10. Carvalho ARR. Eletromiografia dos músculos masséteres e supra-hióideos em mulheres com oclusão normal e com má oclusão classe I de Angle durante a fase oral da deglutição. [Dissertação]. São Paulo. Faculdade de Medicina. Universidade de São Paulo, 2002.

11. Oncins MC, Freire RMAC, Marchesan IQ. Mastigação: análise pela eletromiografia e eletrognatografia. Seu uso na clínica fonoaudiológica. Distúrb. Comum. Ago. 2006; 18(2): 155-65.

12. Park S, Shin WS. Differences in eating and masticatory performances by gender and obesity status. Physiology & Behavior. 2015; 138:69-74.

13. Prates LS, Gois M, Berwig LC, Blanco-Dutra AP, Busanello-Stella AR, Silva AMT. Avaliação clínica e eletromiográfica da mastigação nos diferentes padrões de crescimento facial. Rev. CEFAC]. Aug. 2016; 18(1): 104-12.

14. Genaro KF, Berretin-Felix G, Rehder MIBC, Marchesan IQ. Avaliação Miofuncional Orofacial: Protocolo MBGR. Rev CEFAC. Abr/Jun. 2009; 11(2): 237-55

15. Bianchini EMG. Relações das disfunções da articulação temporomandibular com a articulação da fala. Rev Dental Press Ortodon Ortop Facial. Jan/Fev. 2000; 5(1): 51-9.

16. Rahal A, Goffi-Gomez MVS. Estudo eletromiográfico do músculo masseter durante o apertamento dentário e mastigação habitual em adultos com oclusão dentária normal. Rev Soc Bras Fonoaudiol.2009; 14(2): 160-4.

17. Nascimento GKBO, et al. Protocolo de avaliação eletromiógrafica em mastigação. "In": SILVA, H.J. Protocolos de eletromiografia de superfície em Fonoaudiologia.1ª ed. Barueri: Pró-Fono,2013.p.15-25.

18. Marchetti PH, Duarte M. Instrumentação em eletromiografia. Laboratório de Biofísica, Escola de Educação Física e Esportes da Universidade de São Paulo USP, São Paulo, 2006. Disponível em: http://lob.iv.fapesp.br/ . Acesso em: 17 abr. 2016.

19. Godoi ERAM, et al. Envolvimento macrovascular e esclerose sistêmica. J. Vasc. Bras., Recife, 2009. Disponível em: http://www.scielo.br/pdf/jvb/2009nahead/0509.pdf . Acesso em: 27 fev. 2016.

20. Salem B, Rim BHA, Shini BR,Maher B. Manifestations bucco-dentaires de la sclérodermie systémique. Pan African Medical Journal. 2013;16(114):3065.

21. Baldrighi SEZMB, et al. Esclerodermia Sistêmica: relato de caso. Revista Extensão & Sociedade. Rio Grande do Norte, 2011; 2(3). Disponível em: <http;//www.periodicos.ufrn.br/ojs/index.php/extensaoesociedade>. Acesso em: 18 mai. 2016.

22. Pereira MCMC, et al. Esclerodermia sistêmica: relato de caso clínico. Revista de Odontologia da Universidade Cidade de São Paulo, São Paulo, 2009; 21(1): 69-73. Disponível em: http://arquivos.cruzeirodosuleducacional.edu.br/principal/old/revista_odontologia/pdf/janeiro_abril_2009/Unicid_21(1)_69_73.pdf . Acesso em: 04 mai. 2016.

23. Chifflot H, et al. Incidence and prevalence of systemic sclerosis: a systematic literature review. PubMed, Bethesda MD. Fev. 2008; 37(4): 223-35.

24. Gu YS. et al. The immunobiology of systemic sclerosis. Semin Arthritis Rheum, Elsevier, Filadélfia. Out. 2008; 38(2): 132-60.

25. Collier MD. Esclerose Sistêmica. "In": WEST, S.G. Segredos em Reumatologia: respostas necessárias ao dia a dia em rounds, na clínica, em exames orais e escritos. 1ªed. Porto Alegre: Artmed; 2001.p.160-8.

26. Martini G. et al. Systemic Sclerosis in Childhood. Arthritis Rheum, 2006; 54(12): 3971-8.

27. Monticielo AO, et al. Esclerose sistêmica e níveis séricos elevados de organoclorado: uma associação possível?. Rev. Bras. Reumatol.2008; 48(1): 51-4.

28. Junqueira LCU, Carneiro J. Histologia básica. 10ª. ed. Rio de Janeiro: Guanabara Koogan; 2004. p.165-75.

29. Marchesan IQ. Deglutição: diagnóstico e possibilidades terapêuticas. "In": Marchesan IQ. Fundamentos em Fonoaudiologia: aspectos clínicos da motricidade oral. 2ª. ed. Rio de Janeiro: Guanabara Koogan; 2005.p.51-8.

30. Cattoni DM, Tessitore A. Diagnóstico das alterações de respiração, mastigação e deglutição. "In": Fernandes FDM, Mendes BCA, Navas ALPGP. Tratado de Fonoaudiologia. 2ª. ed. São Paulo: Roca, 2009.p.140-5.