

The rehabilitation of facial involvement in systemic sclerosis: efficacy of the combination of connective tissue massage, Kabat's technique and kinesitherapy: a randomized controlled trial

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Abstract In Systemic Sclerosis (SSc), face involvement causes functional loss as well as aesthetic changes and loss of the self-image. The aim of the work is to evaluate the efficacy of a rehabilitation program based on the combination of Kabat's technique, connective massage and kinesitherapy specifically conceived for the face of SSc patients. Forty SSc patients were enrolled: 20 patients (interventional group) were treated for 9 weeks (twice a week, 1 h per session) with a combined connective tissue massage, Kabat's technique, kinesitherapy and home exercise program, and 20 patients (control group) were assigned only home exercise program. All patients were assessed at baseline (T0), at the end of the treatment (T1) and after 9 weeks of follow-up (T2). They were evaluated with SF-36, HAQ, modified Rodnan skin score, mouth opening in centimeters and Mouth Handicap in Systemic Sclerosis (MHSS) scale. At T1, both groups improved in mouth opening ($P < 0.05$), but the improvement was maintained at T2 only in interventional group. In interventional group, facial skin score

ameliorated at T1 and maintained at T2 ($P < 0.05$ vs. T0), while no change was observed in controls. In both groups, SF-36 and HAQ were not affected by the treatment. MHSS scale improved significantly in interventional group at T1 ($P < 0.001$), while no change was found in controls. The combination of connective tissue massage, Kabat's technique, kinesitherapy and home-based exercises is more effective than a home exercise program alone in the rehabilitative treatment of SSc facial involvement.

Keywords Systemic sclerosis · Rehabilitation · Connective tissue massage · Kabat method · Physiotherapy

Introduction

Systemic Sclerosis (SSc) is characterized by induration of the skin and internal organs, by joint modifications and muscle impairment [1–5]. Skin fibrosis leads to tissue retraction and atrophy, and consequently to disability and reduction of patients' quality of life (QoL).

Though most authors agree that specific SSc rehabilitation program can prevent and reduce disabilities deriving from skin and joint involvement [2], only few studies on this matter are available [6–9]. The involvement of face and oral tissues is a typical feature of SSc patients that causes important esthetic changes and impairment of the self-image [10]. The face becomes amimic, the cutaneous furrows disappear. Around the mouth, vertical wrinkling, due to skin retraction, develops and the nose becomes sharp.

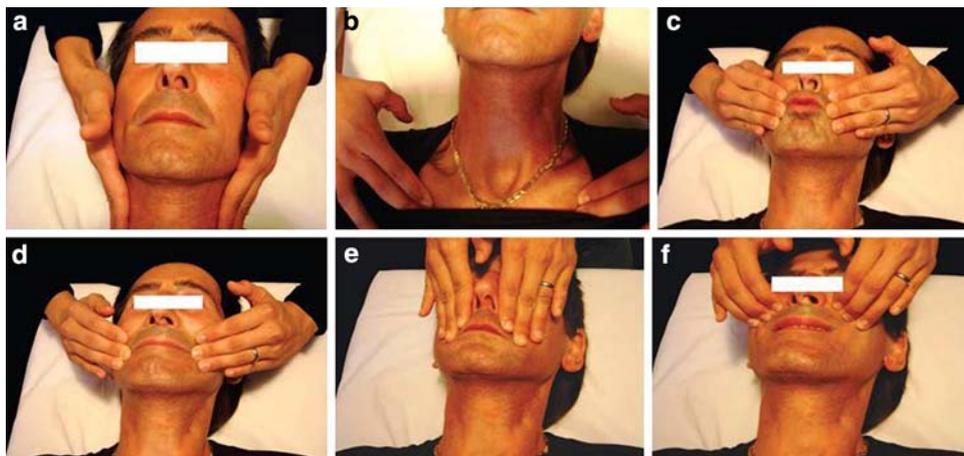
Face changes also include thinning and reduction of mouth width (microcheilia) and opening (microstomia), also favoured by osteolysis of mandibular angles and by fibrosis of soft tissues. These modifications interfere with eating, speaking, oral hygiene measures and dental

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Fig. 1 Physiotherapeutic intervention with connective tissue massage of the neck (a) and clavicular region (b). Kabat's technique for the involvement of the orbicularis oris (c) zygomaticus (d) levator labii (e) and nasalis (f) muscles



treatment, thus deteriorating the quality of life of SSc patients [6, 11–15] and in most severe cases leading to surgical interventions, such as bilateral commissurotomies [6, 11–15].

Rehabilitation management of microstomia is mainly based on self-administered home-based exercise program including mouth-stretching and oral augmentation exercises. This traditional approach has been shown to have some positive effect on mouth opening in SSc patients [6]. Preliminary results from our group suggested that the combination of connective tissue massage, Kabat's technique and a specific kinesitherapy program [8] can be usefully used for the treatment of facial involvement in SSc.

The aim of our work was to evaluate in SSc patients the efficacy of a rehabilitation program for the face based on the sequential combination of connective tissue massage, Kabat's technique, kinesitherapy and traditional home-based exercise program of mimic exercises versus home-based exercise program of mimic exercises alone.

Materials and methods

Forty SSc patients, fulfilling ACR criteria [16] (6 men and 34 women; age: 57.28 ± 11.33 years; disease duration 9.4 ± 4.3 years), were enrolled from the outpatient clinic of the Department of Biomedicine—Division of Rheumatology, of the University of Florence. After a written informed consent, all patients underwent a clinical examination and were assessed according to international guidelines [17]. Entry criterion was face involvement assessed by a Rodnan skin score ≥ 1 [18], no gender or age limits.

The presence of secondary Sjögren syndrome was evaluated by the determination of SSa/Ro and SSb/La autoantibodies, salivary scintigraphy, salivary minor glands biopsy and the ophthalmologic tests including Schirmer e Lissamine green test ().

Patients were assigned, using a random number sequence, to two groups and assessed at baseline (T0), after 9 weeks of treatment (T1) and after 9 weeks of follow-up (T2).

The 20 patients of interventional group were treated for 9 weeks twice a week (each session lasting 1 h) with a combined four-step procedure of connective tissue massage [19–21] during the first 10 min, Kabat's method [21, 22] for about 15 min and kinesitherapy for the following 15 min. Due to the stressful work of this combination of physiotherapy, the remaining 20 min was employed to perform relaxing exercise. All the patients applied also a home program of mimic exercises [6, 12, 23]. Control group (20 patients) performed for 9 weeks a program of daily home exercises for the face only [6, 12, 23].

In both groups, the home program of mimic exercises was continued after the assessment at the end of the treatment (T1) for further 9 weeks till the end of follow-up (T2), for a total duration of 18 weeks.

None of the patients was edentulous, and none presented mandibular osteolysis at X-rays.

All patients continued their pharmacological treatments (alprostadil- α -cyclodextran, calcium channel blockers, topical glyceryl trinitrate, proton pump inhibitors, clebopride, steroids, cyclophosphamide, azathioprine and methotrexate) with no changes throughout the study.

Rehabilitation techniques

Connective tissue massage is a manual technique used to treat altered connective tissues, in order to increase local blood flow and the release of involved tissue by connective tissue stretching [19–21]. For the facial involvement of SSc, it is strictly recommended to treat not only the face but also the neck (Fig. 1a) and the clavicular regions. (Fig. 1b).

Kabat's method is a neurorehabilitation technique that uses spiral and diagonal movement patterns in conjunction with stretch, resistance and other proprioceptive facilitation

Fig. 2 Physiotherapeutic intervention with Kabat's method for the involvement of buccinators (a) frontalis (b, c) and corrugators (d) muscles. Kinesitherapy for the improvement of mouth opening (e) and jaw lateralizing (f)

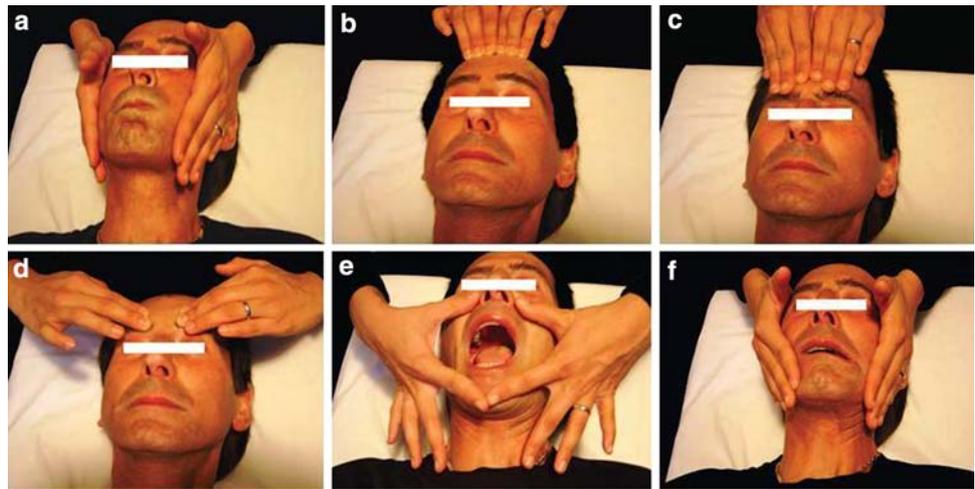
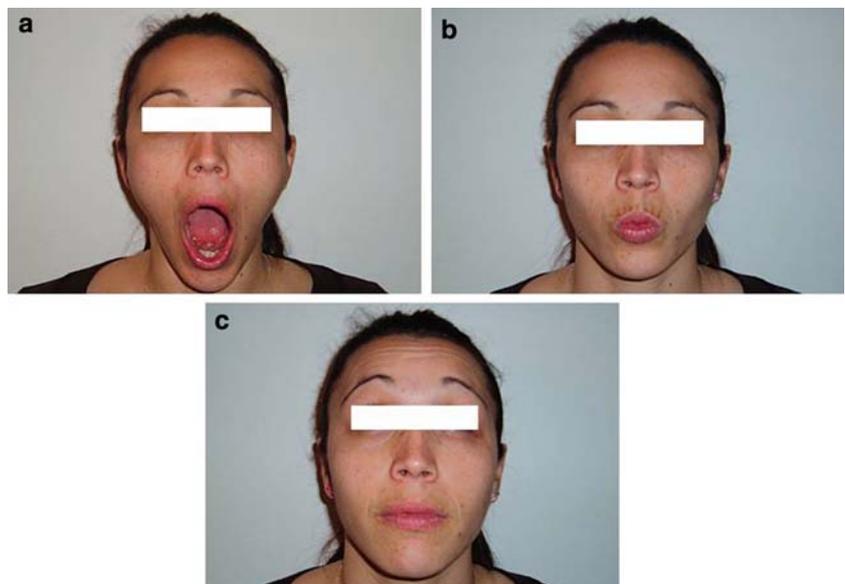


Fig. 3 Program of home daily exercises: mouth stretching (a) and mimic exercises (b, c)



techniques to reinforce neuromuscular recruitment [21, 22]. The Kabat's method concerns the stimulation of the orbicularis oris (Fig. 1c), zygomaticus (Fig. 1d), levator labii (Fig. 1e), nasalis (Fig. 1f) buccinator (Fig. 2a), frontalis (Fig. 2b,c) and corrugator (Fig. 2d) muscles.

Kinesitherapy consisted in specific passive, active and assisted exercises for temporal-mandibular joint (exercises for improving mouth opening and jaw lateralizing) [6, 12]. (Fig. 2e,f).

Program of home daily exercises

All patients were instructed by the same operator (G.L.) to perform daily of mouth stretching and opening exercises [6, 12] and grimacing exercises for mimic muscles [23] (Fig. 3a–c). Home exercises were divided into three sequences. In the first sequence (to be executed at least 5 min, 3 times/day), the patient placed thumbs into the

mouth in order to enlarge the oral angles. The movements were done bilaterally and simultaneously (modified from 6). In the second exercise sequence (to be performed once a day), the patient inserted a number (as high as possible) of tongue depressors between the premolars of one arch toward the molars of the contralateral one to properly open the mouth. The tongue depressors should be maintained for a minimum of 8 min and, after a stop, the exercise should be repeated, if possible, for additional 8 min, by increasing the number of tongue depressors (modified from 6 to 12). The third exercise sequence consisted in mimic exercises to be performed once a day, based on a series of grimaces to exercise (oro)facial muscles [23].

Assessment

Patients were assessed by the same operator at baseline (T0), at the end of the 9 weeks period of treatment (T1) and

Table 1 Patient characteristics

Patient characteristics	SSc patients	Interventional group	Control group	Pvalue
Number	40	20	20	
Sex (M/F)	6/34	2/18	4/16	NS
Age (years)	57.27 ± 11.33	57.20 ± 10.23	57.35 ± 12.60	NS
Disease duration (years)	9.4 ± 43	9.7 ± 42	9.1 ± 46	NS
Active ulcers	11	6	5	NS
Raynaud	40	20	20	NS
Esophageal involvement	13	5	5	NS
Muscle-skeletal involvement	16	8	8	NS
Lung involvement				
ILD	21	11	10	NS
PAH	15		8	NS
Sjogren syndrome	25	13	12	NS
MSI (SF-36)	39.01 ± 9.12	37.19 ± 8.03	40.79 ± 10.35	NS
PSI (SF-36)	37.30 ± 8.12	38.15 ± 7.89	36.50 ± 8.39	NS
HAQ-DI	0.40 ± 0.53	0.50 ± 0/2	0.31 ± 0.34	NS
Mouth opening (on)	3.90 ± 1.07	3.80 ± 1.06	4.00 ± 1.09	NS
Facial skin score	3.72 ± 1.49	3.90 ± 1.55	3.55 ± 1.43	NS
MHISS	17.65 ± 5.25	17.20 ± 5.15	18.10 ± 5.36	NS

ILD interstitial lung disease, PAH pulmonary hypertension, MSI mental synthetic index, PSI physical synthetic index, HAQ-DI health assessment questionnaire disease index, MHISS mouth handicap in systemic sclerosis scale

after 9 weeks of follow-up (T2). The following outcome measures were chosen:

Assessment of global health condition

Health Assessment Questionnaire disability index (HAQ-DI) [24, 25] was used to measure disability; the Physical Synthetic Index (PSI) and the Mental Synthetic Index (MSI) of SF36 were used to assess QoL [10, 26, 27].

Face assessment

The functional involvement of face and mouth was assessed by the Mouth Handicap in Systemic Sclerosis (MHISS) scale [28, 29].

Mouth opening was assessed in centimeters measuring the distance between the tips of upper and lower right incisive teeth (mean of two consecutive measurements).

Skin involvement of the face was assessed by means of a modified Rodnan skin score [18] to be applied to two areas of the face (right and left cheek).

Statistical analysis

Data are presented as mean ± standard deviation. Student's *T*-test or Mann–Whitney tests, when necessary, were used to compare group characteristics at T0. ANOVA for repeated measures was used for all outcome measures. Data

analysis was performed using the SPSS statistical package 12.0 for Windows.

Clinical trial registration

The study was registered with Current Controlled Trials (<http://www.controlled-trials.com>) no ISRCTN43633950.

Results

The general features of the samples are reported in Table 1. No patient dropped out from the study.

Assessment of global health condition

SF36 and HAQ

In both groups, neither the combined treatment nor the home exercise program modified the Physical and Mental Summary Indexes of SF36 and HAQ-DI, as well as at the end of the study (T1) and at follow-up (T2) (Tables 2, 3).

Face assessment

MHISS scale

In interventional group, the combined treatment significantly improved the scores of MHISS scale at T1 versus T0

Table 2 Mean and SD of the items assessed at baseline (T0), at the end of treatment (T1) and after 9 weeks of at follow-up (T2) in interventional group

	T0	T1	T2	T0–T1	T0–T2
	Mean ± SD	Mean ± SD	Mean ± SD	<i>P</i>	<i>P</i>
MSI (SF-36)	37.19 ± 8.03	41.01 ± 7.09	39.22 ± 6.38	NS	NS
PSI (SF-36)	38.15 ± 7.89	39.58 ± 7.84	41.99 ± 6.97	NS	NS
HAQ-DI	0.50 ± 0.72	0.24 ± 0.29	0.16 ± 0.09	NS	NS
Mouth opening (cm)	3.80 ± 1.06	4.28 ± 0.99	4.58 ± 1.16	<i>P</i> < 0.05	<i>P</i> < 0.001
Skin score	3.90 ± 1.55	1.60 ± 0.99	1.75 ± 1.02	<i>P</i> < 0.001	<i>P</i> < 0.001
MHISS	17.20 ± 5.15	16.25 ± 5.64	18.50 ± 5.23	<i>P</i> < 0.001	NS

MSI mental synthetic index, PSI physical synthetic index, HAQ-DI health assessment questionnaire disease index, MHISS mouth handicap in systemic sclerosis scale

Table 3 Mean and SD of the items assessed at baseline (T0), at the end of treatment (T1) and after 9 weeks of at follow-up (T2) in control group

	T0	T1	T2	T0–T1	T0–T2
	Mean ± SD	Mean ± SD	Mean ± SD	<i>P</i>	<i>P</i>
MSI (SF-36)	40.79 ± 10.35	40.98 ± 7.96	41.93 ± 11.42	NS	NS
PSI (SF-36)	36.50 ± 8.39	35.81 ± 7.87	37.52 ± 8.35	NS	NS
HAQ-DI	0.31 ± 0.34	0.33 ± 0.31	0.26 ± 0.20	NS	NS
Mouth opening (cm)	4.00 ± 1.09	4.48 ± 1.04	4.20 ± 1.05	<i>P</i> < 0.001	NS
Skin score	3.55 ± 1.43	3.15 ± 1.63	3.35 ± 1.18	NS	NS
MHISS	18.10 ± 5.36	18.00 ± 4.97	17.90 ± 4.03	NS	NS

MSI mental synthetic index, PSI physical synthetic index, HAQ-DI health assessment questionnaire disease index, MHISS mouth handicap in systemic sclerosis scale

(16.25 ± 5.64 vs. 17.20 ± 5.15; *P* < 0.001). At T2, MHISS scores (18.50 ± 5.23) worsened both versus scores obtained at T1 and at T0. This indicates that the home-based program did not maintain the effect obtained by the combined program during the previous 9 weeks.

In control group, the home exercise program did not provide any significant change in MHISS throughout the study (Tables 2, 3).

Mouth opening

Patients of both groups benefited from the treatments for mouth opening. In interventional group, a significant increase of mouth opening was shown at T1 and still maintained at T2 (*P* < 0.05 and *P* < 0.001 vs. T0, respectively).

In control group, the home exercise program improved mouth opening at T1 (*P* < 0.001 vs. T0), but the significance was lost at T2 (Tables 2, 3).

Skin score

In interventional group, the combined treatment decreased significantly skin score at T1 (*P* < 0.001 vs. T0). The decrease of the skin score was still significant at T2

(*P* < 0.01 vs. T0). In controls, the home exercise program did not modify skin score throughout the study (Tables 2, 3).

Discussion

Our data show that, in SSc, a combined rehabilitation approach is significantly more effective than a home exercise program in reducing skin thickness of the face, in recovering mouth opening and in improving self-reported face and mouth-related symptoms. It is, however, clear that the treatment of the face did not exert any effect on global function and QoL.

SSc leads to fibrosis of facial soft tissues with reduction of mimics and disappearance of the cutaneous furrows. These features, together with diminished mouth opening and width, altered dentition, difficulties in dental intervention, concomitance of sicca syndrome and changes at temporo-mandibular joints often lead to impairment of QoL. In fact, the fibrotic involvement of hands and face becomes one of the SSc patients' greatest complaints [30, 31], with concern about disfigurement [32] and personal self-esteem [33]. It was also shown that,

in SSc, skin deformities are a core stressor of the disease, only preceded by fatigue [33].

In order to deal with the facial involvement of SSc patients, we have chosen to use the combination of connective tissue massage (able to modify, by stretching, blood-stream and releasing the connective tissues) and Kabat's method (a neurorehabilitation technique that reinforces neuromuscular recruitment) [26, 27]. These techniques were added to the traditional kinesitherapy and home exercises for mouth and mimic muscles. At the best of our knowledge, this is the first study combining these techniques in a rehabilitation program specifically conceived for the face of SSc patients.

It is interesting to note that the improvement of mouth opening, the only effect of home exercises, was lost at follow-up. Contrarily, the effects of combined protocol on mouth opening, skin score and self-perceived functional mouth and face handicap were maintained after 9 weeks from the end of the study.

This can be due to the efficacy of the different techniques acting synergically and to the fact that patients continued, for a 9-week period after the end of the treatment, the program of mimic and stretching exercises in a home self-management program.

This evidence suggests that the continuity of care is mandatory in the rehabilitation of patients affected with chronic rheumatic diseases, such as SSc, in order to maintain its efficacy [34].

Home exercises for orofacial muscles were of some efficacy. In fact, in the short-term period, they were able to increase mouth opening in the group of SSc patients that were assigned to the home program only.

In the chronic care model, patients are encouraged to become part of the health care team, and the concept of self-care or self-management plays a central role [35]. Some evidence exists about the effectiveness of self-management in SSc patients. Recently, Mugii et al. studied the efficacy of self-administered stretching of each finger in SSc patients, showing that amelioration of range of motion was present in each finger after 1 month and maintained within 1 year [36].

The amelioration of MHISS scale suggests that, in SSc, the combined approach improved also the subjective perceptions and the self-perceived disability due to face and mouth involvement. This result is important because, although problems related to mouth involvement seem to have less weight than hand-related problems in total disability, mouth problems are felt as an important handicap by SSc patients. MHISS is a new scale with an excellent reliability and good construct validity, in the specific assessment of disability involving the mouth in SSc patients [22]. In our study, MHISS was helpful in following up SSc patients over time for evaluating the outcome due to rehabilitation.

From our data, it is evident that the treatment of the face had no effects on QoL and disability. This is probably due to the relatively short period of the treatment and to the fact that the patients were treated with techniques not aimed at the overall body, but specifically conceived for the involvement of the face.

Due to the complexity and the severity of SSc, the rehabilitation program proposed in the present work should be regarded as a support tool in SSc management. However, this combined program should be integrated with global rehabilitation techniques on top of the pharmacological treatments for the disease [4, 5].

The limitations of our study may be considered the short period of treatment and follow-up that did not allow us to identify how long the effect of the combined treatment could last after the end of the study.

In conclusion, the combination of Kabat's technique, connective tissue massage and kinesitherapy with home-based exercises is effective in the rehabilitative treatment of SSc face. This combination treatment may lead to benefits for mouth opening and skin score, maintained also after the end of the program and is more effective than a simple home exercise program. Further studies are needed to evaluate the long-term effects of this combined program also in the two SSc subsets.

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