EXTENDED REPORT

# Development and validation of a scale for mouth handicap in systemic sclerosis: the Mouth Handicap in Systemic Sclerosis scale

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*Ann Rheum Dis* 2007;**66**:1651–1655. doi: 10.1136/ard.2007.070532

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Accepted 30 April 2007
**Published Online First 9 May 2007**

**Objective:** To develop and assess the reliability and construct validity of a scale assessing disability involving the mouth in systemic sclerosis (SSc).

**Methods:** We generated a 34-item provisional scale from mailed responses of patients (n = 74), expert consensus (n = 10) and literature analysis. A total of 71 other SSc patients were recruited. The test–retest reliability was assessed using the intraclass coefficient correlation and divergent validity using the Spearman correlation coefficient. Factor analysis followed by varimax rotation was performed to assess the factorial structure of the scale.

**Results:** The item reduction process retained 12 items with 5 levels of answers (total score range 0–48). The mean total score of the scale was 20.3 (SD 9.7). The test–retest reliability was 0.96. Divergent validity was confirmed for global disability (Health Assessment Questionnaire (HAQ), r = 0.33), hand function (Cochin Hand Function Scale, r = 0.37), inter-incisor distance (r = −0.34), handicap (McMaster-Toronto Arthritis questionnaire (MACTAR), r = 0.24), depression (Hospital Anxiety and Depression (HAD); HADd, r = 0.26) and anxiety (HADa, r = 0.17). Factor analysis extracted 3 factors with eigenvalues of 4.26, 1.76 and 1.47, explaining 63% of the variance. These 3 factors could be clinically characterised. The first factor (5 items) represents handicap induced by the reduction in mouth opening, the second (5 items) handicap induced by sicca syndrome and the third (2 items) aesthetic concerns.

**Conclusion:** We propose a new scale, the Mouth Handicap in Systemic Sclerosis (MHISS) scale, which has excellent reliability and good construct validity, and assesses specifically disability involving the mouth in patients with SSc.

Systemic sclerosis (SSc) is a connective-tissue disease characterised by excessive collagen deposition and by vascular hyper-reactivity and obliterative microvascular phenomena.[1 2] SSc is responsible for skin, tendon, joint and vessel damage, which leads to handicap.[3] Therefore, outcome measures with good metric properties assessing handicap are needed to assess disease evolution and treatment efficacy in SSc.

Global disability in SSc patients is usually measured by the Health Assessment Questionnaire (HAQ), which correlates well with the extent of skin thickening, loss of ability to close the fist, proximal muscle weakness and tendon friction rubbing, but not digital ulcers.[4 5] Thus, Steen and Medsger proposed the use of the scleroderma HAQ (sHAQ), a more disease-specific disability scale.[6] Five patient-generated visual analogue scales were added to the original HAQ, assessing Raynaud's phenomenon, digital tip ulcers, gastro-intestinal and lung symptoms and overall disease severity from the patient's perspective.[6] Recent assessment has seen the use of more location-specific disability scales in SSc, such as the Cochin Hand Function Scale (CHFS).[7 8] These scales are useful to clinicians in assessing the impact of certain locations of the disease and the efficacy of treatment. For example, hand disability assessed by the CHFS has been shown to contribute to 75% of the HAQ variance, highlighting the need to specifically assess hand disability in SSc when evaluating treatments.[7]

The mouth and face are frequently involved in SSc; patients often complain of aesthetic concerns with skin sclerosis and telangiectasia, diminished mouth opening, altered dentition, difficulties in undergoing dental surgery and/or sicca syndrome. However, no specific assessment tool is available to quantify the handicap associated with mouth disability in SSc patients.

Therefore, we decided to develop and validate a specific mouth handicap scale for use in SSc.

## PATIENTS AND METHODS

### Provisional scale elaboration

One hundred patients followed at the Internal Medicine Department of Cochin Hospital, Paris, were asked by mail to indicate the main situations of daily living (eating, speaking, relationship with relatives) that affected them because of mouth involvement. Seventy-four patients responded. Also, 10 experts were contacted and asked by mail to indicate the main situations of daily living (eating, speaking, relationship with relatives) that patients had complaints about because of mouth involvement. Experts were also asked to define the main domains these items represented. A literature search of published scales concerning the mouth (from PubMed, Embase, and Cochrane library databases) was performed according to EULAR recommendations.[9] A 34-item provisional scale was generated. All items selected were covered by patient answers. No additional items were generated from expert interviews. Although several scales concerning the mouth have been proposed for an oral care setting,[10 11] no relevant item was

**Abbreviations:** CHFS, Cochin Hand Function Scale; dSSc, diffuse systemic sclerosis; HAD, Hospital Anxiety and Depression; HAQ, Health Assessment Questionnaire; ICC, intraclass coefficient correlation; lSSc, limited systemic sclerosis; MACTAR, McMaster-Toronto Arthritis; MHISS, Mouth Handicap in Systemic Sclerosis; SSc, systemic sclerosis

selected from these scales. All experts assumed that handicap induced by mouth involvement in SSc would be due to mouth-opening restriction, mouth dryness and aesthetic concerns.

## Testing the provisional scale

### Patients

To be eligible for the study, patients had to fulfil the American College of Rheumatology criteria[12] and/or the Leroy & Medsger criteria[13] for SSc. SSc patients were classified according to the extent of skin involvement: limited SSc (lSSc), with no detectable skin involvement;[13] limited cutaneous SSc (lcSSc), with skin involvement essentially limited to the hands and face;[13] and diffuse SSc (dSSc), with skin proximal involvement proximal to the elbows and/or knees.[14] All patients were assessed by the same operator (LM) within 48 h (during spring 2005, temperature 20°C) during the annual meeting of the Association des Sclérodermiques de France (ASF), the French SSc patients association. Parameters recorded were age; sex; ethnicity; occupation; sick leave; year of onset of Raynaud's syndrome; age at diagnosis; year of onset of the first non-Raynaud's phenomenon; disease duration; disease form (lSSc, lcSSc or dSSc); body mass index; Karnofsky index score; inter-incisor distance (measured in millimeters); dyspnoea (assessed by the New York Heart Association 4-point scale); pitting scars; digital ulcers; calcinosis; esophagus, joint and/or muscle involvement; heart involvement; interstitial lung disease; pulmonary arterial hypertension; and renal crisis. Evidence of esophagus, joint and/or muscle involvement; heart involvement; interstitial lung disease; pulmonary arterial hypertension; and scleroderma renal crisis was based on patient report.

### Item reduction

The item reduction process retained 12 items with 5 levels of answers (range 0–48) (Appendix 1). Ten items were eliminated because the answer distribution was badly skewed (floor effect), 6 for a lack of reliability (intraclass correlation coefficient (ICC)$<$0.8), 5 because of high correlation with one or several other items (Spearman correlation coefficient (r)$>$0.7), and 5 because of more than one of the above criteria. The final scale was named Mouth Handicap in Systemic Sclerosis (MHISS).

## Testing the final scale

### Other outcome measures used to assess construct validity of the MHISS (see Statistical analysis)

Global disability was assessed using the HAQ,[15] the scale ranging from 0 (no disability) to 3 (maximal disability). The HAQ comprises 20 items divided into 8 domains. Hand disability was evaluated using the CHFS,[16] a questionnaire with 18 items concerning daily activities, each question scored on a scale of 0 (performed without difficulty) to 5 (impossible to do), which is administered by the physician. The total score was obtained by adding the scores of all items (range 0–90). This questionnaire has also erroneously been called the Duruoz Hand Index.[8]

Patients' perceived disability was assessed using the McMaster-Toronto Arthritis questionnaire (MACTAR).[17] Patients were asked to select the 3 situations among activities of daily living that caused them maximal trouble. Each item is scored on an 11-point semiquantitative scale (0–10). The global score ranges from 0 (no disability) to 30 (maximal disability).

Anxiety and depression were assessed using the Hospital Anxiety and Depression scale (HADa and HADd).[18] This scale has 7 questions for each dimension and ranges from 0 (no depression, no anxiety) to 21 (maximal depression, maximal anxiety).

## Statistical analysis

Data analysis involved use of Systat 9. Quantitative variables were described with means, standard deviations (SD) and ranges. Qualitative variables were described with proportions and percentages. The test–retest reliability was assessed with the ICC.

Construct validity was investigated in 2 ways. Divergent validity was assessed by correlating the MHISS scores with scores on variables known to assess dimensions or concepts differing from that assessed by the MHISS. Thus, for example, we hypothesised that MHISS scores would have a weak or fair correlation with global (HAQ) and hand (CHFS) disability, patient's perceived disability (MACTAR), anxiety and depression (HAD), and inter-incisor distance. Because a normal distribution could not be demonstrated for all the parameters studied, we used the nonparametric Spearman r to assess the correlation between 2 quantitative variables, interpreted as excellent ($>$0.91), good (0.90–0.71), moderate (0.70–0.51), fair (0.50–0.31) or little or absent ($<$0.30).[19] Principal-component analysis was used to extract factors. Retained factors had eigenvalues $>$1. Eigenvalues are values obtained by matrix algebra. They represent the part of the whole variation of the data that can be attributed to each factor and indicate the importance of the different factors extracted. Then, independent factors were obtained using the varimax rotation method, an orthogonal rotation method applied to the initial factorial solution, to minimise the number of variables with high loading in each factor and therefore simplify the clinical interpretation of the factors. Factors extracted using this approach were compared with the a priori stratification proposed by experts. Finally, ANOVA was used to assess the weight of mouth handicap assessed with the MHISS and hand disability assessed by CHFS in global disability assessed with the HAQ. Non-parametric tests (Kruskall–Wallis) were used to compare the MHISS total score and subscores according to type of SSc (limited or limited cutaneous vs diffuse), mouth-opening disability (never–occasionally vs often–always) and presence (yes/no) of telangiectasia.

# RESULTS

## Demographic and clinical data (table 1)

Ninety-eight patients from the ASF meeting were asked to participate, and 71 (61 (83.6%) females) agreed. All patients were Caucasian. The mean age at the time of evaluation was 57.6 (11) years and mean disease duration 13.7 (12.3) years.

Clinical parameters are reported in table 1. Thirty-two patients (45.1%) had dSSc, 38 (53.5%) lcSSc and one lSSc. The mean inter-incisor distance was 34.9 (7.6) mm. The mean Karnofsky score was 80 (SD 9) (range 60–100).

## Test–retest reliability

The test–retest reliability was analysed for 40 patients (35 (87.5%) females) with a mean age of 54.3 (SD 10.2) years who completed the questionnaire twice within a mean interval of 24 (range 20–28) h. No specific treatment was introduced between the 2 evaluations. Test–retest reliability analysis gave an ICC of 0.96, indicating excellent reliability.

## Outcome measure scores

The mean MHISS score was 20.3 (SD 9.7) and explained 36.5% of the variance of the HAQ global score. The mean HAQ global disability score was 1.20 (0.68). The mean CHSF score was 24.33 (19.26) and explained 75.5% of the variance of the HAQ global score, which confirms our previous finding in a different sample of patients.[7] The mean perceived handicap score (MACTAR) was 20.10 (8.63). A higher mean anxiety than

**Table 1** Demographic and clinical characteristics of 71 SSc patients

| | |
|---|---|
| Age at the time of evaluation, mean (SD) | 57.6 (11) |
| Sex, F (%) | 61 (83.6) |
| Age at disease onset, mean (SD) | 45.1 (12.5) |
| Disease duration at the time of evaluation, mean (SD) | 13.7 (12.3) |
| Skin involvement, n (%) | 70 (98.6) |
| Limited SSc, n (%) | 1 (1.4) |
| Limited cutaneous SSc, n (%) | 38 (53.5) |
| Diffuse SSc, n (%) | 32 (45.1) |
| Karnofsky index score, mean (SD) | 80 (9) |
| Raynaud's syndrome, n (%) | 69 (97.2) |
| Pitting scars, n (%) | 40 (56.3) |
| Digital ulcers, n (%) | 18 (25.3) |
| Calcinosis | 32 (45.1) |
| Inter-incisor distance (mm), mean (SD) | 34.9 (7.6) |
| Gastrointestinal tract involvement, n (%) | 62 (87.3) |
| Arthralgias, n (%) | 55 (77.5) |
| Myalgias, n (%) | 47 (66.2) |
| Dyspnoea (nYHA), mean (SD) | 1.90 (0.75) |
| Interstitial lung disease, n (%) | 20 (28.2) |
| Pulmonary arterial hypertension, n (%) | 5 (7) |
| Renal crisis, n (%) | 5 (7) |

SSc, systemic sclerosis; SD, standard deviation; F, female; n, number; NYHA, New York Heart Association.

depression score was observed, with values of 9.49 (4.51) and 7.04 (4.36), respectively.

### Construct validity of the MHISS

We confirmed our hypothesis of divergent validity of the MHISS with global disability (HAQ, $r = 0.33$), hand function (CHFS, $r = 0.37$), inter-incisor distance ($r = -0.34$), handicap (MACTAR, $r = 0.11$), depression (HADd, $r = 0.26$) and anxiety (HADa, $r = 0.17$) (table 2). Factor analysis extracted 3 factors with eigenvalues of 4.26, 1.76 and 1.47, respectively, explaining 62.44% of the variance (table 3). These 3 factors could be clinically characterised. The first factor (5 items, items 1, 3, 4, 5 and 6) represents handicap induced by reduced mouth opening, the second (5 items, items 7, 8, 9 and 10) handicap induced by sicca syndrome, and the third (2 items, 11 and 12) aesthetic concerns. The mean score for factor 1 was 7.8 (SD 5.5) (range 0–20); factor 2, 8.6 (4.4) (range 0–20); and factor 3, 3.7 (2.7) (range 0–8). The factorial structure of the scale was similar to the a priori domain stratification proposed by the experts.

The MHISS total score and factor scores were not significantly different between patients with lSSc or lcSSc, or with dSSc (table 4) or inter-incisor distance (data not shown). Thirty-five patients (49.2%) complained of a dry mouth. MHISS total and factor scores were significantly higher for patients reporting problems with mouth opening often or always than for those answering never, rarely or occasionally (table 4). The MHISS total score and factor 3 score (aesthetic concerns) were significantly higher for patients with telangiectasia than for those without, but factor 1 and 2 scores did not differ significantly (table 4).

**Table 2** Divergent validity of the MHISS with other variable scores

| Variable score | Spearman correlation coefficient (r) |
|---|---|
| Mouth opening | 0.34 |
| Cochin hand function scale | 0.37 |
| HAQ | 0.33 |
| Anxiety (HADa) | 0.17 |
| Depression (HADd) | 0.26 |
| McMaster-Toronto Arthritis questionnaire | 0.11 |

HAQ, Health Assessment Questionnaire; HAD, Hospital Anxiety and Depression scale; HADa, HAD anxiety; HADd, HAD depression.

## DISCUSSION

Our data suggest that the MHISS is a reliable and valid scale to assess disability involving the mouth in SSc patients. To our knowledge, this is the first mouth-specific disability outcome measure designed for SSc patients. Although mouth disability seems to have less weight than hand disability in total disability, the MHISS score explained up to 36% of the variance of the HAQ score, which highlights the need to specifically assess disability involving the mouth in patients with SSc when evaluating treatments.

The construct validity of this scale seems acceptable. Construct validity is a major criterion of the validity of a questionnaire.[19] To assess divergent validity, we used disability scales (HAQ and CHFS) that have recently been shown to be valid for SSc patients.[7] Attendant divergent validity was observed with global and hand disability, patients' perceived disability, anxiety, depression and inter-incisor distance. Since mouth disability assessed by the MHISS represents only 36% of the variance of the HAQ score, the only fair correlation observed between the 2 scores is not surprising. Although not negligible, mouth disability represents only a part of total disability in SSc. This observation probably also explains the low correlation between the MHISS and MACTAR score, which is a sum of disabilities in 3 different activities chosen by the patient.[17] Most of the time, patients choose no more than one activity involving mouth disability on this scale. The fair correlation between MHISS and CHFS scores is also not surprising, because although the 2 scales assess the same concept (disability), they are specific to different locations. The fair correlation between the MHISS score and inter-incisor distance seems more surprising. After factor analysis, mouth-opening limitation seemed to be the main factor implicated in disability due to mouth and face involvement for SSc patients. This finding highlights that mobility and disability are 2 distinct concepts. A weak correlation between mobility and disability has been observed in rheumatoid hands,[20] hand[21] and knee[22] osteoarthritis, and SSc hand involvement.[7]

The 3 factors we extracted corresponded to the experience of clinicians dealing with SSc patients. In fact, the 10 experts involved in the selection of items assumed that handicap induced by mouth involvement in SSc would be due to mouth-opening restriction, mouth dryness and aesthetic concerns. This

**Table 3** Factor analysis and varimax rotated factor matrix of the MHISS

| Factors | Eigenvalue | Variance (%) | Cumulative % |
|---|---|---|---|
| F1 | 4.26 | 35.48 | 35.48 |
| F2 | 1.76 | 14.69 | 50.67 |
| F3 | 1.47 | 12.27 | 62.44 |
| **MHISS items** | **Factor 1** | **Factor 2** | **Factor 3** |
| 1 | 0.844 | 0.184 | 0.131 |
| 2 | 0.393 | 0.474 | 0.393 |
| 3 | 0.694 | 0.292 | 0.038 |
| 4 | 0.812 | 0.062 | 0.096 |
| 5 | 0.876 | 0.005 | 0.207 |
| 6 | 0.754 | 0.081 | 0.135 |
| 7 | 0.277 | 0.572 | 0.277 |
| 8 | 0.013 | 0.676 | 0.061 |
| 9 | 0.028 | 0.742 | 0.091 |
| 10 | 0.184 | 0.663 | 0.390 |
| 11 | 0.149 | 0.107 | 0.847 |
| 12 | 0.208 | 0.122 | 0.844 |

The highest loading of each item is shown in bold.

**Table 4** MHISS total score and factor scores according to type of SSc, problems with mouth opening and telangectasia

| | Type of SSc | | | Problems with mouth opening | | | Telangectasia | | |
|---|---|---|---|---|---|---|---|---|---|
| Score | l SSc or lcSSc (n = 39) | dSSc (n = 32) | p value | Never, rarely, occasionally (n = 58) | Often, always (n = 13) | p value | Yes (n = 54) | No (n = 17) | p value |
| Total MHISS (range 0–48) (SD) | 18.44 (9.92) | 22.06 (9.45) | 0.134 | 17.60 (7.74) | 32.23 (8.66) | <0.001 | 21.52 (9.92) | 16.25 (8.04) | 0.038 |
| Factor 1 (range 0–20) (SD) | 7.31 (5.59) | 8.41 (5.60) | 0.428 | 6.14 (4.26) | 15.39 (3.73) | <0.001 | 8.14 (5.63) | 6.87 (5.04) | 0.396 |
| Factor 2 (range 0–20) (SD) | 7.82 (4.34) | 9.23 (4.53) | 0.194 | 7.83 (4.07) | 11.54 (4.52) | 0.015 | 8.98 (4.38) | 7.18 (4.09) | 0.128 |
| Factor 3 (range 0–8) (SD) | 3.23 (2.46) | 4.09 (2.77) | 0.178 | 3.41 (2.57) | 5.31 (2.78) | 0.037 | 4.11 (2.65) | 2.47 (2.43) | 0.024 |

Factor 1, ''mouth opening''; factor 2, ''mouth dryness''; factor 3, ''aesthetic concerns''; lSSc, limited scleroderma; lcSSc, limited cutaneous scleroderma; dSSC, diffuse scleroderma.

finding reinforces the robustness of the factorial structure of our scale.

Factor 1 of the MHISS is related to restriction in mouth opening. Diminished mouth opening results from temporomandibular joint involvement as well as atrophy of lips, skin and subcutaneous tissues. As a consequence, SSc patients complain of difficulties opening the mouth and restricted mouth widening, and experience difficulties during dental surgery. Patients reporting more problems with mouth opening had a higher MHISS total and factor 1 scores than those reporting fewer problems but also seemed to experience more mouth dryness and aesthetic prejudice.

Factor 2 of the scale is related to mouth dryness, even though some items such as items 2 and 9 may also be related to gastro-oesophageal reflux. SSc patients frequently complain of mouth dryness (49% in this study reported mouth dryness occurring often or always), but one prospective study of patients citing mouth dryness found only 13% fulfilling the European criteria for Sjögren's syndrome.[23] Moreover, another study found only 6% of SSc patients complaining of mouth and/or eye dryness with a diagnosis of Sjögren's syndrome.[24] We did not investigate specifically Sjögren's syndrome in our patients and therefore cannot evaluate the exact proportion of these patients with sicca or Sjögren's syndrome. However, a similar proportion of SSc patients (7%) who contributed to elaboration of our provisional scale had Sjögren's syndrome according to the European criteria. Therefore, it is unlikely that items related to sicca syndrome might be over-represented in our scale. Mouth dryness may not be restricted to patients experiencing Sjögren's syndrome and probably constitutes a classic symptom of SSc. Two items of Factor 2, about mouth dryness, concern food intake. A significant proportion of patients avoid certain drinks (sparkling, alcoholised, acidic) and compose their meals according to what they can eat, not what they would like to eat. The reasons why patients complain of these difficulties are probably intricate. Sicca syndrome probably plays an important role, but gingival abnormalities and gastro-oesophageal reflux may also be involved.

Factor 3 is related to the aesthetic concerns. This factor is probably related to skin retraction and telangiectasia. In fact, patients with telangiectasia had a higher score on this factor than those without. Patients with lcSSc more often showing telangiectasia and those with dSSc more often showing pronounced skin sclerosis of the face may explain the lack of difference in Factor 3 score between patients with lcSSc and dSSc.

Our work has several limitations. The list of items was generated largely from a sample of patients followed in one internal medicine department of a tertiary care hospital, whereas the item-selection process involved members of a patient association. Concerning the former sample, even though the department is a national referral centre for the management of SSc, we cannot exclude a recruitment bias, particularly that patients from this sample might be more likely to have a severe form of the disease than the general population of SSc patients. Concerning the latter sample, HAQ scores observed for patient members of this association are quite high and comparable with those reported from a previous study conducted in a tertiary care setting,[25] and patients are long-standing, which could contribute to more symptoms. Therefore, a further evaluation in other cohorts of SSc patients is necessary to confirm the reliability and construct validity of the MHISS.

Another limitation could be the relatively small number of patients tested with the scale. No consensus exists on the minimum number of subjects needed for principal-component analysis. A minimum of 100–300 subjects has been proposed,[26 27] or 5–10 times the number of variables.[28] However, since the required sample size also depends on the magnitude of real correlations and real number of factors in the population, with strong correlations (ie, $r>0.7$) and few distinct factors, a smaller sample size is adequate,[29] and 50 cases should be sufficient.[30 31]

Finally, we did not assess sensitivity to change in MHISS. This property should be confirmed before the scale is used to assess treatment efficacy.

In conclusion, we propose a new scale, the MHISS, which has excellent reliability and good construct validity, to assess disability involving the mouth in SSc patients. This scale has 3 factors that more specifically explore disability due to mouth opening limitation, mouth dryness and the aesthetic concerns. The MHISS might be considered as a measure of outcome in future scleroderma trials.

## ACKNOWLEDGEMENTS

We thank patients followed at the Department of Internal Medicine of Cochin Hospital and those from the Association des Sclérodermiques de France (ASF) for their participation in this study. We also thank members of the ASF for their logistical help. Luc Mouthon, Alice Bérezné, Jean Cabane and Loïc Guillevin are membres of the Groupe Français de Recherche sur la Sclérodermie.

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Competing interests: None declared.

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This work was presented at the 70th ACR/ARHP Annual Scientific Meeting, 10–15 November 2006, Washington, DC, USA.

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## APPENDIX 1

**Table A1** MHISS scale

| | | Never | Rarely | Occasionally | Often | Always |
|---|---|---|---|---|---|---|
| 1 | I have difficulties opening my mouth | 0 | 1 | 2 | 3 | 4 |
| 2 | I have to avoid certain drinks (sparkling, alcohol, acidic) | 0 | 1 | 2 | 3 | 4 |
| 3 | I have difficulties chewing | 0 | 1 | 2 | 3 | 4 |
| 4 | My dentist has difficulties taking care of my teeth | 0 | 1 | 2 | 3 | 4 |
| 5 | My dentition has become altered | 0 | 1 | 2 | 3 | 4 |
| 6 | My lips are retracted and/or my cheeks are sunken | 0 | 1 | 2 | 3 | 4 |
| 7 | My mouth is dry | 0 | 1 | 2 | 3 | 4 |
| 8 | I must drink often | 0 | 1 | 2 | 3 | 4 |
| 9 | My meals consist of what I can eat and not what I would like to eat | 0 | 1 | 2 | 3 | 4 |
| 10 | I have difficulties speaking clearly | 0 | 1 | 2 | 3 | 4 |
| 11 | The appearance of my face is modified | 0 | 1 | 2 | 3 | 4 |
| 12 | I have trouble with the way my face looks | 0 | 1 | 2 | 3 | 4 |

As you are probably aware, your systemic sclerosis might involve your face and your mouth. This questionnaire is aimed at assessing how much your face and mouth involvement affects your daily life.