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Title: Validity and reliability of the Mouth Handicap of Systemic Sclerosis questionnaire in a UK population

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Structured abstract:

Background: The objective of this study was to explore the psychometric properties of the Mouth Handicap in Systemic Sclerosis (MHISS) specifically exploring aspects of validity and reliability of MHISS in UK SSc patients.

Methods: 150 questionnaires were distributed in this study at the Oral Medicine Department of UCLHT Eastman Dental Hospital, the Outpatient Rheumatology Clinic of the Royal Free Hospital and Scleroderma Family Day. Participants were asked to complete three questionnaires (SF-36, OHIP-14 and MHISS) along with a proforma of demographic and disease-specific questions. Both convergent validity and internal consistency reliability were examined. The convergent validity was assessed by the Spearman's correlation coefficient and internal consistency reliability was assessed by Cronbach's alpha.

Results: Of the 150 questionnaires distributed to participants, 68 respondents included. With regard to construct validity, a low to moderate degree of convergent validity was found between MHISS total score and all SF-36 subscales. However, the total MHISS score was highly correlated to all OHIP subscales.

Conclusion: MHISS questionnaire is a self-administered patient-reported outcome measure that assesses the mouth disability in SSc patients. However, improving the global and oral health of SSc patients is important as there is no cure for the disease thus instruments that record the impact of the condition and indicate the effect of treatment on the lives of patients are of paramount importance. Current results demonstrate good preliminary psychometric properties (validity and reliability) in a UK population with further exploration of psychometric properties with an emphasis on interpretability required.

Key words:

Systemic sclerosis, Oral health, Quality of life, Outcome measures.

Introduction:

Systemic sclerosis (SSc) is a rare multisystem autoimmune disease of unknown cause characterised by induration of the skin, joint modification and muscle impairment ¹. It has a propensity to affect the skin as well as internal organs leading to a reduction in physical function, quality of life and survival ². The prevalence of systemic sclerosis is estimated to be 1 – 15/100.000 of the population, affecting middle to late age, with predominance among females ³. Approximately 90% of patients with SSc have a variety of orofacial features such as the fibrosis of the facial skin, microstomia, salivary gland dysfunction and xerostomia, dysphagia as well as an increased risk of dental caries and periodontal disease ⁴. A recent systematic review has demonstrated a correlation between orofacial features of SSc and impaired quality of life of patients with SSc ⁵.

Due to the nature of the disease and the lack of a definitive cure, the management of SSc can be challenging, and patients may face significant physical, social and psychological impacts on daily life ⁶. SSc is commonly associated with anxiety, fear and depression that might be related to the unpredictable course of the disease and the concern about the future not only due to the disease manifestation but also due to the increased mortality reported in this cohort of patients ⁷.

Living and coping with the disease itself and its subsequent complications usually need both pharmacological and non-pharmacological therapeutic efforts and referral to multiple healthcare clinicians. The goal of treatment is to improve health-related quality of life (HRQoL) and reduce the morbidity of the disease. Quality of life is a multidimensional concept incorporating areas such as physical function, social function, psychological, emotional, cognitive, spiritual, work-related and financial aspects of health and disease. Since early 1990, the utility of patient-reported outcome measures (PROMS) in the medical field, both in clinical practice and in research has grown significantly. Efforts have been made over the last two decades to enhance the acceptance of PROMS in routine clinical practice and research ⁸. However, more than 250 PROMS exist in the field of rheumatology ⁹. Although the use of PROMS is thought to be helpful in recording the impact of the disease from the perspective of the patient, further research is needed to refine disease-specific PROMS in SSc and to investigate psychometric properties.

As highlighted in the literature, the majority of patients with SSc have impairment of health-related quality of life (HRQoL) and oral health-related quality of life (OHRQoL) that

should not be underestimated when considering the spectrum of systemic manifestations of the disease ¹⁰. Generic and specific oral health-related quality of life measures have been used to assess various aspects of SSc (e.g. The Oral Health Impact Profile and the 36-item Short-Form Health Survey). Although these have been used frequently they may not successfully assess the magnitude and impact of the oral features of the disease as they were not developed in a population with SSc.

The Mouth Handicap in Systemic Sclerosis questionnaire (MHISS) was developed in a French population with SSc and has proven to be valid and reliable scale to assess disability involving the mouth in SSc patients ¹¹. However, given the necessity to explore the cross-cultural sensitivity of health-related quality of life measures researchers have examined the psychometric properties of MHISS in Italian and Dutch populations with supporting outcomes ^{12,13}. The MHISS has not been tested in the UK population, hence the aims of this study were to explore the psychometric properties of MHISS, namely validity and reliability, in an English-speaking UK population.

Material and Methods:

Recruitment and data collection:

This observational study had a cross-sectional design. A total of 150 questionnaires were distributed at 3 sites, Oral Medicine Department of UCLHT Eastman Dental Hospital, the Outpatient Rheumatology Clinic of the Royal Free Hospital and Scleroderma Family day, between 2016 and 2018. Patients over the age of 18 years with a diagnosis of SSc and who had an adequate command of the English language to comprehend the questionnaires were invited to complete the study questionnaire pack. This pack included the Oral Health Impact Profile (OHIP-14), the 36-item Short-Form Health Survey (SF-36) and the Mouth Handicap in Systemic Sclerosis questionnaire (MHISS).

The Mouth Handicap in Systemic Sclerosis questionnaire (MHISS) was developed by Mouthon et al. in 2007 to identify and evaluate the limitations of the oral manifestations of condition SSc on individuals and aims to improve the oral health care mainly by measuring the degree of restriction of mouth opening, dryness of the mouth and the aesthetic appearance. It has been developed in a French population and subsequently validated in French, Italian and Dutch languages ¹¹⁻¹³. It consists of 12 items (with five levels of answers), divided into 3 subscales as subscale one examines impact related to

reduced mouth opening, subscale 2 examines impact related to dryness of the mouth and subscale 3 examines aesthetic concerns. The total score ranges from 0 to 48 by summing the score for all items.

The Oral Health Impact Profile (OHIP) is a questionnaire designed by Slade and Spencer to measure OHRQoL in adults with oral disease ¹⁴. It has been used widely in patients with SSc ^{10,15}. The theoretical basis for the development of this tool was Locker's model of oral health, which includes aspects of the influence of the disability of disease on oral health-related quality of life. It was originally developed as a 49-item tool representing seven domains including functional limitation, physical pain, psychological discomfort, physical disability, psychological disability, social disability and handicap. This was subsequently refined to the 14 item OHIP-14 by including two questions in each of the seven OHIP aspects ¹⁶. Each question is rated on a 5-point Likert scale to record the frequency of the oral problems. Patients are invited to answer questions by choosing from 0-4 scale while 0=never and 4=very often. The total score ranges from 0 to 56 by summing the score for all items.

The 36-item Short-Form Health Survey (SF-36) is designed as a generic assessment of health status with a wide range of types and severity of conditions and has shown good psychometric properties with SSc patients ^{17,18}. The RAND 36-item Health Survey 1.0 measures eight aspects of health including physical functioning, role-physical, bodily pain, general health, vitality, social functioning, role-emotional and mental health. Each item is scored on a 0 (worst health) to 100 (best health) scale so that the lowest and highest possible scores are 0 and 100, respectively. Items Scores represent the percentage of total possible score achieved. Selected items averaged together to create the eight scale scores ¹⁹.

Participants were asked to return the completed questionnaire on-site or in a stamped addressed envelope to one of the authors (SRP). After completion of the data collection, all data were transferred to Excel spreadsheets, tabulated and later interpreted where appropriate using the SPSS statistical software package (version 25).

Ethical approval was sought for this study; however, as this was considered to be an evaluation of service, ethical approval was not deemed necessary.

Psychometric testing:

Convergent validity, a type of construct validity, was assessed. Construct validity explores whether the instrument can reflect theories and traits of SSc. Convergent validity is the extent to which MHISS relates to other instruments of a similar construct to which it is proposed to be related ²⁰. The initial proposal explored in this study was that patients with poor general HRQoL, as measured by SF-36, would have poorer scores as recorded by MHISS. Secondly, as Mouthon et al. indicates that MHISS measures mouth-related disability, which is a component of Locker's model of oral health, the theoretical basis of OHIP-14, we anticipated that patients with poor OHRQoL, as measured by OHIP-14, would have poorer scores as recorded by MHISS. Spearman's rho correlations were used to assess the relationship between MHISS and SF-36 and between MHISS and OHIP-14. The following grading of the degree of correlation was applied 0.1 – 0.29 low correlation, 0.3 – 0.49 moderate correlation, 0.5 – 1.0 good high correlation ^{13,21}.

Internal consistency reliability refers to the degree to which items in an instrument correlate with each other and should also correlate with the overall scale score ²². Cronbach's alpha coefficient was used to determine internal consistency reliability which can range from 0.0 to 1.0. In this study, Cronbach's alpha of 0.6 to 0.69 was considered acceptable, 0.7 or higher were required before an instrument is considered adequately reliability, however, a minimum value of 0.8 was necessary for 'good' internal consistency reliability ¹³.

Results:

Patient characteristics:

Of the 150 questionnaires distributed to patients, 68 responded, giving a response rate of 45.3%. Of the 68 study participants, 64 were female (94%) and 4 were male. The mean age was 62.5 (± 10) years and mean disease duration was 13.2 (± 11) years. Most of the study sample were White British (73.5%). All the participants were non-smokers, and 33.8% of the sample consumed alcohol. Approximately a third of the patients had diffuse cutaneous SSc (dcSSc) (35.3%), 19.1% had limited cutaneous SSc (lcSSc), 7.4% had overlap SSc and 38.2% were reported with unknown disease subtype. With regard to orofacial manifestations of SSc, 48% of the participants reported experience of facial skin tightness and telangiectasia, 44% reported having dysphagia, 42% reported having

microstomia, 32% reported having gingival diseases, and 30% reported having teeth mobility and oral ulceration. Patient demographics, disease characteristics and symptoms are summarised in Table 1 and 2.

The response frequencies to MHISS, OHIP-14 and SF-36 tended towards skewed distributions, with some subscales demonstrating normal distribution. Due to the mixture between normal and skewed data the median, mean, standard deviation, standard error and interquartile range scores for each subscale in MHISS, and the OHIP-14 and for SF-36 have been reported in Table 3.

Validity:

With respect to convergent validity, all Spearman's rho correlation coefficients were significant at the $p < 0.05$ and $p < 0.01$ levels as seen in Table 4 and 5. The negative correlation seen in Table 4 reflect the fact that the scores of SF-36 and MHISS are inversely related; a higher score in SF-36 reflects a better HRQoL whilst a higher score in MHISS indicates a worse impact of SSc on the patient. Results show low correlation between the following - all MHISS domains and SF-36 Role-physical domain; MHISS Mouth opening restriction and SF-36 Social functioning; MHISS Mouth dryness and both SF-36 Vitality and Bodily pain; MHISS Aesthetic concerns and SF-36 Role-emotional and Vitality. A moderate correlation was found between MHISS Mouth opening restriction and SF-36 Role-emotional, Vitality and Bodily pain domains. Also, moderate correlation was found between MHISS Mouth dryness and both SF-36 Role-emotional and Social-functioning. MHISS total scores were found moderately correlated to all SF-36 except for both Physical functioning and Mental health domains. However, Spearman's rho correlation coefficients indicated that all MHISS subscales were moderately correlated to SF-36 General health domain.

Most of the MHISS domains were found to positively and highly correlated to the OHIP subscales with Spearman's rho correlation coefficients between 0.543 and 0.725 at the $p < 0.01$ level as seen in (Table 5). However, data shows some degree of moderate correlation was found between MHISS Mouth opening restriction and both OHIP Functional limitation and Social disability and between MHISS Mouth dryness and OHIP Psychological disability, Handicap and OHIP total score. MHISS Aesthetic concerns

domain was only highly correlated to OHIP (Psychological discomfort, disability and total score). The total MHISS score was highly correlated to all OHIP subscales except the Social disability score that reflect moderate level of correlation.

Reliability:

With regard to internal consistency reliability, the mouth dryness domain with Cronbach's alpha value of 0.76 demonstrated adequate internal consistency reliability. The other two domains including mouth opening restriction and aesthetic concerns indicated good internal consistency reliability level with Cronbach's alpha values of 0.878 and 0.874 respectively. The overall MHISS Cronbach's alpha value of 0.894 indicated very good internal consistency reliability of the 12-item instrument. The Cronbach's alpha values of 0.898, 0.918 and 0.905, respectively, indicated good internal consistency reliability with respect to the 'Psychological discomfort' and 'Physical disability' subscales and the OHIP-14 total score. However, the Cronbach's alpha values of all other OHIP-14 subscales were ranged between (0.716 – 0.779) indicated adequate internal consistency reliability. All SF-36 subscales Cronbach's alpha values indicated good internal consistency reliability except the Cronbach's alpha values of 0.733 and 0.745, respectively, indicated adequate internal consistency reliability with respect to the 'Mental health' and 'Social functioning'. (Table 6).

Discussion:

SSc is a heterogeneous disease with high morbidity and impacts upon patients' HRQoL⁶. Given the impact of the widespread of orofacial manifestations on both HRQoL and OHRQoL, continuous clinical evaluation should be performed routinely in parallel to the continuous assessment of disease-related mouth disability⁴.

Although the value and merit of PROMS use in chronic disease have long been established, the importance of selecting and administering appropriately valid and reliable instrument cannot be underestimated. It can be challenging for clinicians and researcher to select appropriate tools. Streiner and Norman recommended the use of both a generic and a disease-specific outcome measure when evaluating the impact of a disease on the daily life of a patient²³. Generic instruments can demonstrate a change in the HRQoL, but they cannot detect small clinical important changes in specific disease. On the other hand, specific measures are more accurately predict most of the clinical changes but do

not allow for comparison between diseases. However, evidence shows that using both generic and specific HRQoL measures when evaluating the impact of a disease is recommended ²⁴.

Oral health-related quality of life measures has been applied among different groups of oral diseases demonstrating valid and reliable outcomes ⁸. Both SF-36 and OHIP were found to be the most frequently used PROMS when assessing the impact of different oral diseases, it is reasonable to consider using these tools when evaluating the impact of the oral manifestations of SSc on patients ²⁵. However, concerns have been expressed regarding the suitability of OHIP-14 as an OHRQoL measure in certain patient cohorts ²⁶. Therefore, using specific disease HRQoL measures could offer more accurate and reliable results in both clinical and research settings as seen in recent reports ^{27,28}.

MHISS was developed as a disease-specific tool to specifically evaluate the 'Mouth Handicap' in patients with SSc, this may be a viable alternative to OHIP-14 in this patient population. The oral health-related quality of life has been assessed by MHISS scale in many studies ^{28,29}. MHISS has been reported with acceptable validity and reliability in different languages over different countries ¹¹⁻¹³. To the best of our knowledge, there is no study has investigated the psychometric properties of the MHISS questionnaire among patients with SSc in the UK. Therefore the aim of this study was to explore the psychometric properties of MHISS in a cohort of UK patients with SSc.

Current results demonstrate a moderate correlation between MHISS total score and SF-36 general health ($r=-0.446$) and other domains. However, the low correlation level that has been reported among different SF-36 subscales highlighting that the SF-36 questionnaire may not comprehensively capture and evaluate specific orofacial impairments related to SSc including mouth opening reduction, mouth dryness and impaired aesthetics and therefore OHRQoL should be evaluated using disease-specific HRQoL measures. As MHISS represents only a part of total disability in SSc ¹¹, the mouth disability measured by MHISS in the present study was not significantly related to both SF-36 mental health and general physical functioning domains. However, as in previous studies, our data showed some lack of correlation between MHISS and different SF-36 subscales that may explain the generic nature of the SF-36 questionnaire ¹².

When considering oral health related quality of life, the results demonstrated that the total MHISS score has high correlation with total OHIP-14 ($r=0.725$) in addition to most of the

subscales of the tool. However, low to moderate level of correlation has been noticed between different MHISS and some of the OHIP-14 subscales emphasising the need for using a more sensitive HRQoL measures related to mouth disability among patients with SSc. Although these overall values reflecting moderate to high convergent validity, variations among different subscales could be further explored in a more general study sample of patients with SSc undertaking qualitative assessment regarding the MHISS instrument and the impact of the disease on their lives and daily activities.

Cronbach's alpha should approach a good level for the instrument to be considered reliable for use in clinical practice. However, MHISS has shown a good level of internal consistency reliability as the values of Cronbach's alpha for the MHISS total score was 0.894 and this was also mirrored in the study of Dutch version of MHISS reporting good internal consistency reliability level of MHISS with total MHISS score of 0.88¹³. Although these values are considered good but they are lower than the values found in a previous study using an Italian translated version of MHISS in patients with SSc demonstrated excellent reliability (Cronbach's alpha = 0.99)¹² however, these values of reliability could be further investigated in a different setting to obtain precise estimates as it is dependent on the number of items as well as the level of correlation between them.

This study has some limitations, which include the low response rate (n=68) to the survey. A number of studies published in the literature have explored methods of improving response rates to surveys in patient and physician populations, with paper, postal and online surveys. Suggestions such as incentives and reminders were made, which should have been considered in our study design. Recruitment in a tertiary referral centre certainly facilitated access to this patient population but the findings may not reflect the general SSc patient population. Also, the present study is limited by its observational single-point in time cross-sectional design, restricting the number of psychometric properties that could be explored. Ideally, a clinical examination could have been to explore the exact orofacial features of SSc thus facilitating comparisons between OHRQoL scores and the orofacial features are presented.

Conclusion:

Given the impact of the widespread of orofacial manifestations of SSc on HRQoL, healthcare providers caring for patients with SSc should be aware of the availability of such reliable and valid instrument and its essential role to improve OHRQoL. Current results demonstrate good preliminary psychometric properties (validity and reliability) in a UK population with further exploration of psychometric properties with an emphasis on interpretability required.

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Disclosure statement:

No conflict of interest was reported by the authors.

Author contributions:

Ismail Abdouh and Richeal Ni Riordain designed the study. Ismail Abdouh collected data from patients, carried out the statistical analyses and drafted the manuscript. Stephen Porter, Stefano Fedele and Richeal Ni Riordain edited and contributed comments on the manuscript.

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Table 1. Sociodemographic and disease characteristics of SSc patients (n=68)

Variable	Value
Age (year), mean (SD)	62.5 (10)
Disease duration (years), mean (SD)	13.2 (10.9)
Female, n %	64 (94)
Smoking, n %	0 (0)
Alcohol, n %	23 (33.8)
Ethnicity	British White, n %
	50 (73.5)
	Other White, n %
	6 (8.8)
	Indian, n %
	4 (5.9)

	Black African, n %	1 (1.5)
	Black Caribbean, n %	2 (2.9)
	Pakistani, n %	1 (1.5)
	Other Asian backgrounds, n %	1 (1.5)
	Other ethnicity, n %	3 (4.4)
Disease subtype	Diffuse cutaneous SSc, n %	24 (35.3)
	Limited cutaneous SSc, n %	13 (19.1)
	Overlap SSc, n %	5 (7.4)
	Unknown, n %	26 (38.2)

Table 2. Orofacial features related to SSc that experienced by patients (n=68)

Item	N (%)
Microstomia	29 (42.6)
Bleeding/recession gums	22 (32.3)
Loose/mobile teeth	20 (29.4)
Loose/mobile denture	8 (11.7)
Bruising/ulceration of the lining of the mouth (oral mucosa)	20 (29.4)

Tightness of facial skin/oral mucosa	33 (48.5)
Altered breath smell (halitosis)	8 (11.7)
Difficult root canal treatment (endodontics)	11 (16.1)
Difficulties with dental extractions	15 (22)
Oral infection	12 (17.6)
Speech impairment (dysarthria)	7 (10.2)
Swallowing difficulty (dysphagia)	30 (44.1)
Altered taste sensation (dysgeusia)	8 (11.7)
Tongue atrophy / ankylosis / rigidity	11 (16.1)
Salivary gland swelling/hypofunction	14 (20.5)
Facial/oral telangiectasia (pigmentation)	33 (48.5)
Fissured/cracked lips	19 (27.9)

Table 3. Descriptive statistics for the SF-36, OHIP-14 and MHISS scales of SSc patients

Instrument	Scale	Median	IQR	Mean (SD)	Std. Error
SF-36	Physical Functioning	45	20-70	48.05 (28.6)	3.47
	Role-physical	25	0-69	32.72 (40.3)	4.89

	Role-emotional	100	33-100	63.53 (41.4)	5.02
	Vitality	34	15-55	39.34 (28.2)	3.43
	Mental health	72	57-87	70.04 (20.4)	2.48
	Social functioning	62.5	41-88	61.95 (27.5)	3.33
	Bodily pain	56.2	37.5-78	58.04 (24.9)	3.02
	General health	30	20-60	40.59 (26.1)	3.17
OHIP-14	Functional limitations	3	1-4	2.97 (2.1)	0.26
	Physical pain	4	2-5	3.63 (2.2)	0.27
	Psychological discomfort	4	0.25-7	3.97 (2.9)	0.36
	Physical disability	2.5	0-4	2.85 (2.5)	0.31
	Psychological disability	3	0-4	2.81 (2.4)	0.29
	Social disability	1	0-3	1.63 (1.8)	0.22
	Handicap	1.5	0-3	1.79 (1.9)	0.23
	OHIP total	20	7.25-31	19.74 (13.0)	1.57
MHISS	Mouth opening restriction	12	4.25-18	11.31 (7.3)	0.89
	Mouth dryness	10	8-12	9.16 (4.0)	0.48
	Aesthetic concerns	4	1-6	4.03 (2.8)	0.34
	MHISS total	27	12.5-35	24.59 (12.1)	1.47

Table 4. Matrix of Spearman's rho coefficients between pairs of responses to the MHISS and SF-36 (n=68)

Instrument	SF-36 Physical Functioning	SF-36 Role- physical	SF-36 Role- emotional	SF-36 Mental health	SF-36 Vitality	SF-36 Social functioning	SF-36 Bodily pain	SF-36 General health
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MHISS Mouth opening restriction	-.158	-.262*	-.307*	-.046	-.300*	-.254*	-.331**	-.362**
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MHISS Aesthetic concerns	-.136	-.257*	-.247*	-.033	-.283*	-.187	-.092	-.316**
MHISS total score	-.221	-.315**	-.365**	-.058	-.372**	-.351**	-.310**	-.446**

** . Correlation is significant at the 0.01 level (2-tailed).

* . Correlation is significant at the 0.05 level (2-tailed).

Table 5. Matrix of Spearman's rho coefficients between pairs of responses to the MHISS and OHIP-14 (n=68)

Instrument	OHIP Functional limitations	OHIP Physical pain	OHIP Psychological discomfort	OHIP Physical disability	OHIP Psychological disability	OHIP Social disability	OHIP Handicap	OHIP total
MHISS Mouth opening restriction	.487**	.595**	.618**	.604**	.622**	.455**	.551**	.689**
MHISS Mouth dryness	.573**	.513**	.281*	.548**	.325**	.234	.453**	.489**
MHISS Aesthetics	.383**	.399**	.562**	.407**	.585**	.449**	.438**	.582**
MHISS total score	.575**	.628**	.609**	.652**	.623**	.456**	.602**	.725**

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Table 6. Internal consistency reliability of the MHISS, OHIP-14 and SF-36 scales

Instrument	Scale	Number of items (n=68)	Cronbach's alpha
MHISS	Mouth opening restriction	6	0.878
	Mouth dryness	4	0.760
	Aesthetic concerns	2	0.874
	Total score	12	0.894
OHIP-14	Functional limitations	2	0.714
	Physical pain	2	0.779
	Psychological discomfort	2	0.898
	Physical disability	2	0.918
	Psychological disability	2	0.775
	Social disability	2	0.730
	Handicap	2	0.716
	Total score	14	0.905
SF-36	Physical Functioning	10	0.927
	Role-physical	4	0.903
	Role-emotional	3	0.843
	Vitality	4	0.837
	Mental health	5	0.733
	Social functioning	2	0.745
	Bodily pain	2	0.868
	General health	5	0.816

Figure legends:

Table 1, Sociodemographic and disease characteristics of 68 SSc patients.

Table 2, Orofacial features related to SSc that experienced by patients (n=68).

Table 3, Descriptive statistics for the SF-36, OHIP-14 and MHISS scales of 68 SSc patients.

Table 4, Matrix of Spearman's rho coefficients between pairs of responses (n=68) to the MHISS and SF-36.

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Table 6, Internal consistency reliability of the MHISS, OHIP-14 and SF-36 scales.

Table 1. Sociodemographic and disease characteristics of SSc patients (n=68)

Variable		Value
Age (year), mean (SD)		62.5 (10)
Disease duration (years), mean (SD)		13.2 (10.9)
Female, n %		64 (94)
Marital status	Single, n %	10 (14.7)
	Married, n %	27 (39.7)
	Divorced, n %	10 (14.7)
	Widowed, n %	2 (2.9)
	Unknown, n %	19 (28)
Education level	No degree-level	6 (8.8)
	At degree-level or above	44 (64.7)
	Unknown	18 (26.5)
Work status	Working/Not working, %	42/58
	Paid employee/Self-employed, %	66.7/33.3
Work-time (Full/Part time), %		45.8/54.2
Smoking, n %		0 (0)
Alcohol, n %		23 (33.8)
Ethnicity	British White, n %	50 (73.5)
	Other White, n %	6 (8.8)
	Indian, n %	4 (5.9)
	Black African, n %	1 (1.5)
	Black Caribbean, n %	2 (2.9)
	Pakistani, n %	1 (1.5)
	Other Asian backgrounds, n %	1 (1.5)
	Other ethnicity, n %	3 (4.4)

Disease subtype	Diffuse cutaneous SSc, n %	24 (35.3)
	Limited cutaneous SSc, n %	13 (19.1)
	Overlap SSc, n %	5 (7.4)
	Unknown, n %	26 (38.2)

Table 2. Orofacial features related to SSc that experienced by patients (n=68)

Item	N (%)
Microstomia	29 (42.6)
Bleeding/recession gums	22 (32.3)
Loose/mobile teeth	20 (29.4)
Loose/mobile denture	8 (11.7)
Bruising/ulceration of the lining of the mouth (oral mucosa)	20 (29.4)
Tightness of facial skin/oral mucosa	33 (48.5)
Altered breath smell (halitosis)	8 (11.7)
Difficult root canal treatment (endodontics)	11 (16.1)
Difficulties with dental extractions	15 (22)
Oral infection	12 (17.6)
Speech impairment (dysarthria)	7 (10.2)
Swallowing difficulty (dysphagia)	30 (44.1)
Altered taste sensation (dysgeusia)	8 (11.7)
Tongue atrophy / ankylosis / rigidity	11 (16.1)
Salivary gland swelling/hypofunction	14 (20.5)
Facial/oral telangiectasia (pigmentation)	33 (48.5)
Fissured/cracked lips	19 (27.9)

Table 3. Descriptive statistics for the SF-36, OHIP-14 and MHISS scales of SSc patients

Instrument	Scale	Median	IQR	Mean (SD)	Std. Error
SF-36	Physical Functioning	45	20-70	48.05 (28.6)	3.47
	Role-physical	25	0-69	32.72 (40.3)	4.89
	Role-emotional	100	33-100	63.53 (41.4)	5.02
	Vitality	34	15-55	39.34 (28.2)	3.43
	Mental health	72	57-87	70.04 (20.4)	2.48
	Social functioning	62.5	41-88	61.95 (27.5)	3.33
	Bodily pain	56.2	37.5-78	58.04 (24.9)	3.02

	General health	30	20-60	40.59 (26.1)	3.17
OHIP-14	Functional limitations	3	1-4	2.97 (2.1)	0.26
	Physical pain	4	2-5	3.63 (2.2)	0.27
	Psychological discomfort	4	0.25-7	3.97 (2.9)	0.36
	Physical disability	2.5	0-4	2.85 (2.5)	0.31
	Psychological disability	3	0-4	2.81 (2.4)	0.29
	Social disability	1	0-3	1.63 (1.8)	0.22
	Handicap	1.5	0-3	1.79 (1.9)	0.23
	OHIP total	20	7.25-31	19.74 (13.0)	1.57
MHISS	Mouth opening restriction	12	4.25-18	11.31 (7.3)	0.89
	Mouth dryness	10	8-12	9.16 (4.0)	0.48
	Aesthetic concerns	4	1-6	4.03 (2.8)	0.34
	MHISS total	27	12.5-35	24.59 (12.1)	1.47

Table 4. Matrix of Spearman's rho coefficients between pairs of responses to the MHISS and SF-36 (n=68)

Instrument	SF-36 Physical Functioning	SF-36 Role- physical	SF-36 Role- emotional	SF-36 Mental health	SF-36 Vitality	SF-36 Social functioning	SF-36 Bodily pain	SF-36 General health
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