

## Concise report

**The Canadian Systemic Sclerosis Oral Health Study II: the relationship between oral and global health-related quality of life in systemic sclerosis**

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**Abstract**

**Objective.** Both oral and global health-related quality of life (HRQoL) are markedly impaired in SSc. In this study we aimed to determine the degree of association between oral HRQoL and global HRQoL in SSc.

**Methods.** Subjects were recruited from the Canadian Scleroderma Research Group registry. Global HRQoL was measured using the Medical Outcomes Trust 36-item Short Form Health Survey (SF-36) and oral HRQoL with the Oral Health Impact Profile (OHIP). The Medsger Disease Severity Score was used to determine organ involvement. Multivariate regression models determined the independent association of the OHIP with the SF-36 after adjusting for confounders.

**Results.** This study included 156 SSc subjects. The majority (90%) were women, with a mean age of 56 years, mean disease duration 13.8 years (s.d. 8.5) and 29% of the subjects had dcSSc. Mean total OHIP score was 40.8 (s.d. 32.4). Mean SF-36 mental component summary (MCS) score was 49.7 (s.d. 11.1) and physical component summary (PCS) score was 37.0 (s.d. 10.7). In adjusted analyses, the total OHIP score was significantly associated with the SF-36 MCS and PCS, accounting for 9.7% and 5.6% of their respective variances. Measures of disease severity were not related to OHIP score.

**Conclusion.** Oral HRQoL in SSc is independently associated with global HRQoL. Oral HRQoL, however, is not related to physician-assessed disease severity. This suggests that physicians may be disregarding issues related to oral health. HRQoL is an additional dimension of HRQoL not captured by generic instruments such as the SF-36.

**Key words:** systemic scleroderma, oral health, oral pathology, oral hygiene, quality of life, questionnaires.

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## Introduction

SSc is a multisystem disease in which functional impairment and work disability are common. Oral manifestations of SSc include microstomia (decreased oral aperture and opening), xerostomia (dry mouth), caries, gingival recession, periodontal disease and bone resorption of the mandible, sometimes leading to fractures. Instruments have been developed to assess oral health-related quality of life (HRQoL) or that portion of a person's sense of well-being that may be diminished specifically by problems resulting from poor oral health [1]. We have recently demonstrated, using the Oral Health Impact Profile [1], a well-validated instrument, that oral HRQoL is markedly impaired in SSc compared with controls [2]. Although oral HRQoL has been previously studied in SSc [3, 4], to our knowledge, ours is the first study to compare oral HRQoL in SSc with a control population.

Global HRQoL is significantly impaired in SSc. There is some evidence from studies in non-SSc subjects that oral HRQoL is associated with global HRQoL [5]. We hypothesized that in SSc oral HRQoL would be significantly associated with global HRQoL. The purpose of this study was therefore to determine the relationship between oral HRQoL, measured using the OHIP, and global HRQoL in SSc measured using the Medical Outcomes Trust 36-item Short Form health Survey (SF-36) mental (MCS) and physical (PCS) component summary scores, beyond sociodemographic and disease variables known to account for impaired HRQoL in SSc.

## Patients and methods

### Study design and subjects

This multisite, cross-sectional study was conducted between 2008 and 2011. The research ethics board of each site approved the study and all study subjects provided informed consent in compliance with the Helsinki Declaration. All SSc patients were enrolled in the Canadian Scleroderma Research Group (CSRG) registry, had a diagnosis of SSc confirmed by a recruiting rheumatologist and were  $\geq 18$  years of age. Details of the recruitment methods are reported elsewhere [2].

### Demographics and disease variables

Information regarding sex, age, ethnicity, post-secondary education and smoking status was obtained by patient self-report. SSc disease duration was determined as the time from the onset of the first non-RP manifestation. Skin involvement was assessed using the modified Rodnan skin score (mRSS), ranging from 0 to 51 [6]. lcSSc was defined as skin involvement distal to the elbows and knees with or without face involvement and dcSSc was defined as skin involvement proximal to the elbows and knees, with or without truncal involvement [7]. Overall disease severity was assessed by physician global assessment, using a 0–10 numerical rating scale. The Medsger Disease Severity Score was used to determine severity of organ involvement [8]. In this scale, a severity score

ranging from 0 (normal) to 4 (end stage) is generated for each of nine organ systems.

### Measures of global HRQoL

Global HRQoL was measured using the SF-36, a widely used and evaluated health outcomes measure. It consists of eight domains, which can be scored separately, with scores ranging from 0 (the worst health state) to 100 (the best health state). Domain scores can also be summarized into MCS and PCS scores. Version 2 of the SF-36, which has been shown to be responsive to change in SSc [9], was used.

### Measures of oral HRQoL

Oral HRQoL was measured using the OHIP [1], which consists of 49 questions on the frequency of adverse oral conditions such as toothache, mouth pain, difficulty chewing or pronouncing words and discomfort related to appearance. Respondents indicate on a 5-point scale how frequently they experience each problem in their daily life. Response categories were coded using the following scale: 4, very often; 3, fairly often; 2, occasionally; 1, hardly ever; 0, never or not applicable. The items can be organized into seven subscales: functional limitation, physical pain, psychological discomfort, physical disability, social disability and handicap [1]. Additive scoring of the OHIP ranging from 0 to 196, with higher scores indicating worse oral HRQoL, was used in this study [1].

### Statistical analysis

Associations between sociodemographic characteristics, disease variables and OHIP scores and between the SF-36 MCS and SF-36 PCS and total OHIP score were assessed using Spearman's correlation coefficients for continuous variables and Student's *t*-test or Mann-Whitney *U* test for binary variables, as appropriate. Hierarchical multivariate linear regression models were used to determine the independent association of total OHIP score and each OHIP subscale with the SF-36 PCS and SF-36 MCS, after adjusting for age, sex, ethnicity (white vs non-white), education (more than high school vs high school or less), disease duration and disease severity (using the physician global assessment of severity). We repeated these models using study site as random effect and no differences in parameter estimates were found. All statistical analyses were performed with SAS 9.2 (SAS Institute, Cary, NC, USA). *P*-values  $< 0.05$  were considered as significant.

## Results

### Study subjects

Among the 163 SSc subjects who participated in the first study [2], we report here the results of the 156 subjects who had no missing data for any of the variables used for the current analysis. The 156 SSc subjects included in this study were compared with the 1221 non-participating CSRG subjects (Table 1). Study participants had longer disease duration [13.8 years (s.d. 8.5) vs 10.7 (s.d. 9.6),

**TABLE 1** Baseline characteristics of SSc study subjects and comparison with non-participating SSc subjects in the CSRG cohort

	SSc patients (n = 156) Mean (s.d.)	CSRG (n = 1221) Mean (s.d.)	P-value
Sociodemographic variables			
Female, n (%)	141 (90.4)	1047 (85.8)	0.113
White, n (%)	144 (92.3)	1006 (89.2)	0.232
Current smoker, n (%)	15 (9.6)	168 (14.9)	0.077
Education (>high school), n (%)	78 (50.0)	535 (47.7)	0.587
Age, mean (s.d.), years	56.1 (10.7)	55.8 (12.4)	0.683
Disease variables			
dcSSc, n (%)	39 (28.5)	432 (35.9)	0.084
Disease duration, mean (s.d.), years	13.8 (8.5)	10.7 (9.6)	<b>&lt;0.001</b>
mRSS (range 0–51), mean (s.d.)	8.4 (8.6)	9.7 (9.5)	0.074
Physician global assessment of severity (range 0–10), mean (s.d.)	2.9 (2.2)	2.8 (2.3)	0.297
Disease severity score (range 0–4), mean (s.d.)			
General	0.5 (0.7)	0.9 (1.2)	<b>0.002</b>
Gastrointestinal tract	2.1 (0.7)	1.9 (0.8)	<b>0.018</b>
Heart	0.5 (1)	0.5 (1)	0.604
Joint/tendon	0.7 (1.1)	0.7 (1.2)	0.774
Kidney	0 (0.1)	0.1 (0.6)	0.054
Lung	1.3 (1.2)	1.3 (1.1)	0.513
Muscle	0.2 (0.5)	0.3 (0.8)	0.217
Peripheral vascular	1.3 (1.2)	1.6 (1.3)	<b>0.002</b>
Skin	1.1 (0.7)	1.2 (0.7)	0.152
SF-36 MCS, mean (s.d.)	49.7 (11.1)	48.5 (12.4)	0.410
SF-36 PCS, mean (s.d.)	37 (10.7)	36.4 (9.3)	0.458

Significant *P*-values are highlighted in bold. CSRG: Canadian Scleroderma Research Group; MCS: mental component summary; mRSS: modified Rodnan skin score; PCS: physical component summary; SF-36: 36-item Short Form Health Survey.

$P < 0.001$ ], lower general and peripheral vascular disease severity scores [0.5 (s.d. 0.7) vs 0.9 (s.d. 1.2),  $P = 0.002$ , and 1.3 (s.d. 1.2) vs 1.6 (s.d. 1.3),  $P = 0.002$ , respectively] and higher gastrointestinal tract disease severity scores [2.1 (s.d. 0.7) vs 1.9 (s.d. 0.8),  $P = 0.018$ ] compared with non-participating subjects.

#### Baseline characteristics

The majority of the study subjects (90%) were women, mean age 56 years (s.d. 11), 92% were white and 50% had more than high school education (Table 1). Twenty-nine per cent had dcSSc. The mean score on the SF-36 MCS was 49.7 (s.d. 11.1) and on the SF-36 PCS was 37.0 (s.d. 10.7). The mean total OHIP score (range 0–196) was 40.8 (s.d. 32.4) (Table 2). The total OHIP score was 37.5 in subjects with dcSSc vs 41.4 in those with lcSSc ( $P = 0.623$ ).

#### Bivariate associations

The total OHIP score had a negative association with SF-36 MCS ( $r = -0.30$ ,  $P < 0.001$ ) and SF-36 PCS ( $r = -0.29$ ,  $P < 0.001$ ) scores, indicating that worse oral HRQoL was associated with worse overall HRQoL. All of the OHIP subscale scores were also negatively correlated with the SF-36 MCS and PCS scores. As expected, the OHIP was related to smoking ( $P = 0.012$ ). However, we found no relationship between any of the disease variables (dcSSc,

disease duration, mRSS, physician global assessment of severity, Medsger disease severity score) and the total OHIP score.

#### Hierarchical multivariate regression models

In adjusted analyses (Table 2), the total OHIP score had a statistically significant association with both the SF-36 MCS and the SF36-PCS after adjustment for possible confounding variables ( $P < 0.001$  and  $P = 0.002$ , respectively). Inclusion of the total OHIP score into the model accounted for 9.7% of the variance of the SF-36 MCS and 5.6% of the variance of the SF-36 PCS.

In separate multivariate analyses of the relationship between each OHIP subscale and both the SF-36 PCS and SF36-MCS, all subscales of the OHIP were significantly associated with a decrease in both the MCS and PCS except social disability, which was not associated with the PCS.

#### Sensitivity analysis

To confirm the robustness of the findings obtained by hierarchical regressions, we also conducted multivariate linear regressions replacing the physician global assessment of severity with the nine Medsger disease severity subscales. These analyses confirmed the relationships between the total OHIP score and the SF-36 MCS and SF36-PCS (data not shown).

**TABLE 2** Linear regression analysis showing the relationship between OHIP and SF-36 MCS and PCS scores

	MCS			PCS		
	$\beta$	95% CI	<i>P</i> -value	$\beta$	95% CI	<i>P</i> -value
Age, years	0.14	−0.04, 0.32	0.125	−0.18	−0.35, −0.01	<b>0.036</b>
Female vs male	−0.91	−6.69, 4.88	0.757	−0.46	−6.01, 5.09	0.870
White	1.42	−5.22, 8.06	0.673	6.42	0.05, 12.78	<b>0.048</b>
Education (>high school)	3.14	−0.33, 6.61	0.075	0.30	−3.03, 3.62	0.859
Current smoker	−3.84	−9.77, 2.08	0.202	−2.91	−8.59, 2.77	0.313
Disease duration, years	−0.04	−0.25, 0.16	0.681	−0.09	−0.28, 0.11	0.376
Mean physician global assessment of severity (range 0–10)	−0.23	−1.02, 0.56	0.568	−0.89	−1.64, −0.13	<b>0.021</b>
Total OHIP score	−0.11	−0.16, −0.06	<b>&lt;0.001</b>	−0.08	−0.13, −0.03	<b>0.002</b>

Significant *P*-values are highlighted in bold. For MCS,  $R^2$  model = 16.6%, total OHIP score accounted for 9.7% of the total  $R^2$ . For PCS,  $R^2$  model = 16.5%, total OHIP score accounted for 5.6% of the total  $R^2$ . Significant *P*-values are highlighted in bold. MCS: mental component summary; OHIP: Oral Health Impact Profile; PCS: physical component summary; SF-36: 36-item Short Form Health Survey.

## Discussion

We have previously shown in a study of 163 SSc patients compared with 231 controls that oral HRQoL was substantially reduced in SSc patients compared with controls (mean OHIP score: SSc, 41.6; controls, 26.7) [2]. In the current study we found that OHIP scores were associated with the SF-36 MCS and PCS and, after adjusting for possible confounders, predicted 9.7% and 5.6% of the variance of the MCS and PCS scores, respectively. One previous study of oral HRQoL in SSc assessed the association between the Mouth Handicap in Systemic Sclerosis (MHSS) scale with global HRQoL in 40 subjects and did not find any association between the two [4], perhaps because of the smaller sample size and a different measure of oral HRQoL.

Oral disorders may impact daily living through work loss, reduction in social interaction, disruption of family life and dietary restriction. Consequently, subjective oral health status indicators have been developed to capture the effects of oral disease on quality of life. The OHIP [1], one of these measures, has excellent measurement properties, is sensitive to change and is available in several languages [10]. Scores show small to moderate correlations with a wide range of traditional clinical indicators and self-perceived oral conditions, such as xerostomia [11], caries [12], periodontal disease [13] and number of missing teeth [14]. Thus, although it has not yet been compared with the MHSS, a recently developed [4] specific SSc oral HRQoL measure, there is strong rationale to use it in SSc. On the other hand, the OHIP has not been specifically validated in SSc. It may have been preferable to use an SSc-specific instrument in our study, but it was not available when we started to collect our data. Also, it has not been validated in any other oral conditions and thus using it in normal controls, as we were required to do in the initial controlled study [2], may have been questionable. The fact that in our first report [2] we found a large difference in OHIP scores between SSc subjects and

controls along with significant differences in saliva production, oral aperture, number of missing teeth and extent of periodontal disease suggests that the OHIP is also a valid measure of oral HRQoL in this population.

There is some evidence from non-SSc studies that oral HRQoL is associated with global HRQoL. This issue was systematically reviewed in 2006 [5]. Four of seven studies reviewed showed significant associations between oral health status and global HRQoL. A search of more recent literature published after 2004 identified a few additional studies that demonstrated an association between oral and global HRQoL [15, 16].

The observation that certain disease parameters such as the physician global assessment of disease severity and several of the Medsger disease severity scales are related to the SF-36 but not to OHIP scores lends support to the concept that those aspects of the disease that we have chosen to assess either semi-quantitatively (Medsger scales) or subjectively (physician global assessments) do not include oral issues that are important to the patient in terms of quality of life. It also suggests that oral HRQoL may represent an additional dimension of HRQoL that is not captured by traditional HRQoL measures such as the SF-36. This is supported by the fact that despite the association between measures of oral HRQoL and global HRQoL in other non-SSc studies, the OHIP has been found to be a better instrument than the SF-36 for its ability to discriminate between various oral health states [15, 17].

This study is not without limitations. First, the cross-sectional design of the study precludes determination of causality. Indeed, it is possible that an underlying construct, such as depression, may lead to worse self-report on multiple measures, including both the OHIP and the SF-36. Second, our study subjects had long disease duration and more than two-thirds had lcSSc. Our findings are thus generalizable only within the sampling frame of this study. Also, as mentioned above, the OHIP has not been validated specifically in SSc.

The strengths of this study include the large, multisite sample of SSc subjects, the assessment of both oral and global HRQoL at the same time, the concurrent assessment and adjustment for disease characteristics such as severity and the robust statistical analysis, making it the most definitive study of oral HRQoL in SSc to date.

In summary, we found that oral HRQoL in SSc, measured with the OHIP, was independently associated with global HRQoL. However, oral HRQoL was not related to various measures of disease severity. This suggests that physicians, in their assessment of global disease severity in SSc, are disregarding issues related to oral health. Given the impact of poor oral health on HRQoL, physicians caring for SSc patients should pay more attention to oral health, as has been previously suggested [18], as interventions to improve oral health in SSc have the potential to improve overall HRQoL [19, 20].

#### Rheumatology key messages

- Oral health-related quality of life (HRQoL) and global health-related quality of life are impaired in SSc.
- There is a significant association between oral HRQoL and global HRQoL in SSc.
- Oral HRQoL is not captured by physician assessment of disease severity in SSc.

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### References

- 1 Slade GD. The Oral Health Impact Profile (OHIP). In: Slade GD, ed. *Measuring Oral Health and Quality of Life*. Chapel Hill, NC, USA: Department of Dental Ecology, School of Dentistry, University of North Carolina, 1997:93–104.
- 2 Baron M, Hudson M, Tatibouet S *et al.* The Canadian systemic sclerosis oral health study: orofacial manifestations and oral health-related quality of life in systemic sclerosis compared with the general population. *Rheumatology* 2014;53:1386–94.
- 3 Mouthon L, Rannou F, Berezne A *et al.* Development and validation of a scale for mouth handicap in systemic sclerosis: the Mouth Handicap in Systemic Sclerosis scale. *Ann Rheum Dis* 2007;66:1651–5.
- 4 Maddali Bongi S, Del Rosso A, Miniati I *et al.* The Italian version of the Mouth Handicap in Systemic Sclerosis scale (MHISS) is valid, reliable and useful in assessing oral health-related quality of life (OHRQoL) in systemic sclerosis (SSc) patients. *Rheumatol Int* 2012;32:2785–90.
- 5 Naito M, Yuasa H, Nomura Y *et al.* Oral health status and health-related quality of life: a systematic review. *J Oral Sci* 2006;48:1–7.
- 6 Clements P, Lachenbruch P, Siebold J *et al.* Inter and intraobserver variability of total skin thickness score (modified Rodnan TSS) in systemic sclerosis. *J Rheumatol* 1995;22:1281–5.
- 7 LeRoy EC, Black C, Fleischmajer R *et al.* Scleroderma (systemic sclerosis): classification, subsets and pathogenesis. *J Rheumatol* 1988;15:202–5.
- 8 Medsger TA Jr, Bombardieri S, Czirjak L *et al.* Assessment of disease severity and prognosis. *Clin Exp Rheumatol* 2003;21(Suppl 29):S42–6.
- 9 Khanna D, Furst DE, Clements PJ *et al.* Responsiveness of the SF-36 and the Health Assessment Questionnaire Disability Index in a systemic sclerosis clinical trial. *J Rheumatol* 2005;32:832–40.
- 10 Slade GD, Spencer AJ. Development and evaluation of the oral health impact profile. *Community Dent Health* 1994; 11:3–11.
- 11 Baker SR, Pankhurst CL, Robinson PG. Utility of two oral health-related quality-of-life measures in patients with xerostomia. *Community Dent Oral Epidemiol* 2006;34: 351–62.
- 12 Busato IM, Ignacio SA, Brancher JA, Moyses ST, Azevedo-Alanis LR. Impact of clinical status and salivary conditions on xerostomia and oral health-related quality of life of adolescents with type 1 diabetes mellitus. *Community Dent Oral Epidemiol* 2012;40:62–9.
- 13 Ng SK, Leung WK. Oral health-related quality of life and periodontal status. *Community Dent Oral Epidemiol* 2006; 34:114–22.
- 14 Lawrence HP, Thomson WM, Broadbent JM, Poulton R. Oral health-related quality of life in a birth cohort of 32-year olds. *Community Dent Oral Epidemiol* 2008;36: 305–16.
- 15 Lee IC, Shieh TY, Yang YH, Tsai CC, Wang KH. Individuals' perception of oral health and its impact on the health-related quality of life. *J Oral Rehabil* 2007;34:79–87.
- 16 Jones JA, Kressin NR, Kazis LE *et al.* Oral conditions and quality of life. *J Ambul Care Manage* 2006;29:167–81.
- 17 Allen PF, McMillan AS, Locker D. An assessment of sensitivity to change of the Oral Health Impact Profile in a clinical trial. *Community Dent Oral Epidemiol* 2001;29: 175–82.
- 18 Alantar A, Cabane J, Hachulla E *et al.* Recommendations for the care of oral involvement in patients with systemic sclerosis. *Arthritis Care Res* 2011;63:1126–33.
- 19 Yuen HK, Weng Y, Bandyopadhyay D *et al.* Effect of a multi-faceted intervention on gingival health among adults with systemic sclerosis. *Clin Exp Rheumatol* 2011; 29(Suppl 65):S26–32.
- 20 Yuen HK, Marlow NM, Reed SG *et al.* Effect of orofacial exercises on oral aperture in adults with systemic sclerosis. *Disabil Rehabil* 2012;34:84–9.