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Original article

The Canadian systemic sclerosis oral health study: orofacial manifestations and oral health-related quality of life in systemic sclerosis compared with the general population

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Abstract

Objective. The aim of this study was to compare oral abnormalities and oral health-related quality of life (HRQoL) of patients with SSc with the general population.

Methods. SSc patients and healthy controls were enrolled in a multisite cross-sectional study. A standardized oral examination was performed. Oral HRQoL was measured with the Oral Health Impact Profile (OHIP). Multivariate regression analyses were performed to identify associations between SSc, oral abnormalities and oral HRQoL.

Results. We assessed 163 SSc patients and 231 controls. SSc patients had more decayed teeth (SSc 0.88, controls 0.59, $P = 0.0465$) and periodontal disease [number of teeth with pocket depth (PD) >3 mm or clinical attachment level (CAL) $\geqslant 5.5$ mm; SSc 5.23, controls 2.94, $P < 0.0001$]. SSc patients produced less saliva (SSc 147.52 mg/min, controls 163.19 mg/min, $P = 0.0259$) and their interincisal distance was smaller (SSc 37.68 mm, controls 44.30 mm, $P < 0.0001$). SSc patients had significantly reduced oral HRQoL compared with controls (mean OHIP score: SSc 41.58, controls 26.67, $P < 0.0001$). Multivariate regression analyses confirmed that SSc was a significant independent predictor of missing teeth, periodontal disease, interincisal distance, saliva production and OHIP scores.

Conclusion. Subjects with SSc have impaired oral health and oral HRQoL compared with the general population. These data can be used to develop targeted interventions to improve oral health and HRQoL in SSc.

Key words: systemic sclerosis, quality of life, oral health, dental caries, periodontal disease, Sjögren's syndrome, tooth loss.

CLINICAL SCIENCE

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Introduction

Oral abnormalities are common in SSc [1] but may be overshadowed by other serious systemic symptoms. The most frequent oral problems include dental (decayed, filled and missing teeth), periodontal and orofacial (microstomia, xerostomia and bone resorption of the mandible) abnormalities [2–4]. Studies of oral health in SSc have been performed with small samples, often without appropriate controls. Oral health-related quality of life (HRQoL) in SSc has not been robustly estimated [2–5]. The aim of this study was to assess the oral abnormalities and oral HRQoL of a large sample of SSc subjects and compare these to general population controls using a standardized oral examination and a validated measure of oral HRQoL, the Oral Heath Impact Profile (OHIP) [6]. We hypothesized that SSc subjects would have more oral abnormalities on clinical exam and worse oral HRQoL compared with the general population.

Methods

Study design and subjects

This multisite, cross-sectional study was conducted between 2008 and 2011. The research ethics board of each participating centre approved the study and all study subjects provided informed consent in compliance with the Declaration of Helsinki. All SSc patients were enrolled in the Canadian Scleroderma Research Group (CSRG) registry, had a diagnosis of SSc confirmed by a recruiting rheumatologist, were $\geqslant$18 years of age and were fluent in either English or French. Seven of the 15 CSRG sites took part in the study and eight dentists participated. Prior to recruitment of SSc patients, the participating sites' lists of study subjects were randomly reordered and separated into subgroups of 25 patients. The first 25 patients on the list (subgroup 1) were approached sequentially by the site coordinators until 20 patients agreed to participate. If patients in the first subgroup were all approached and the 20-patient target was not met, patients of the second subgroup were approached in a similar manner. The two largest sites, Montreal, Quebec and London, Ontario had a 40-patient target.

SSc disease duration was measured as the time between the onset of the first non-RP symptoms and the first oral health study visit. lcSSc was defined as skin involvement distal to the elbows and knees, with or without face involvement. dcSSc was defined as skin involvement proximal to the elbows and knees, with or without truncal involvement. SSc global disease severity was rated by the study physicians on a 0–10 numerical rating scale, a valid measure of severity in SSc [7]. Medication use was recorded by study physicians and medications known to be associated with dry mouth, according to the manufacturers' product monographs, were identified (supplementary Table S1, available at *Rheumatology* Online). Controls were recruited by site coordinators from the same health centres as SSc patients. Controls were patients consulting for mechanical joint disease (e.g. OA) and were sampled such that they were of similar sex and age as the spectrum of SSc patients.

Outcome measures

Three sets of outcomes were identified: (i) dental (decayed, filled, missing teeth) and periodontal abnormalities, (ii) orofacial abnormalities commonly associated with SSc (i.e. interincisal distance and saliva production), and (iii) oral HRQoL. During dental and periodontal examinations the number of decayed, missing and filled teeth was noted. Decay was determined as severe caries (less than one-third of the crown remaining or progression halfway or more through the root), pit and fissure and/or smooth surfaces caries. Filled teeth included those with amalgam or other material. Wisdom teeth were not examined.

Periodontal disease was determined by assessing periodontal pocket depth (PD) and clinical attachment level (CAL) (Fig. 1). PD is the distance from the gingival margin to the base of the gingival sulcus [8]. It was measured in millimetres with a Williams probe (Hu-Friedy 945188PW) at six sites (mesio-buccal, mid-buccal, disto-buccal, mesio-lingual, mid-lingual, disto-lingual) [9]. All teeth were measured twice for PD. When two PD values differed by $>$2 mm, the measurement was performed a third time and the two closest values were retained. The PD values at each site were averaged. The CAL is the distance on the buccal or labial surface from the cemento-enamel junction to the base of the sulcus measured using a Williams probe [8]. This was measured once for each tooth. The presence of periodontal disease in a given tooth was defined as either a PD $>$3 mm or a CAL $\geqslant$5.5 mm [8, 10]. The extent of periodontal disease in the entire mouth was calculated as the number of teeth with either a PD $>$3 mm or a CAL $\geqslant$5.5 mm. Mouth aperture was originally evaluated, but results were not consistent for each site, therefore these data were not included.

Interincisal distance was assessed at the beginning of the examination to avoid bias induced by prolonged stretching of the mouth. Subjects were asked to open their mouths as wide as they could and the interincisal distance was measured as the distance between the incisal edges of the lower central tooth and the upper central tooth [11]. The same was done for edentulous patients, with dentures in the mouth.

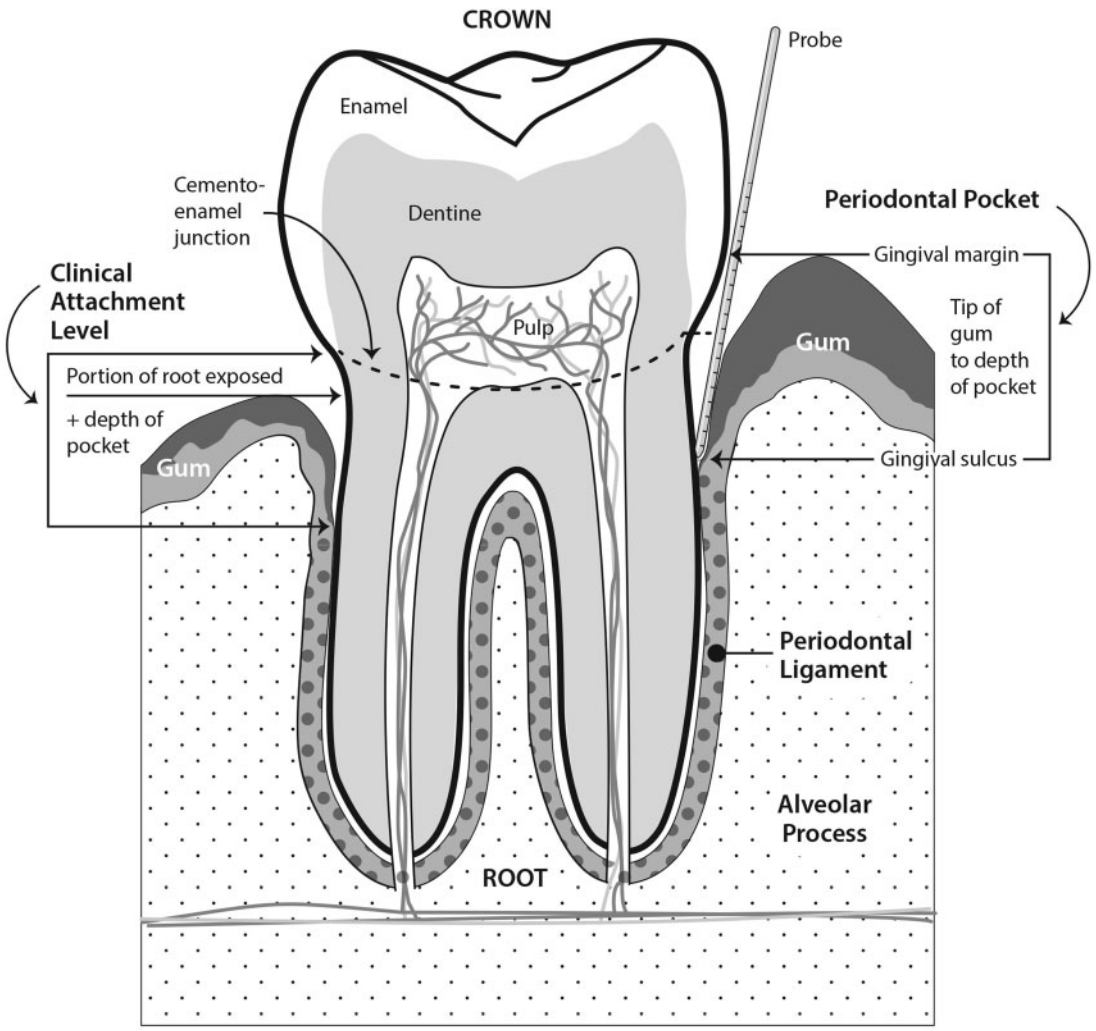
Xerostomia

The Saxon test was used to determine the subject's unstimulated saliva production in milligrams per minute [12]. Briefly, subjects were asked to keep a piece of preweighed gauze in their mouths for 1 min. They were instructed to move it around but not to chew or swallow. The gauze was then reweighed to determine the quantity of unstimulated saliva present in the mouth.

Oral HRQoL

The OHIP-49 was chosen because it is the most widely used and investigated oral HRQoL instrument [6, 13–21]. It shows acceptable stability across a 3-month test–retest

Fig. 1 Anatomy of a tooth



Pocket depth (PD) is the distance from the top of the gingival margin to the bottom of the gingival sulcus. In a normal tooth the periodontal ligament is attached to the root at the cemento-enamel junction. Cementum is the bony covering of the root. PD is increased because inflammation of the gingiva elevates the gingival margin and/or the attachment of the periodontal ligament to the cemento-enamel junction is destroyed as the alveolar process is resorbed. CAL is the distance from the cemento-enamel junction to the bottom of the gingival sulcus. CAL cannot be assessed if the cemento-enamel junction is not exposed. It reflects bone resorption, an effect of periodontal disease. Either an abnormal PD or an abnormal CAL reflect periodontal disease, although different stages in the disease process.

period with an intraclass coefficient (ICC) ranging from 0.50 to 0.86 [22]. This instrument consists of 49 questions on the frequency of a number of adverse oral problems, such as toothache, mouth pain, difficulty chewing or pronouncing and discomfort related to appearance. Respondents are asked to indicate how frequently they have experienced each problem in their daily life on a five-point scale coded from 4 (very often) to 0 (never or not applicable). Three questions pertain only to denture wearers and have a not-applicable option. Six subscales are calculated: functional limitation, physical pain, psychological discomfort, physical disability, psychological disability, social disability and handicap [6]. The OHIP revealed levels of dysfunction, discomfort and disability in various populations that are consistent with clinical conditions and dental care accessibility [6]. Edentulous patients have increased levels of functional and physical limitation [6]. Higher scores are noted for physical pain, functional limitation and physical disability in denture wearers [6]. There are several methods of scoring the OHIP, including additive and weighted scores [6, 23]. Since addition of raw scores has been shown to perform well and is simpler [24, 25], we used this approach for scoring. Summed OHIP scores range from 0 to 196, with higher scores indicating worse oral HRQoL.

Calibration workshop

Prior to the start of the study, eight dentists, including the primary investigator (M.G.), participated in a 1-day calibration workshop. Eight patients with SSc volunteered to participate in the workshop. M.G. instructed the dentists on the examination protocol. Each of the dentists then examined two randomly assigned SSc patients. The sequence for each exam was Saxon test, interincisal distance, PD, CAL, missing teeth, filled teeth, second Saxon, second interincisal distance and second PD. Decayed teeth cannot be reliably visually noted and require radiographic assessment. This was done and will be the subject of future analyses. Written and instructional Power Point presentations were provided to all dentists to ensure compliance with the study protocol.

Statistical analysis

Descriptive statistics were used to summarize the demographic characteristics, oral characteristics and OHIP scores of the SSc subjects and controls. Chi-squared tests, Fisher's exact tests and Mann–Whitney *U* tests were used, as appropriate. Intrarater reliability was calculated for saliva production and interincisal distance. Filled and missing teeth were not checked twice because these are straightforward to measure and the exams were already quite long. In addition, since PD values were the mean of two measurements, intrarater reliability for the number of teeth with a PD >3 mm was not calculated because this would have meant performing this exam four times by two dentists on each subject and would have been too onerous for the subjects. Interrater reliability was calculated for saliva production, interincisal distance, number of teeth with a PD >3 mm or a CAL $\geqslant 5.5$ mm, number of missing teeth and number of filled teeth. Intra- and interrater reliability were measured with ICCs because the variables of interest were continuous. The ICCs were calculated using the variance component estimates of linear and generalized linear mixed models. The ICCs were interpreted using the following guidelines: 0 = poor, 0.01–0.20 = slight, 0.21–0.40 = fair, 0.41–0.60 = moderate, 0.61–0.80 = substantial and 0.81–1 almost perfect [26].

Linear mixed models for continuous outcome data and generalized linear mixed models using negative binomial distribution for count data were used to examine associations between SSc and outcomes of interest, namely the number of decayed, missing and filled teeth, periodontal disease, saliva production, interincisal distance, OHIP total and subscale scores. Models were adjusted for potential confounders identified from the 2002 annual report of Oral Health U.S. [27], and included age, gender, ethnicity (white *vs* other), education (greater *vs* less than high school) and current smoking. In addition, the number of teeth was added as an offset term variable in the models for the number of decayed and filled teeth and for periodontal disease, since the results for decayed and filled teeth and periodontal disease could be affected by the number of teeth [8, 10, 28]. Edentulous patients were also excluded from these models. The total OHIP scores did not have a normal distribution. Thus all models using the OHIP score were repeated using log-transformed scores and yielded similar results as the models that used raw scores (data not shown). The study site was added as a random effect in all mixed models. *P*-values <0.05 were considered statistically significant. All statistical analyses were performed using SAS version 9.2 (SAS Institute, Cary, NC, USA).

Results

Calibration workshop

The intrarater reliability for saliva production and interincisal distance were substantial (ICC = 0.64) and almost perfect (ICC = 0.94), respectively. The interrater reliability was almost perfect for saliva production (ICC = 0.83) and substantial for interincisal distance (ICC = 0.63). The interrater reliability was almost perfect for the number of teeth with a PD >3 mm or a CAL $\geqslant 5.5$ mm (ICC = 0.90) and the number of missing teeth (ICC = 0.99). Interrater reliability for the number of filled teeth was moderate (ICC = 0.49). However, one dentist reported no filled teeth in one quadrant, whereas the second recorded six in that quadrant. We suspect that the first dentist erred in not recording the results for that quadrant. If this patient is removed, the ICC for the number of filled teeth is 0.80.

Study results

Study subjects

A total of 163 SSc patients and 231 controls were included in the study. There were no significant differences in the two groups with regards to age, gender, education, ethnicity and smoking status (Table 1). However, SSc subjects were more likely to be edentulous and to be using drugs associated with dry mouth (supplementary Table S1, available at *Rheumatology* Online).

To assess the generalizability of the study findings, the SSc study subjects were compared with the 1159 non-participating CSRG subjects. Study subjects had a longer disease duration [13.9 years (S.D. 8.5) *vs* 10.9 years (S.D. 9.7), $P < 0.0001$] and more had limited SSc compared with the rest of the CSRG population (72.4% *vs* 63.6%, $P = 0.028$). There was no significant difference between physician global assessments of disease severity between the study subjects and the non-participating CSRG subjects [2.9 (S.D. 2.2) *vs* 2.8 (S.D. 2.3), respectively, $P = 0.3027$).

Oral abnormalities

SSc patients had significantly more decayed teeth (0.88 *vs* 0.59, $P = 0.0465$) and periodontal disease (number of teeth with a PD >3 mm or a CAL $\geqslant 5.5$ mm, 5.23 *vs* 2.94, $P < 0.0001$) compared with controls (Table 2). SSc patients produced less saliva (147.52 mg/min *vs* 163.19 mg/min, $P = 0.0259$) and their interincisal distance was smaller (37.68 mm *vs* 44.30 mm, $P < 0.0001$) compared with controls.

Oral HRQoL

SSc subjects had significantly higher OHIP scores than controls in all subscales of the instrument ($P < 0.01$), as well as in the overall score (mean OHIP scores 41.58 *vs*

Table 1 Baseline characteristics of SSc subjects ($n = 163$) and controls ($n = 231$)

Variable	SSc subjects	Controls	*P*-value
Female, *n* (%)	146 (89.6)	209 (90.5)	0.7669
Age, mean (S.D.), years	56.20 (10.56)	58.01 (10.63)	0.0985
White, *n* (%)	150 (92.6)	207 (89.6)	0.3131
Aboriginal, *n* (%)	9 (5.6)	7 (3.0)	0.2124
Education >high school, *n* (%)	80 (49.4)	135 (58.4)	0.0758
Current smoker, *n* (%)	15 (9.5)	29 (12.7)	0.3268
Edentulous, *n* (%)	21 (12.88)	6 (2.60)	**<0.0001**
Any dentures, *n* (%)	42 (25.77)	44 (19.05)	0.1118
Use of drugs associated with dry mouth, *n* (%)	120 (73.6)	137 (59.3)	**0.0046**

Statistically significant results are shown in bold.

Table 2 Univariate associations between SSc and oral abnormalities and HRQoL

Variable	SSc subjects		Controls		*P*-value
	Mean	S.D.	Mean	S.D.	
Number of decayed teeth[a]	0.88	1.82	0.59	1.75	**0.0465**
Number of filled teeth[a]	11.22	6.08	11.81	5.83	0.2937
Number of missing teeth	7.90	9.44	5.66	7.04	0.1055
Number of teeth with a PD >3 mm or a CAL ⩾5.5 mm[a]	5.23	5.63	2.94	4.11	**<0.0001**
Saliva, mg/min	147.52	95.07	163.19	81.34	**0.0259**
Interincisal distance, mm	37.68	8.36	44.30	6.59	**<0.0001**

	Mean	S.D.	Median	IQR	Mean	S.D.	Median	IQR	*P*-value
Total OHIP score	41.58	32.53	34	16–58	26.67	25.15	19	10–34	**<0.0001**
OHIP subscales									
Functional limitation	10.29	7.39	9	5–16	6.08	5.23	4	2–8	**<0.0001**
Physical pain	10.39	6.46	10	5–15	8.39	5.90	7	4–11	**0.0013**
Psychological discomfort	6.20	5.23	6	1–10	4.29	4.72	3	0–7	**0.0001**
Physical disability	6.68	6.77	4	1–11	2.84	4.33	1	0–4	**<0.0001**
Psychological disability	4.52	5.06	3	0–7	3.17	4.20	2	0–4	**0.0066**
Social disability	1.33	2.74	0	0–1	0.68	1.89	0	0–0	**0.0072**
Handicap	2.17	3.84	0	0–2	1.22	3.01	0	0–1	**0.001**

[a]Excluding completely edentulous patients. Statistically significant results are shown in bold. PD: periodontal pocket depth; IQR: interquartile range.

26.67, $P < 0.0001$), consistent with worse oral HRQoL in SSc (Table 2).

Multivariate analyses

Multivariate regression analyses were performed to assess the relationship of SSc to oral abnormalities, adjusting for possible confounders (Table 3). SSc was a significant independent predictor of the number of missing teeth [relative risk (RR) 1.41, 95% CI 1.11, 1.79] and of periodontal disease (number of teeth with either a PD >3 mm or a CAL ⩾5.5 mm; RR 1.84, 95% CI 1.39, 2.43). SSc was a significant independent predictor of saliva production ($\beta = -20.05$, 95% CI −36.72, −3.37), interincisal distance ($\beta = -7.18$, 95% CI −8.67, −5.69), total OHIP score ($\beta = 13.98$, 95% CI 8.27, 19.68) and all OHIP subscale scores (Table 3).

Sensitivity analyses

More patients with SSc had dentures (Table 2). It is possible that dentures are associated with worse oral HRQoL, thus we repeated the main analysis adjusting for dentures. The results were unchanged, suggesting that dentures were not a significant confounder in the relationship between SSc and oral HRQoL (data not shown). Adjusting for medication associated with dry mouth (supplementary Table S1, available at *Rheumatology* Online) also did not change the results of any of the models (data not shown).

Discussion

This study revealed that patients with SSc have significantly worse dental health, including more missing teeth

Table 3 Multivariate regression analyses to determine the independent associations between SSc and oral abnormalities and HRQoL

Outcome variable	Estimates for SSc	
	RR	95% CI
Number of decayed teeth	1.54	0.87, 2.72[a]
Number of filled teeth	1.01	0.92, 1.12[a]
Number of missing teeth	**1.41**	**1.11, 1.79**[b]
Number of teeth with a PD >3 mm or a CAL ⩾5.5mm	**1.84**	**1.39, 2.43**[a]
	β	95% CI
Saliva, mg/min	**−20.05**	**−36.72, −3.37**[c]
Interincisal distance, mm	**−7.18**	**−8.67, −5.69**[c]
Total OHIP score	**13.98**	**8.27, 19.68**[c]
OHIP subscales		
Functional limitation	**4.09**	**2.85, 5.32**[c]
Physical pain	**1.77**	**0.54, 3.00**[c]
Psychological discomfort	**1.87**	**0.86, 2.88**[c]
Physical disability	**3.73**	**2.63, 4.83**[c]
Psychological disability	**1.24**	**0.31, 2.18**[c]
Social disability	**0.52**	**0.05, 0.98**[c]
Handicap	**0.81**	**0.12, 1.49**[c]

All models were adjusted for age, gender, ethnicity, education and smoking status. Statistically significant results are in bold. [a]Generalized linear mixed models (negative binomial distribution) using the number of teeth as an offset term excluding completely edentulous patients). [b]Generalized linear mixed models (negative binomial distribution). [c]Linear mixed models. PD: periodontal pocket depth.

and more periodontal disease. They are also more likely to have decreased saliva production and decreased oral opening. Finally, oral HRQoL of SSc patients was significantly impaired overall and in all seven subscales of the OHIP as noted in Table 2. This important latter finding has not been previously reported in SSc. Implications from these results will allow health care professionals and researchers to focus on specific remedies to improve psychological and physical limitations associated with SSc [29, 30].

Our study thus provides robust data to confirm previous findings from small studies of dental and periodontal disease in SSc. Table 4 summarizes the results of other studies reporting oral findings in at least 15 SSc subjects compared with a normal control sample. None of the studies specified who performed the examinations. All oral examinations for the current study were performed by dentists who had participated in a prior calibration session. These previous studies are limited by small sample sizes. The total number of SSc subjects and controls included in all four controlled studies were 141 and 125, respectively, whereas the current multisite study included a total of 163 SSc subjects and 231 controls. In one of the four studies there were only seven controls [2]. Furthermore, multivariate regression analyses to adjust

Table 4 Comparison of previous oral health studies

Study	Sample size, *n*	Age, years	Number of decayed teeth	Number of filled teeth	Number of missing teeth	Pocket depth, mm	CAL, full mouth, mm	Salivary flow, ml/min	Interincisal distance, mm
Wood *et al.* [3]	SSc: 31	50.3 (12.2)	3.2 (4.3)[a]	NR	NR	PD score: **10.1 (8.9)***	NR	NR	**34.2 (8.1)***
				DMF-S: 95 (37)**					
	Controls: 30	Age matched	1.8 (2.6)	NR	NR	PD score: 3.9 (5.3)	NR	NR	46.4 (3.8)
				DMF-S: 77 (35)					
Nagy *et al.* [2]	SSc: 32	49.3 (11.6)		DMF-T: 21.9 (6.5)[a]		2.8 (0.7)[a]	NR	0.2 (0.04)[a]	**36.8 (9)***
	Controls: 17	45.2 (15.2)		DMF-T: 18.7 (6.1)		2.4 (0.8)	NR	0.26 (0.06)	41.9 (6.4)
Chu *et al.* [30]	SSc: 42	54.0 (12.2)	2.1 (2.4)[a]	4.3 (4.2)[a]	4.1 (5.5)[a]	>3 mm: 76%[a]	NR	**0.18 ±(0.17)*****	**40.1 (6.5)****
	Controls: 42	Age matched	1.5 (2.5)	5.2 (4.2)	4.9 (5.5)	>3 mm: 55%	NR	0.31 (0.21)	43.6 (7.0)
Leung *et al.* [5]	SSc: 36	50.6 (11.7)	NR	NR	NR	**2.52 (0.58)***	3.19 (0.94)[a]	NR	NR
	Controls: 36	Age matched	NR	NR	NR	1.92 (0.44)	3.17 (1.29)	NR	NR

Unless otherwise indicated, values are expressed as mean (S.D.). [a]Not statistically different from the control group. *$P < 0.001$; **$P < 0.05$; ***$P < 0.01$. Statistically significant results are shown in bold. NR: not reported; DMF-S: number of decayed, missing or filled tooth surfaces; DMF-T number of decayed, missing and filled teeth; PD: periodontal pocket depth. PD scores of 0, 1, or 2 were allocated for gingival sulcus depths of <3 mm, ⩾3 mm but <5 mm, and ⩾5 mm, respectively.

for confounders were not performed in any of the previous studies.

Only two studies [3, 31] examined the number of decayed teeth in SSc patients, and neither showed significant differences. In multivariate analyses we also did not find an excess number of missing teeth in SSc. All studies measuring the number of filled or missing teeth reported similar non-significant results, whereas we did find the number of missing teeth increased. When the number of decayed, filled and missing teeth were collapsed within a single measure (i.e. the DMF score), one of four studies revealed a significant difference between SSc and controls [3]. Because of varying definitions of periodontal disease, it is difficult to compare the studies either to each other or to ours. However, two of four studies reported significant abnormalities in PD and thus more periodontal disease [3, 5]. Our study revealed significantly more periodontal disease in SSc than in controls. Salivary flow was assessed in two studies [2, 31], with one reporting a significant difference [31] and the other not, whereas we noted a significant difference. Estimates of the prevalence of clinical xerostomia in SSc vary from 22% to 70% based on mostly small studies [2, 3, 32, 33]. SS may co-exist with SSc [33, 34] and the xerostomia may certainly be due to that condition. This will be further investigated in the future in studies concerned with the relationship between the clinical and serological features of SSc and oral abnormalities.

Interincisal distance was reported in three studies, all of which reported a significant reduction similar to our study [2, 3, 31]. The average interincisal distance of our SSc patients was 37.7 mm (S.D. 8.4), which is consistent with previous findings [2, 3, 31, 35]. The current study is the largest and most comprehensive study to date. The fact that at least some of the previous small studies found an increased DMF, more periodontal disease, decreased saliva and decreased interincisal distance lends support to our findings.

Although it is known that overall HRQoL is impaired in SSc [36], this study is the first to demonstrate impaired oral HRQoL. Various measures have been developed to assess oral HRQoL using self-administered questionnaires [37, 38]. The OHIP [6] has excellent measurement properties. It can distinguish between dentate and edentulous individuals, and correlates with traditional clinical indicators such as xerostomia [39], caries [40], periodontal disease [41] and the number of missing teeth [42]. It thus appears to be a reasonable instrument to use in cross-sectional and longitudinal studies in SSc [24, 25, 43]. The minimal important difference (MID) has been estimated to be 6 points (95% CI 2, 9) [14]. The OHIP scores for the controls in this study were similar to those previously reported in unselected populations and in subjects with corrected oral abnormalities [44, 45]. More importantly, the OHIP scores of the SSc subjects were 15 points higher than those of the controls, well above the MID, indicating worse oral HRQoL in SSc.

There are some limitations to this study. By chance, our study subjects had longer disease duration and proportionately more lcSSc than the general SSc population. Also, SSc is well known to overlap with other autoimmune diseases [46], including SS [47, 48], hypothyroidism and inflammatory myositis, which could conceivably have a relationship with oral health. Future studies are planned to explore the clinical and serological correlates of oral health in SSc, including overlap disease, as well as hand problems, gastro-oesophageal reflux, overall disease severity and autoantibodies.

A disease-specific oral health instrument, the Mouth Handicap in SSc Scale (MHISS), was developed and validated by Mouthon *et al*. in 2007 [49]. Unfortunately it was developed after this study was designed and funded and was not included in our data collection protocol. Nonetheless, the MHISS has several advantages over the OHIP as it is disease specific. Thus future research in oral health of SSc should consider using both the generic (OHIP) and specific (MHISS) instruments.

The strengths of this study include the large, multisite sample of SSc subjects and the comparison with a large control group, as well as the robust statistical analysis, making it the definitive study of oral health in SSc to date [4–6, 21]. Future reports will examine the radiological findings in our cases compared with controls and will explore the relationship between multiple aspects of SSc and oral abnormalities.

In conclusion, SSc subjects have more missing teeth, more periodontal disease, less saliva production, smaller interincisal distance and poorer oral HRQoL than controls. These data can be used to develop targeted interventions to improve oral health and HRQoL in SSc. We recommend the use of adaptive devices such as flossers, powered oscillating-rotating toothbrushes and orofacial exercise to improve oral health [29]. Aesthetics represented by facial changes may be ameliorated by various home-based exercise programmes. These include mouth stretching, connective tissue massage, Kabat's technique and a kinesitherapy programme, which may improve mouth functioning and opening [50].

Rheumatology key messages

- SSc patients have significantly impaired oral health-related quality of life (HRQoL).
- SSc subjects have decreased saliva production and interincisal distance with more dental and periodontal disease.
- These data can help develop targeted interventions to improve oral health and HRQoL in SSc.

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Supplementary data

Supplementary data are available at *Rheumatology* Online.

References

1 Albilia JB, Lam DK, Blanas N *et al*. Small mouths. Big problems? A review of scleroderma and its oral health implications. J Can Dent Assoc 2007;73:831–6.

2 Nagy G, Kovacs J, Zeher M *et al*. Analysis of the oral manifestations of systemic sclerosis. Oral Surg Oral Med Oral Pathol 1994;77:141–6.

3 Wood RE, Lee P. Analysis of the oral manifestations of systemic sclerosis (scleroderma). Oral Surg Oral Med Oral Pathol 1988;65:172–8.

4 Vincent C, Agard C, Barbarot S *et al*. [Orofacial manifestations of systemic sclerosis: a study of 30 consecutive patients]. Rev Stomatol Chir Maxillofac 2010;111:128–34.

5 Leung WK, Chu CH, Mok MY *et al*. Periodontal status of adults with systemic sclerosis: case-control study. J Periodontol 2011;82:1140–5.

6 Slade GD. The Oral Health Impact Profile (OHIP). In: Slade GD, ed. Measuring Oral Health and Quality of Life. Chapel Hill, NC: University of North Carolina: Department of Dental Ecology, 1997:93–104.

7 Hudson M, Steele R, Canadian Scleroderma Research Group. In: Baron M. Update on indices of disease activity in systemic sclerosis. Semin Arthritis Rheum 2007;37: 93–8.

8 Page RC, Eke PI. Case definitions for use in population-based surveillance of periodontitis. J Periodontol 2007; 78(Suppl):1387–99.

9 Buduneli E, Aksoy O, Kose T *et al*. Accuracy and reproducibility of two manual periodontal probes. An in vitro study. J Clin Periodontol 2004;31:815–9.

10 Savage A, Eaton KA, Moles DR *et al*. A systematic review of definitions of periodontitis and methods that have been used to identify this disease. J Clin Periodontol 2009;36: 458–67.

11 Cox SC, Walker DM. Establishing a normal range for mouth opening: its use in screening for oral submucous fibrosis. Br J Oral Maxillofac Surg 1997;35:40–2.

12 Kohler PF, Winter ME. A quantitative test for xerostomia. The Saxon test, an oral equivalent of the Schirmer test. Arthritis Rheum 1985;28:1128–32.

13 Allen PF, O'Sullivan M, Locker D. Determining the minimally important difference for the oral health impact profile-20. Eur J Oral Sci 2009;117:129–34.

14 John MT, Reissmann DR, Szentpetery A *et al*. An approach to define clinical significance in prosthodontics. J Prosthodont 2009;18:455–60.

15 John MT, LeResche L, Koepsell TD *et al*. Oral health-related quality of life in Germany. Eur J Oral Sci 2003;111: 483–91.

16 Allen PF, Locker D. Do item weights matter? An assessment using the oral health impact profile. Community Dent Health 1997;14:133–8.

17 Kieffer JM, Hoogstraten J. Item-order effects in the Oral Health Impact Profile (OHIP). Eur J Oral Sci 2008;116: 245–9.

18 Kieffer JM, Verrips GH, Hoogstraten J. Instrument-order effects: using the oral health impact profile 49 and the Short Form 12. Eur J Oral Sci 2011;119:69–72.

19 Reissmann DR, John MT, Schierz O. Influence of administration method on oral health-related quality of life assessment using the oral health impact profile. Eur J Oral Sci 2011;119:73–8.

20 John MT, Reissmann DR, Schierz O *et al*. No significant retest effects in oral health-related quality of life assessment using the oral health impact profile. Acta Odontol Scand 2008;66:135–8.

21 Allison P, Locker D, Jokovic A *et al*. A cross-cultural study of oral health values. J Dent Res 1999;78:643–9.

22 Yuen HK, Nelson SL. Test–retest reliability of oral health impact profile (OHIP-49) in adults with systemic sclerosis. Spec Care Dentist 2013;33:1–7.

23 Slade GD, Spencer AJ, Locker D *et al*. Variations in the social impact of oral conditions among older adults in South Australia, Ontario, and North Carolina. J Dent Res 1996;75:1439–50.

24 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.

25 Locker D. Health outcomes of oral disorders. Int J Epidemiol 1995;24(Suppl 1):S85–9.

26 Landis JR, Koch GG. The measurement of observer agreement for categorical data. Biometrics 1977;33: 159–74.

27 Dental Oral and Craniofacial Data Resource Center. Oral Health US, 2002. Bethesda, MD: U.S. Department of Health and Human Services, 2002.

28 Gelman A, Hill J. Poisson regression, exposure, and overdispersion. Data Analysis Using Regression & Multilevel/Hierarchical Models. Cambridge: Cambridge University Press, 2007:110–2.

29 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.

30 Chu CH, Yeung CM, Lai IA *et al*. Oral health of Chinese people with systemic sclerosis. Clin Oral Investig 2011;15: 931–9.

31 Andonopoulos AP, Drosos AA, Skopouli FN *et al*. Sjogren's syndrome in rheumatoid arthritis and progressive systemic sclerosis. A comparative study. Clin Exp Rheumatol 1989;7:203–5.

32 Avouac J, Sordet C, Depinay C *et al*. Systemic sclerosis-associated Sjogren's syndrome and relationship to the limited cutaneous subtype: results of a prospective study

of sicca syndrome in 133 consecutive patients. Arthritis Rheum 2006;54:2243–9.

33 Salliot C, Mouthon L, Ardizzone M *et al*. Sjogren's syndrome is associated with and not secondary to systemic sclerosis. Rheumatology 2007;46:321–6.

34 Poole J, Conte C, Brewer C *et al*. Oral hygiene in scleroderma: the effectiveness of a multi-disciplinary intervention program. Disabil Rehabil 2010;32:379–84.

35 Hudson M, Thombs BD, Steele R *et al*. Quality of life in patients with systemic sclerosis compared to the general population and patients with other chronic conditions. J Rheumatol 2009;36:768–72.

36 Locker D, Slade G. Oral health and the quality of life among older adults: the oral health impact profile. J Can Dent Assoc 1993;59:830–3, 837–8, 844.

37 McGrath C, Bedi R. Measuring the impact of oral health on life quality in two national surveys – functionalist versus hermeneutic approaches. Community Dent Oral Epidemiol 2002;30:254–9.

38 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.

39 Busato IM, Ignacio SA, Brancher JA *et al*. 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.

40 Ng SK, Leung WK. Oral health-related quality of life and periodontal status. Community Dent Oral Epidemiol 2006; 34:114–22.

41 Lawrence HP, Thomson WM, Broadbent JM *et al*. Oral health-related quality of life in a birth cohort of 32-year olds. Community Dent Oral Epidemiol 2008;36: 305–16.

42 Slade GD, Spencer AJ. Development and evaluation of the oral health impact profile. Community Dent Health 1994; 11:3–11.

43 Mehl C, Kern M, Freitag-Wolf S *et al*. Does the Oral Health Impact Profile questionnaire measure dental appearance? Int J Prosthodont 2009;22:87–93.

44 Mason J, Pearce MS, Walls AW *et al*. How do factors at different stages of the lifecourse contribute to oral-health-related quality of life in middle age for men and women? J Dent Res 2006;85:257–61.

45 Hudson M, Rojas-Villarraga A, Coral-Alvarado P *et al*. Polyautoimmunity and familial autoimmunity in systemic sclerosis. J Autoimmunity 2008;31:156–9.

46 Fox PC, Bowman SJ, Segal B *et al*. Oral involvement in primary Sjogren syndrome. J Am Dent Assoc 2008;139: 1592–601.

47 Lundstrom IM, Lindstrom FD. Subjective and clinical oral symptoms in patients with primary Sjogren's syndrome. Clin Exp Rheumatol 1995;13:725–31.

48 Soto-Rojas AE, Villa AR, Sifuentes-Osornio J *et al*. Oral manifestations in patients with Sjogren's syndrome. J Rheumatol 1998;25:906–10.

49 Mouthon L, Rannou F, Bérezné A *et al*. Development and validation of a scale for mouth handicap in systemic sclerosis: the mouth handicap in systemic scerosis scale. Ann Rheum Dis 2007;66:1651–5.

50 Maddali-Bongi S, Landi G, Galluccio F *et al*. 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. Rheumatology Int 2011;31:895–901.