Review

Work participation in patients with systemic sclerosis: a systematic review

S. Decuman[1,2], V. Smith[2], S.T.L. Verhaeghe[3], A. Van Hecke[3], F. De Keyser[2]

[1]Department of Internal Medicine,
[2]Department of Rheumatology, and
[3]Department of Public Health,
Faculty of Medicine and Health Sciences,
Ghent University, Belgium.

Saskia Decuman, MSc*
Vanessa Smith, PhD*
Sofie T.L. Verhaeghe, PhD
Ann Van Hecke, PhD
Filip De Keyser, PhD

*These authors contributed equally to the study.

This work should be attributed to:
Department of Rheumatology,
Ghent University Hospital, Belgium.

Please address correspondence to:
Saskia Decuman,
Department of Rheumatology,
Ghent University Hospital 0K12-IB,
De Pintelaan 185,
B-9000 Gent, Belgium.
E-mail: saskia.decuman@ugent.be

Received on October 18, 2013; accepted in revised form on December 11, 2013.

Clin Exp Rheumatol 2014; 32 (Suppl. 86): S206-S213.

Key words: scleroderma, systemic, work, sick leave, disability

Funding: this work was supported by a grant from the 'Nationale Vereniging voor Steun aan Gehandicapte Personen (National association for support to the disabled people, Belgium)' to S. Decuman.

Competing interests: none declared.

ABSTRACT

Objective. *With this systematic review an overview is given of what is known about work participation in patients with systemic sclerosis (SSc).*

Methods. *The databases Pubmed, Cinahl, Nursing and Allied Health and PsychARTICLES have been checked from 1980 onwards. The search string consisted of all combinations of key words for work participation and SSc. Two investigators evaluated the eligibility for the articles. Reference lists were searched for other studies.*

Results. *Eight quantitative and one qualitative study were scrutinised in depth. The percentage of patients not working ranges from 18% to 61%. A meta-analysis of the percentage patients not working was performed and a weight mean of 37% was found. The following parameters are associated with the work variable in multivariate analysis (number of studies in which the variable was independently associated with the work variable/number of studies in which the variable was multivariately assessed): global disability (4/5), health (3/5), educational level (2/4), disease duration (3/3), skin/lung involvement (1/3), age/fatigue/muscle involvement/hand function (1/2) and having a decreased income/race/social support/physically demanding job (1/1). In the qualitative study, management of the work situation, disclosure of limitations at the work force and adaptation of resources in daily life are discussed.*

Conclusion. *Most studies concerning work participation are at this very moment quantitative and cross-sectionally designed. Longitudinal studies are needed to assess causality and qualitative research may be opportune to have a more comprehensive view on the topic of work participation in patients with* SSc.

Introduction

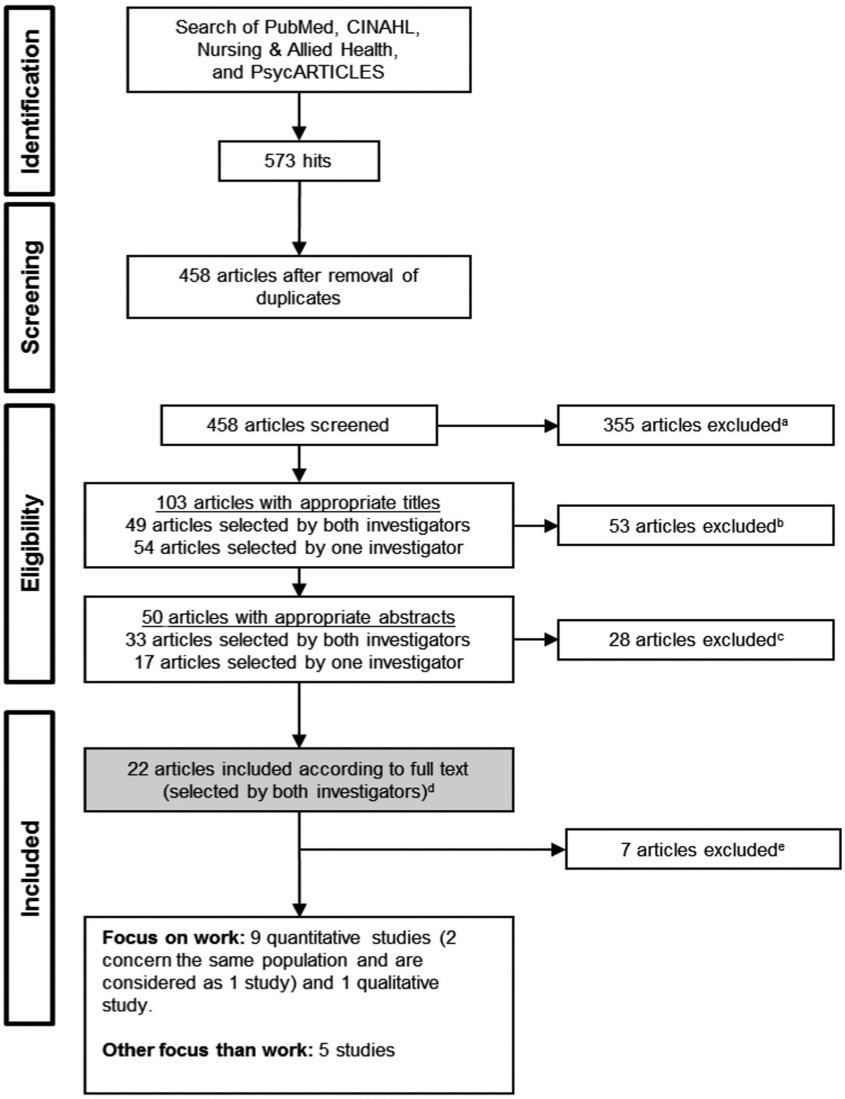
Systemic sclerosis (SSc) is an auto-immune connective tissue disease characterised by abnormal fibrotic processes that can affect multiple organ systems, including the skin, gastro-intestinal tract, lungs, heart and kidney, and can cause immune dysfunction and vascular injury (1). SSc can limit the capacity of performing daily life activities and participation. Consequently patients may have reduced quality of life (2-7). Working is one of those activities of daily living negatively affected by SSc as the disease affects mostly patients in the working age (3-6, 8-19). Not only may patients be unable to work; those still in the labor force can have problems performing their job which both require follow-up (20). Several studies in the field of rheumatology and beyond have already shown the importance of being able to work for the individual (8, 14, 21-27). As different countries have a negative economic situation with high rates of unemployment (and associated costs), keeping people at work is from a society's point of view important (28, 29).

In the last five years there has been a growing number of publications concerning work in patients with SSc. A systematic review may be opportune to provide both quantitative data and qualitative concepts relevant to the field of job participation in patients with SSc and to identify further research questions. Such data may be the backbone for development of interventions and policy guidelines. We therefore performed a systematic literature review to summarise findings on how SSc affects the ability to work and the effects of existing interventions on the ability to stay at or return to work.

Methods

Search strategy

The databases PubMed, Cumulative



[a]When articles were screened on the basis of the title, they are included when the raters have the idea the article is original research about work in patients with SSc. When in doubt the article was selected.
[b]Reasons for exclusion: on the basis of the abstract, the raters decided the article was not original research about work participation (problems) in patients with SSc (n=51) or the study did not include more than 5 patients (n=2). When in doubt the article was selected.
[c]In depth reading of the full text of the articles revealed that the article was not original research, that work participation (problems) was not assessed, or that prevalence rates of not working due to health are not given.
[d]Of those articles the reference list was checked and no additional articles are selected.
[e]Studies do not clearly describe classification criteria.

Fig. 1. Result of the search strategy and selection process.

Index to Nursing and Allied Health Literature (Cinahl), Nursing & Allied Health, and PsycARTICLES were searched for articles in English or French from 1980, the year the preliminary criteria for SSc (scleroderma) were published (30). The search included key words related to work participation as recommended by Haafkens *et al*. (31) combined in all possible combinations with key words and medical subject headings (MeSH) for SSc (see supplementary file 1). Reference lists of the included articles were scrutinised for additional articles.

Selection process

Two investigators (S.D., V.S.) independently evaluated the eligibility of the articles. Articles identified by the database search were selected first according to their titles and in a second step according to their abstracts. At both steps, articles considered eligible by one or both of the investigators were included in the next stage. Finally, the articles were selected based on their full texts. Articles were included in the analysis if they were original studies and addressed one or more of the following issues: prevalence of work participation (problems) and related concepts in patients with SSc; demographics, disease-related, work-related, or other factors associated with work participation and related concepts; interventions developed for patients with SSc related to staying at or returning to work and their components and including ≥5 patients. Studies with mixed patient populations were included if they reported data on patients with SSc separately. Both quantitative and qualitative research designs were allowed. If classification criteria were not clearly described the study was excluded.

Data extraction and analysis

The following data were independently extracted by the two investigators: number of patients, age, sex, educational level, marital status, children, race, diagnostic criteria used in the study, percent of each subset (limited systemic sclerosis [lSSc], limited cutaneous systemic sclerosis [lcSSc] or diffuse cutaneous systemic sclerosis [dcSSc]), disease duration, work variable (definition used, prevalence rate) and associated variables (including how measured).

A forest plot and meta-analysis was performed for the proportion of patients not working (statistical software package metaphor version 1.9-0 [a meta-analysis package for R]) (32).

Quality assessment

No consensus exists in literature about the criteria that have to be assessed in studies and how to score them. Quality assessment of the quantitative studies was based on those used in previous systematic reviews (33-35). The set of criteria used for the qualitative studies are a modified version of a framework developed by researchers of the UK (36). The results of quality assessment can be found in Supplementary file 2.

Results

Results of the literature search

The result of the search strategy and selection process is summarised in Figure 1. A total of 22 articles were selected. Of these, 13 had work as the main focus, of which 10 included patients fulfilling the ACR and/or CREST criteria and/or Leroy/Medsger criteria for SSc (4, 8, 9, 12-16, 18, 19). Nine of these 10 studies, contained quantitative data. Two of these 9 reported results from the same study population and were therefore considered a single study (4, 15). Thus, 8 quantitative studies were scrutinised in depth, including 7 cross-sectional studies (4, 8, 9, 12, 13, 15, 16, 19) and 1 study that was both longitudinal and cross-sectional (18). The characteristics of these studies are summarised in Table I. One article used a qualitative methodology (14). No studies concerning interventions were found. An additional 5 articles were selected that did not have work as main focus but that included relevant data (3, 27, 29, 37, 38). For quality assessment of the studies see Supplementary file 2.

Quantitative studies in which work was the main focus

– Patient characteristics

Patients included in the 8 quantitative studies were 48 to 54 years old and 76% to 100% were women. The educational level, described in 6 of 8 studies, was high for most of the patients (4, 8, 9, 12, 13, 15, 16, 18, 19). Cohabitation was described in 4 studies, and in 3/4, at least 75% lived with a partner (4, 9, 15, 16, 18). The frequency of children at home, described in 3 studies, was between 32% and 46% (4, 9, 15, 16). Race, described in 4 studies, was non-Caucasian for 0% to 53% of the patients (8, 12, 18, 19).

– Disease-related characteristics

Disease classification was described in all studies. Three studies calculated disease duration from the first non-Raynaud's phenomenon, and 1 study described disease duration from the onset of skin involvement (9, 16, 18, 19). In the other 4 studies, disease duration was not defined (4, 8, 12, 13, 15).

– Work variables: prevalence of not working and other work variables

Between 18% and 61% of patients were not working (Table IIA) (4, 8, 9, 12, 13, 15, 16, 18, 19). Figure 2 represents a forest plot based upon a meta-analysis of the prevalence of patients not working. The meta-analysis resulted in an overall percentage of 37% of patients not working. Due to heterogeneity amongst studies it may be opportune to interpret this result with caution.

One study additionally assessed the proportion of patients working at baseline and who became work-disabled at follow-up: after a mean ± SD of 4.4±3.8 years of follow-up, 26.7% (35/131) became work-disabled (18).

The proportion of patients who made work changes (not further defined) since start of SSc, reported in 2 of the 8 quantitative studies, was 31% and 35% (8, 12). Loss of work productivity, described in one of the studies, was 2.5±6.1 days per month of decreased work productivity and 2.2±2.9 days per month of SSc interference with work productivity (19). Working hours were reported in 5 studies: in 4 of the studies, between 14% and 40% reported reduced working hours (4, 9, 12, 15, 16) and in 1 study, the percentage with reduced working hours was not given (18). Changing occupation was reported in 2 studies: in 1 study, patients with SSc changed their occupation 0.6±1.1 times per lifetime after diagnosis (12), and in the other study, 2% of patients changed occupation (9).

– Influence of patient characteristics on work variables

Table IIB summarises the frequency of studies assessing parameters by univariate and multivariate analysis. In Table IIA results of the multivariate analysis are reported. The latter are discussed hereunder.

Four studies assessed the influence of educational level, in 2 of them a significant association was found: one found that patients who had a higher level of education had a lower risk of being work disabled (OR: 0.22, CI: 0.12–0.43, $p<0.001$) (18), and the other found that those unable to work or who missed work at least 1 day per month had a lower educational level ($p=0.01$; $p=0.014$), although the OR was not reported (19). Two studies examined the influence of age, 1 of which found that older patients had a higher risk of being work-disabled (OR: 1.10, CI: 1.05–1.20, $p=0.0006$) [8]. Decreased income was assessed in 1 study and found to be independently associated with being on a disability pension (OR: 8.19, CI: 2.67–25.12, p-value: not mentioned) [12]. Race was also assessed in 1 study. Caucasians have a lower risk on becoming work-disabled at follow-up (OR: 0.46, CI: 0.22–0.96, $p=0.038$) (18).

– Influence of disease-related characteristics on work variables

Three of the 8 quantitative studies examined the influence of disease duration by multivariate analysis. Patients with a longer disease duration were at higher and similar risk of not working (2 studies) (OR: 1.04, CI: not mentioned, $p=0.069$; OR: 1.09, CI: 1.01–1.20, $p=0.03$) or of having made a work transition (OR: 1.01, CI: 1.002–1.022, $p=0.014$) (8, 9, 13). The influence of skin and lung involvement was assessed in 3 studies and of muscle involvement in 2. The following parameters were independently associated with the work variable in a single study: having a DcSSc classification was associated with having stopped working due to illness (OR: 3.72, CI: not mentioned, $p=0.022$), having more severe lung disease as measured by the Medsger severity scale was associated with work disability (OR: 1.50, CI: 1.14–1.99, $p=0.004$) and the presence of myalgia was associated with being on full-time sick leave (OR: 3.19, CI: 1.19–8.57, p-value: not mentioned) (12, 13, 18). Patients with a better diffusing capacity of the lung for carbon monoxide had less risk on becoming work disabled at follow-up (OR: 0.98, CI: 0.97–0.99, $p=0.038$) (18).

– Influence of psychological factors on work variables

Fatigue was assessed in 2 of the 8 quantitative studies. In 1 study, patients with fatigue had higher risk of work disability (OR: 2.18, CI: 1.51-3.14, $p<0.001$), and fatigue was in that study an independent predictor of becoming work disabled at follow-up (OR: 1.96, CI: 1.19-3.25, $p=0.009$) (18).

Table I. Characteristics of the 8 quantitative work-related studies.

Variable	Country	Study design	n	Age y[a]	Female n/N (%)	Educational level n/N (%)	Marital status n/N (%)	Children at home n/N (%)	Race n/N (%)	Classification	Subset n/N (%)	Disease duration
Sandqvist *et al.* (2008)	Sweden	Cross-sectional	44	Median (range): 52 (24–60)	44/44 (100.0)	Compulsory, 8/44 (18.0) Upper secondary, 24/44 (55.0) University, 12/44 (27.0)[c]	Living with a partner, 39/44 (88.6)	19/44 (43.2)	NR	ACR	LcSSc, 44/44 (100)	Median (range): 8 (2–44) y[d]
Ouimet *et al.* (2008)	Canada	Cross-sectional	61	52.00±1.18	52/61 (85.2)	<High (secondary) school, 10/61 (16.3)[e]	NR	NR	NR	ACR or CREST criteria. % ACR: NR	LcSSc, 35/61 (57.3) DcSSc, 26/61 (42.6)	11.02 ± 1.22 y[f]
Nguygen *et al.* (2010)	France	Cross-sectional	87	48.6 ± 8.5	72/87 (82.7)	NR	NR	NR	Caucasian, 87/87 (100.0)	ACR or Leroy/ Medsger criteria % ACR: NR	LSSc, 5/87 (5.7) LcSSc, 52/87 (59.8) DcSSc, 30/87 (34.5)	8.1 ± 6.4 y[d]
Sandqvist *et al.* (2010)	Sweden	Cross-sectional	57	Median (IQR): 58 (47-62)	53/57 (93.0)	Compulsory, 21/57 (36.8) Upper secondary, 23/57 (40.4) University, 13/57 (22.8)[c]	Married/ Cohabiting, 45/57 (78.9)	18/57 (31.6)	NR	ACR	LcSSc, 47/57 (82.4) DcSSc, 10/57 (17.5)	Median (IQR): 14 (9–19) y[g]
Bérezné *et al.* (2011)	France	Cross-sectional	189	54.1 ± 13.3	164/189 (86.8)	NR	NR	NR	Caucasian, 164/187 (87.7) Afr Am, 15/187 (8.0) Asian, 8/187 (4.3)[h]	ACR of Leroy/ Medsger criteria. % ACR: NR	LSSc, 25/179 (14) LcSSc, 76/179 (42) DcSSc, 78/179 (44)[i]	9.3 ± 8.4 y[d]
Sharif *et al.* (2011)	USA	Longitudinal/ cross-sectional	284	48.7 ± 13.2	237/284 (83.5)	≤High school diploma (12th grade), 168/284 (59.2) ≥Associate's degree, 116/284 (40.8)[j]	Married/ marriage-like relationship, 161/284 (56.7)	NR	Caucasian, 133/284 (46.8) Afr Am, 58/284 (20.4); Hispanic, 83/284 (29.2) Other, 10/284 (3.5)	ACR	LcSSc, 122/284 (43) DcSSc, 162/284 (57)	2.5 ± 1.6 y[k]
Singh *et al.* (2012)	USA	Cross-sectional	162	51.8 ± 14.2	131/162 (80.9)	≤Grade 8, 5/160 (3.1) Grade 9–11, 2/160 (1.3) High school graduate, 18/160 (11.3) Some college, 56/160 (35.0) College graduate, 36/160 (22.5) Post-college, 43/160 (26.9)[l,m]	NR	NR	Caucasian, 110/159 (69.2) Afr Am, 10/159 (6.3) Asian, 21/159 (13.2) Am Indian/ Alaskan native, 4/159 (2.5) >1 race, 10/159 (6.3) Unknown, 4/159 (2.5)[n]	ACR	LcSSc, 84/162 (51.9) DcSSc, 67/162 (41.3) Overlap, 11/162 (6.8)	7.6 ± 8.2 y[k]
Decuman *et al.* (2012)	Belgium	Cross-sectional	84	47.8 ± 8.9	64/84 (76.2)	≤Lower secondary: 22/84 (26.2) ≤higher secondary: 42/84 (50.0) Higher education certificate: 20/84 (23.8)[o]	64/84 (76.2)	30/84 (35.7)	NR	ACR and/or Leroy/ Medsger criteria. % ACR: 45.2%	LSSc, 24/84 (28.6) LcSSc, 48/84 (57.1) DcSSc, 12/84 (14.3)	Median (range): 56.50 (0–520) m[k]

ACR: American College of Rheumatology; LcSSc: limited cutaneous systemic sclerosis; DcSSc: diffuse cutaneous systemic sclerosis; NR: not reported; y: year; CREST: Calcinosis-Raynaud-Eosophagus-Sclerodactylia-Teleangictasia; W: working; WD: working-disabled; IQR: Interquartile range; Afr Am: African-American; m: month.

[a] Mean ± standard deviation except where noted; [b] Classification criteria used in the study; [c] Compulsory education = 6–16 years of age; upper secondary education = 16 –18 years of age; university = ≥18 years of age; [d] Calculation of disease duration not defined; [e] 6-18 years of age; [f] Self-reported disease duration, not further defined; [g] Disease duration calculated since the onset of skin involvement; [h] Race is missing for 2 patients; [i] subset is missing for 10 patients; [j] High school diploma and below = 17–18 years of age; Associates degree and above = ≥18 years of age; [k] Disease duration defined by onset of first non-Raynaud disease manifestation; [l] Educational level is missing for 2 patients; [m] Grade 8 or less: 6–14 years of age; grade 9-11: 6–17 years of age; high school graduate: 6–18 years of age; college (some tertiary education): 18-22 years of age, ages vary; college graduate (obtained grade of tertiary education); [n] Race is missing for 3 patients; [o] Maximum lower secondary school = 6–15 years of age; maximum higher secondary school = 6 – 18 years of age; higher education certificate = ≥18 years of age.

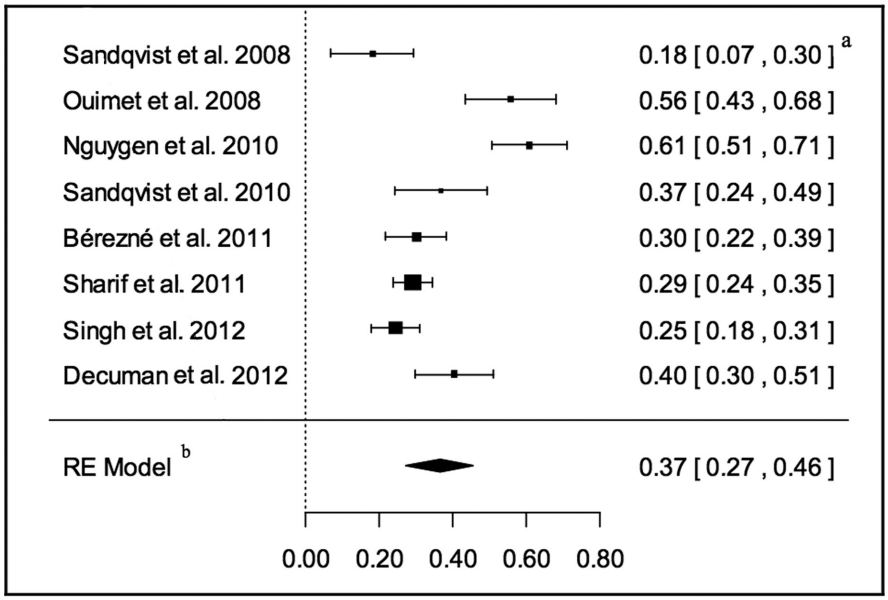
Table IIA. Work variables and significant correlates in the 8 quantitative work-related studies.

Variable	Work variable	Category or measure	Value	Correlates[a]	Odds ratio (95% CI)	*p*-value
Sandqvist *et al*. (2008)	Working ability	No sick leave (working full-time)	21/44 (47.7%)	Not performed	–	–
		Partial sick leave (sick leave 25-50% of a working day)	15/44 (34.0%)			
		Temporarily full-time sick leave[b]	2/44 (4.5%)			
		Full disability pension[b]	6/44 (14.6%)			
	Time spent working	Median time working per day (range), hours	6.25 (0–11) h	Not performed	–	–
		Percent time working	23			
Ouimet *et al*. (2008)	Work disability	Stopped working due to illness[d]	34/61 (55.7%)	Global disability[c]	6.41	<0.001
				Disease duration[d]	1.04	0.069
				LcSSc	3.45	0.024
				DcSSc	3.72	0.022
				Physically demanding job[d]	3.06	0.013
Nguygen *et al*. (2010)	Sick leave status	On full-time sick leave at the time of the study	53/87 (60.9%)	Presence of myalgias[d]	3.19 (1.19, 8.57)	–
				Perceived health status[e]	0.92 (0.88, 0.98)	–
	Disability pension	Officially disabled (receiving a disability pension)	31/87 (35.6%)	Decreased income[d]	8.19 (2.67, 25.12)	–
Sandqvist *et al*. (2010)	Degree of sick leave	No sick leave (working full-time)	16/57 (28.1%)	Not performed	–	–
		Partial sick leave	20/57 (35.1%)			
		Full-time sick leave or disability pension	21/57 (36.8%)			
	Work ability[f]	Good or excellent (WAI[f] >36)	13/48 (27.1%)	Not performed	–	–
		Less good (WAI[f] = 28–36)	15/48 (31.2%)			
		Poor (WAI[f]<28)	20/48 (41.6%)			
Bérezné *et al*. (2011)	Work disability[g]	Any reason	46/113 (40.7%)	Age	1.10 (1.05-1.2)	0.0006
		Retired early due to SSc	7/113 (6.2%)	Disease duration	1.09 (1.01-1.2)	0.03
		Receiving a full disability pension	36/113 (31.9%)	Global disability[c]	6.1 (2.2-16.7)	0.0005
		On sick leave due to SSc	27/113 (23.9%)	Hand disability[h]	0.97 (0.93-1)	0.13
Sharif *et al*. (2011)	Work disability	Any reason	124/284 (43.6%)	*Cross-sectional*		
		Retired early, unemployed, or part-time due to health problems	41/284 (14.4%)	Educational level > Associate's degree	0.22 (0.12, 0.43)	<0.001
		Work disabled	83/284 (29.2%)	Lung disease severity[i]	1.50 (1.14, 1.99)	0.004
				Fatigue[j]	2.18 (1.51, 3.14)	<0.001
				Social support[k]	0.66 (0.54, 0.82)	<0.001
				Longitudinal		
				Caucasian race	0.46 (0.22, 0.96)	0.038
				Fatigue[j]	1.96 (1.19, 3.25)	0.009
				Diffusing capacity of the lung for carbon monoxide	0.98 (0.97, 0.99)	0.038
Singh *et al*. (2012)	Work disability[l]	Unable to work due to SSc	39/159 (24.5%)	Educational level	–	0.01
				Global disability[c]	–	0.01
	Loss of productivity[m]	Missed ≥1 day of work/month	24/58 (41.4%)	Education level	–	0.014
				Global health, physician assessment	–	0.026
Decuman *et al*. (2012)	Work transition	Made a health-related work transition	47/84 (56.0)	Disease duration	1.010–1.013	0.009-0.016
		-Any change in work	13/84 (15.5)			
		Reduced work hours	11/84 (13.1)	Global disability[c]	7.82–10.11	0.001-0.007
		Changed job	1/84 (1.2)			
		Reduced work hours and changed job	1/84 (1.2)	Self-rated health[n]	0.94 (0.90-0.98)	0.002
		-Left labour force	34/84 (40.5)			

[a]Result of multivariate analysis; [b]Groups taken together for analysis; [c]Assessed by the Health Assessment Questionnaire-Disability Index; [d]Self-reported; [e]Assessed by the Karnofsky Performance Scale. [f]Work Ability Index (WAI, range 7-49: higher scores indicating better work ability). Score on the WAI is missing for 9 patients. [g]For patients in the work force (18–61 years of age). [h]Assessed by the Cochin Hand Function Scale. [i]Assessed by the Medsger Disease Severity Scale. [j]Assessed by Fatigue Severity Scale. [k]Assessed by Interpersonal Support Evaluation List. [l]Three patients did not mention their employment status so data are provided for n=159. [m]Missing for two patients. [n]Assessed by Medical Outcomes Study Short Form-36.

– Influence of functional factors on work variables

Global disability was assessed in 5 of the 8 studies. In 4 of these, patients with a higher Health Assessment Questionnaire Disability index (HAQ-DI) had a higher risk of being work disabled (OR: 6.41, CI: not mentioned, $p<0.001$; OR: 6.10, CI: 2.20-16.7, $p=0.0005$; OR and CI: not mentioned, $p<0.01$) or having made a work transition (OR: 7.82-10.11, $p=0.001$-0.007) (8, 9, 13, 19). In 5 of 8 studies, the influence of health/wellbeing was examined. In 3 of the studies, health/wellbeing was associated with the work variable: higher perceived health status was independently associated with not being on full-time sick



[a] Confidence intervals are given between brackets. [b] The weighted proportion of patients not working is obtained from a DerSimonian-Laird random-effects model (DERSIMONIAN R, LAIRD N: Meta-analysis in clinical trials. Control Clin Trials 1986; 7: 177-88).

Fig. 2. Forest plot of percentage patients not working in the included studies (n=8).

leave (OR: 0.92, CI: 0.88-0.98, *p*-value: not mentioned) (12), higher self-rated health with not having made a work transition (OR: 0.94, CI: 0.90-0.98, *p*=0.002) (9), and a lower physician's assessment of overall health with loss of productivity (OR and CI: not mentioned, *p*=0.026) [19]. Hand function was assessed in 2 studies, and in 1 of the studies, patients with better hand function had a lower risk of work disability (OR: 0.97, CI: 0.93-1, *p*=0.13) (8).

– Influence of social network factors on work variables

Patients who experience more social support are less likely to be work disabled (OR: 0.66, CI: 0.54-0.82, *p*<0.001) (18).

– Influence of job-related characteristics on work variables

Only 1 study assessed the influence of job-related factors: patients with a physically demanding job had higher risk of being work disabled (OR: 3.06, CI: not mentioned, *p*=0.013) (13).

Qualitative study with work as main focus

The qualitative study collected data from interviews of four focus groups, which included 17 patients currently working (see Supplementary file 3) (14). Work was influenced by symptoms such as fatigue, pain, cold intolerance, stiffness and impaired dexterity. Patients perceived their work role as important and managed their working life by reduced working hours, striving for work flexibility and simplifying/prioritising meaningful activities (as work and private life interact). Work adjustments depend on the employer, coworkers and the individual's attitude towards talking about limitations. Some patients do not disclose limitations due to SSc because they are afraid of the consequences. Nevertheless, patients who disclose limitations had good experiences with it.

Quantitative studies where work was not the main focus

– Prevalence of not working and reduced working hours

The prevalence of not working or of reduced working hours due to health status was described in 3 studies. One of these studies included 80 patients, of whom only 7 (8.75%) were in a full-time job, 1 (1.25%) was on permanent sick leave but 39 (48.8%) received a disability allowance (29). In the second study 23/94 (25%) were disabled (38). In the third study, which included only women, 5/36 (14%) had a full sick pension and 12/36 (33%) combine a pension with work (3).

– Costs due to SSc

The costs due to productivity loss (indirect costs as measured by the human capital method) constitute 56% of total costs of patients with SSc (29, 39). The disability-related productivity loss (55.2%) and hospitalisation (28.3%) were the highest among the cost items. There was a significant correlation between the indirect costs and the European Scleroderma Study Group Activity Index/peripheral vascular disease severity as measured by the disease severity scale of Medsger (Spearman's rank correlation coefficient 0.23 for both) (29).

– Other findings

One study reported that women with SSc worked less and were less satisfied with work. The latter was significantly correlated with general life satisfaction, domain-specific life satisfaction, and self-rated health (3). Work was amongst the 5 (8) or 10 (27) activities most often cited by patients as affected by disability. In a study about health care needs and preferences, of 64 patients, 46 (72%) had problems with maintaining job or study performance and 32 (70%) had not discussed this with a health care provider, although for the latter, only 4 (13%) considered this an unmet need (37).

Discussion

This systematic review, which included 8 quantitative studies focused on work, found that 18% to 61% patients stopped working. The pooled estimate of the percentage not working is 37%. Between 14% and 40% have reduced working hours (8, 9, 12, 13, 18, 19). The percentage of working patients becoming work disabled during follow-up was described in 1 study and found to be 27% (18). Independent factors for reduced work vary in strength and frequency of the relationships found. With global disability there was a strong and frequently found association. Less frequently found but with a strong relationship is having a lower income, the presence of myalgia's, the extend of skin in-

volvement and having a physically demanding job. A less stronger association was found with health/hand function and disease duration but the relationship was found in all but one/all studies respectively assessing this parameter in multivariate analysis. As for educational level not all odds are given interpretation of the strength of the relationship is difficult (8, 9, 12, 13, 18, 19).

Race, decreased DLCO, and fatigue were associated with becoming work disabled at follow-up (18).

These findings are partially in line with a former published systematic review (17). Employment rates varied between 11% and 82%, the lower value from a study that we considered out of scope as the focus was on costs due to SSc. Both the previous systematic review and the current analyses suggested that work disability is related to functioning or global disability and a poorer quality of life, health, or wellbeing but less evidence was found for a relationship with age and sex. The former review found inconsistent evidence for an association with education and disease duration. Contrarily, we found that disease duration was associated with the work variable in 3/3 studies. The relationship with educational level was also in our review not frequently found (2/4).

The only qualitative study included in our review, examined the importance of managing the work situation, disclosure of limitations due to SSc, and using one's own resources to adapt to the disease (14).

There are three studies (two quantitative/one qualitative) which have work as scope but with no clear classification criteria and were therefore excluded (10, 11, 40). The percentage of patients not working lied within the range we found (21% and 33%, respectively) (10, 11). One of the studies found that disease duration, having comorbidities and global disability were independently correlated with being work disabled (10). In the qualitative study patients with SSc, currently employed underwent a structured audiotaped phone interview. Work was perceived as a daily challenge on the level of the work environment, career planning and support from others (40).

Table IIB. Frequency of assessed parameters and results of univariate and multivariate analysis in 8 quantitative work-related studies.

Characteristic	Frequency univariate assessment n/N	Frequency significant relationships n/N	Frequency of multivariate assessment n/N	Frequency of significant relationships n/N
Patient characteristics				
Educational level	6/8	3/6	4/8	**2/4**
Age	8/8	3/8	2/8	**1/2**
Marital status	4/8	1/4	2/8	0/2
Having a decreased income	1/8	1/1	1/8	**1/1**
Race	1/8	1/1	1/8	**1/1**
Region of residence	1/8	1/1	1/8	0/1
Sex	7/8	0/7	0/8	NA
Disease-related characteristics				
Disease duration	8/8	3/8	3/8	**3/3**
Skin involvement	8/8	2/8	3/8	**1/3**
Lung involvement	6/8	4/6	3/8	**1/3**
Muscle involvement	4/8	1/4	2/8	**1/2**
Cardial involvement	4/8	1/4	2/8	0/2
Gastro-intestinal involvement	3/8	2/3	2/8	0/2
Joint involvement	4/8	1/4	2/8	0/2
Comorbidity	1/8	1/1	1/8	0/1
SSc general	3/8	2/3	1/8	0/1
Pitting scars/ulcers/Raynaud	6/8	2/6	1/8	0/1
Valentini disease activity index	1/8	1/1	1/8	0/1
General stiffness	2/8	1/2	0/8	NA
Psychological factors				
Depression	3/8	2/3	3/8	0/3
Fatigue	4/8	4/4	2/8	**1/2**
Pain	6/8	3/6	1/8	0/1
Learned helplessness	1/8	1/1	1/8	0/1
Coping with illness	1/8	1/1	1/8	0/1
Will to follow education	1/8	1/1	1/8	0/1
Feelings of discrimination	1/8	1/1	1/8	0/1
Lack of advancement	1/8	1/1	1/8	0/1
Satisfaction with life/occupations	2/8	2/2	0/8	NA
Empowerment	1/8	1/1	0/8	NA
Functional factors				
Global disability	6/8	6/6	5/8	**4/5**
'Health/Well being	7/8	6/7	5/8	**3/5**
Handfunction	4/8	4/4	2/8	**1/2**
Mouth handicap	1/8	1/1	1/8	0/1
Performing occupations	2/8	2/2	0/8	NA
Social network factors	*2/8*	*2/2*	*1/8*	**1/1**
Job-related factors	*4/8*	*3/4*	*1/8*	**1/1**

Values are the number of studies meeting the criteria (n) over the number of total studies (N). NA: not assessed in multivariate analysis.

One recently published study assessed how work impairment varies by number of digital ulcers (DU) (categorised in 0, 1-2 and ≥3 DUs at enrolment). The authors found that the number of work hours missed due to DUs, impairment due to DUs while working and mean overall work impairment, increased with number of DUs. We did not include the article in this review because a mixed patient group was included (LcSSc, DcSSc, overlap/mixed connective tissue disease, other specified, other not specified) and work-related data were not given separately for the subgroups (41).

Interpretation of the results of this review was complicated by variability in the characteristics of the sample (although we only included studies which clearly describe classification criteria), the work variable and parameters under study and differences in social security systems (42). Furthermore, the studies that have been performed are mostly cross-sectional and therefore the relationship between work disability and self-reported patients outcomes (*e.g.*

HAQ-DI) can be overestimated due to the bi-directional nature of the relationship. For example the higher HAQ-DI in patients on work disability may also be due to the fact that those patients do not work and not merely the cause/predictor of not working. More longitudinal studies are needed with clear descriptions of the work variable and of the measurements made, to make conclusions about causality. In addition, further qualitative research is needed to provide a more comprehensive view on this topic. Finally, we did not find any literature on the impact of interventions on the ability to work in patients with SSc. Studies on the ability of interventions to improve work variables are a priority given the costs of work disability (29).

References

1. BOIN F, WIGLEY F: Clinical Features and Treatment of Scleroderma. *In* FIRESTEIN G, BUDD R, GABRIEL S, MCINNES I, O'DELL J (Eds.): *Kelley's Textbook Of Rheumatology*. 9th ed. Philadelphia, Elsevier 2013: 1366-403.
2. DEL ROSSO A, BOLDRINI M, D'AGOSTINO D *et al*.: Health-related quality of life in systemic sclerosis as measured by the Short Form 36: relationship with clinical and biologic markers. *Arthritis Rheum* 2004; 51: 475-81.
3. SANDQVIST G, AKESSON A, EKLUND M: Daily occupations and well-being in women with limited cutaneous systemic sclerosis. *Am J Occup Ther* 2005; 59: 390-7.
4. SANDQVIST G, EKLUND M: Daily occupations - performance, satisfaction and time use, and relations with well-being in women with limited systemic sclerosis. *Disabil Rehabil* 2008; 30: 27-35.
5. SANDQVIST G, EKLUND M, AKESSON A, NORDENSKIOLD U: Daily activities and hand function in women with scleroderma. *Scand J Rheumatol* 2004; 33: 102-7.
6. SANDQVIST G, HESSELSTRAND R, EBERHARDT K: A longitudinal follow-up of hand involvement and activities of daily living in early systemic sclerosis. *Scand J Rheumatol* 2009; 38: 304-10.
7. GOLEMATI C, MOUTSOPOULOS H, VLACHOYIANNOPOULOS P: Psychological characteristics of systemic sclerosis patients and their correlation with major organ involvement and disease activity. *Clin Exp Rheumatol* 2013; 31 (Suppl. 76): S37-S45.
8. BÉREZNÉ A, SEROR R, MORELL-DUBOIS S *et al*.: Impact of systemic sclerosis on occupational and professional activity with attention to patients with digital ulcers. *Arthritis Care Res* 2011; 63: 277-85.
9. DECUMAN S, SMITH V, VERHAEGHE S, DESCHEPPER E, VERMEIREN F, DE KEYSER F: Work participation and work transition in patients with systemic sclerosis: a cross-sectional study. *Rheumatology* (Oxford) 2012; 51: 297-304.
10. HUDSON M, STEELE R, LU Y, THOMBS BD, THE CANADIAN SCLERODERMA RESEARCH GROUP, BARON M: Work disability in Systemic Sclerosis. *J Rheumatol* 2009; 36: 1-6.
11. MAU W, LISTING J, HUSCHER D, ZEIDLER H, ZINK A: Employment across chronic inflammatory rheumatic diseases and comparison with the general population. *J Rheumatol* 2005; 32: 721-8.
12. NGUYEN C, POIRAUDEAU S, MESTRE-STANISLAS C *et al*.: Employment status and socioeconomic burden in systemic sclerosis: a cross-sectional survey. *Rheumatology* 2010; 49: 1-8.
13. OUIMET JM, POPE JE, GUTMANIS I, KOVAL J: Work disability in scleroderma is greater than in rheumatoid arthritis and is predicted by high HAQ Scores. *Open Rheumatol J* 2008; 2: 44-52.
14. SANDQVIST G, HESSELSTRAND R, SCHEJA A, HAKANSSON C: Managing work life with systemic sclerosis. *Rheumatology* (Oxford) 2012; 51: 319-23.
15. SANDQVIST G, SCHEJA A, EKLUND M: Working ability in relation to disease severity, everyday occupations and well-being in women with limited systemic sclerosis. *Rheumatology* 2008; 47: 1708-11.
16. SANDQVIST G, SCHEJA A, HESSELSTRAND R: Pain, fatigue and hand function closely correlated to work ability and employment status in systemic sclerosis. *Rheumatology* 2010; 49: 1739-46.
17. SCHOUFFOER AA, SCHOONES JW, TERWEE CB, VLIET VLIELAND TP: Work status and its determinants among patients with systemic sclerosis: a systematic review. *Rheumatology* (Oxford) 2012; 51: 1304-14.
18. SHARIF R, MAYES MD, NICASSIO PM *et al*.: Determinants of work disability in patients with systemic sclerosis: a longitudinal study of the GENISOS cohort. *Semin Arthritis Rheum* 2011; 41: 38-47.
19. SINGH MK, CLEMENTS PJ, FURST DE, MARANIAN P, KHANNA D: Work productivity in scleroderma: analysis from the University of California, Los Angeles scleroderma quality of life study. *Arthritis Care Res* (Hoboken) 2012; 64: 176-83.
20. BASSEL M, HUDSON M, BARON M *et al*.: Physical and occupational therapy referral and use among systemic sclerosis patients with impaired hand function: results from a Canadian national survey. *Clin Exp Rheumatol* 2012; 30: 574-7.
21. WADDELL G, BURTON AK: *Is work good for your health and well-being*? 2nd ed., London, The Stationery Office, 2007: 246.
22. GRONNING K, RODEVAND E, STEINSBEKK A: Paid work is associated with improved health-related quality of life in patients with rheumatoid arthritis. *Clin Rheumatol* 2010; 29: 1317-22.
23. ROBINSON SM, WALKER DJ: Negotiating targets with patients: choice of target in relation to occupational state. *Rheumatology* (Oxford) 2012; 51: 293-6.
24. ROBINSON D, JR., AGUILAR D, SCHOENWETTER M *et al*.: Impact of systemic lupus erythematosus on health, family, and work: the patient perspective. *Arthritis Care Res* (Hoboken) 2010; 62: 266-73.
25. LASTOWIECKA E, BUGAJSKA J, NAJMIEC A, RELL-BAKALARSKA M, BOWNIK I, JEDRYKA-GORAL A: Occupational work and quality of life in osteoarthritis patients. *Rheumatol Int* 2006; 27: 131-9.
26. CHORUS AM, BOONEN A, MIEDEMA HS, VAN DER LINDEN S: Employment perspectives of patients with ankylosing spondylitis. *Ann Rheum Dis* 2002; 61: 693-9.
27. MOUTHON L, RANNOU F, BEREZNE A *et al*.: Patient preference disability questionnaire in systemic sclerosis: a cross-sectional survey. *Arthritis Rheum* 2008; 59: 968-73.
28. BERNATSKY S, HUDSON M, PANOPALIS P *et al*.: The cost of systemic sclerosis. *Arthritis Rheum* 2009; 61: 119-23.
29. MINIER T, PENTEK M, BRODSZKY V *et al*.: Cost-of-illness of patients with systemic sclerosis in a tertiary care centre. *Rheumatology* (Oxford) 2010; 49: 1920-8.
30. AMERICAN RHEUMATISM ASSOCIATION: Preliminary criteria for the classification of systemic sclerosis (scleroderma). Subcommittee for scleroderma criteria of the American Rheumatism Association Diagnostic and Therapeutic Criteria Committee. *Arthritis Rheum* 1980; 23: 581-90.
31. HAAFKENS J, MOERMAN C, SCHURING M, VAN DIJK F: Searching bibliographic databases for literature on chronic disease and work participation. *Occup Med* (Lond) 2006; 56: 39-45.
32. DERSIMONIAN R, LAIRD N: Meta-analysis in clinical trials. *Control Clin Trials* 1986; 7: 177-88.
33. DE CROON EM, SLUITER JK, DIJKMANS BAC, LANKHORST GJ, FRINGS-DRESEN MHW: Predictive factors of work disability in rheumatoid arthritis: a systematic literature review. *Ann Rheum Dis* 2004; 63: 1362-7.
34. PINCUS T, VOGEL S, BURTON AK, SANTOS R, FIELD AP: Fear avoidance and prognosis in back pain - a systematic review and synthesis of current evidence. *Arthritis Rheum* 2006; 54: 3999-4010.
35. THOMBS BD, TAILLEFER SS, HUDSON M, BARON M: Depression in patients with systemic sclerosis: a systematic review of the evidence. *Arthritis Rheum* 2007; 57: 1089-97.
36. MACEACHEN E, CLARKE J, FRANCHE R, IRVIN E, WORKPLACE BASED RETURN TO WORK LITERATURE REVIEW GROUP: Systematic Review of the Qualitative literature on return to work after injury. *Scan J Work Environ Health* 2012; 32: 257-69.
37. SCHOUFFOER AA, ZIRKZEE EJM, HENQUET SM *et al*.: Needs and preferences regarding health care delivery as perceived by patients with systemic sclerosis. *Clin Rheumatol* 2011; 30: 815-24.
38. MOSER DK, CLEMENTS PJ, BRECHT ML, WEINER SR: Predictors of psychosocial adjustment in systemic sclerosis - the influence of formal education level, functional ability, hardiness, uncertainty, and social support. *Arthritis Rheum* 1993; 36: 1398-405.
39. TRANMER J, GUERRIERE D, UNGAR W, COYTE P: Valuing patient and caregiver time: a review of the literature. *Pharmacoeconomics* 2005; 23: 449-59.
40. MENDELSON C, POOLE JL, ALLAIRE S: Experiencing work as a daily challenge: the case of scleroderma. *Work* 2013; 44: 405-13.
41. GUILLEVIN L, HUNSCHE E, DENTON C *et al*.: Functional impairment of systemic scleroderma patients with digital ulcerations: results from the DUO Registry. *Clin Exp Rheumatol* 2013; 31: S71-S80.
42. DE BOER WEL, BESSELING JJM, WILLEMS JHBM: Organisation of disability evaluation in 15 countries. *Pratiques et organisation des soins* 2007; 38: 205-17.