Validity of the Workers Productivity and Activity Impairment Questionnaire: Specific Health Problem (WPAI:SHP) in patients with systemic sclerosis

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ABSTRACT

Objective. *To evaluate the construct validity of the Workers Productivity and Impairment Activity Index: Specific Health Problem (WPAI:SHP) in Australian systemic sclerosis (SSc) patients.*

Methods. *SSc patients, identified through the Australian Scleroderma Cohort Study database, completed the WPAI:SHP and a quality of life instrument (PROMIS-29) cross-sectionally. The construct validity of the WPAI:SHP was assessed by the correlations between the WPAI:SHP and a range of SSc health states. Non-parametric correlation, including Spearman's correlation (ϱ), was used to test the validity of WPAI:SHP and ability to distinguish between different health states.*

Results. *A total of 476 completed questionnaires was returned, equating to a response rate of 63.7%. Among those under 65 years of age, 155 patients (55.2%) were in paid employment. Employed patients had a mean (± SD) age of 56.5 (9.8) years and were predominantly female (87.3%) with limited disease subtype (75.6%). The WPAI:SHP showed construct validity based on moderate to strong correlations with health status as assessed by a range of health outcome measures including disease activity (ϱ=0.34-0.39, p=0.001), physical function (ϱ=0.55-0.62, p=0.001), disease severity(ϱ=0.55-0.62, p=0.001), fatigue (ϱ= 0.62-0.63, p=0.001), pain (ϱ=0.68-0.71, p=0.001), and breathlessness (ϱ=0.39-0.46, p=0.001). Furthermore, according to the effect size, the WPAI:SHP scores have a large discriminative ability (d=1.26-1.47) for distinguishing SSc patients with different health outcomes.*

Conclusion. *The WPAI is a valid questionnaire for assessing impairments in paid employment and social activities in SSc patients, and for measuring the relative differences between SSc patients with varying health states.*

Introduction

Systemic sclerosis (SSc) is a chronic autoimmune multi-organ disease that occurs during peak working age (1). SSc is characterised by fibrosis of the skin and internal organs leading to joint contractures, ischaemic digital ulcers, hand dysfunction and altered physical appearance (1, 2). As the disease progresses, these manifestations cause functional limitations and psychological burden which can impact negatively on employment and patient reported health related quality of life (HRQoL) (3). Therefore, SSc has the potential to have a significant impact on work productivity with an associated economic burden.

There is a paucity of literature assessing work productivity in SSc. In 2014, the Outcome Measures in Rheumatoid Arthritis Clinical Trials (OMERACT) Worker Productivity Working Group reviewed all available work ability questionnaires and recommended only two for use in rheumatic diseases, the Rheumatoid Arthritis Specific–Work Productivity Survey (WPS-RA) and the Work Productivity and Activity Impairment questionnaire (WPAI) (4). Neither of these instruments has been validated for use in the SSc population.

To our knowledge, there is only one study assessing work productivity in SSc using a validated work productivity instrument, the WPS-RA (5). This found that only 37% of SSc patients were employed outside of the home and those patients reported missing 2.6 days of work per month of work, and had productivity reduced by half on 2.5 days per month. Additionally, they found that of the unemployed patients, 39.4% were unable

to work due to their SSc, highlighting the impact SSc can have on employment and work productivity. We recently assessed work productivity in a large cohort of Australian SSc patients using the WPAI and found a high frequency of unemployment (44.8%) and substantial work and activity impairment amounting to a high annual indirect economic cost per patient (6).

The WPAI is available in the public domain and can be used without fee or permission from the authors (7). It has been validated for use in a broad range of non-rheumatic conditions such as asthma (8) and gastro-oesphageal reflux disease (9) and also rheumatic conditions such as rheumatoid arthritis (RA) and ankylosing spondylitis (AS) (10, 11). Additionally, the WPAI has been used to compare work impairment between individuals with different disease severity levels (10).

A validated questionnaire is one that has been shown to accurately measure what it is intended to, regardless of who responds, when they respond and to whom they respond (12). Construct validity or the extent to which a measuring instrument accurately measures a theoretical construct it is designed to measure, is the most important type of validity. It can be determined either by correlating the performance on the questionnaire with performance on a test for which construct validity has been established or by demonstrating that the scores of the new test differ among people with different disease states or outcomes (12).

The primary objective of this study was to evaluate the construct validity of the WPAI in Australian patients with SSc and to determine its ability to differentiate between various health states in SSc as determined by patient-reported outcomes relating to HRQoL and physician assessment of disease status.

Appendix 1. Work Productivity and Activity Impairment Questionnaire: Specific Health Problem V2.0 (WPAI:SHP)

The following questions ask about the effect of your Scleroderma on your ability to work and perform regular activities. Please fill in the blanks or circle a number, as indicated.

1. Are you currently employed (working for pay)? _____ NO ___ YES
If NO, check "NO" and skip to question 6.

The next questions are about the past seven days, not including today.

2. During the past seven days, how many hours did you miss from work because of problems associated with your Scleroderma? Include hours you missed on sick days, times you went in late, left early, etc., because of your Scleroderma. Do not include time you missed to participate in this study. _____ HOURS

3. During the past seven days, how many hours did you miss from work because of any other reason, such as vacation, holidays, time off to participate in this study? __HOURS

4. During the past seven days, how many hours did you actually work? ____HOURS (If "0", skip to question 6.)

5. During the past seven days, how much did your Scleroderma affect your productivity while you were working? Think about days you were limited in the amount or kind of work you could do, days you accomplished less than you would like, or days you could not do your work as carefully as usual. If Scleroderma affected your work only a little, choose a low number. Choose a high number if Scleroderma affected your work a great deal.Consider only how much Scleroderma affected productivity while you were working.

	CIRCLE A NUMBER										
PROBLEM had no effect on my work	0	1	2	3	4	5	6	7	8	9	10 PROBLEM completely prevented me from working

6. During the past seven days, how much did your Scleroderma affect your ability to do your regular daily activities, other than work at a job? By regular activities, we mean the usual activities you do, such as work around the house, shopping, childcare, exercising, studying, etc. Think about times you were limited in the amount or kind of activities you could do and times you accomplished less than you would like. If Scleroderma affected your activities only a little, choose a low number. Choose a high number if Scleroderma affected your activities a great deal. Consider only how much PROBLEM affected your ability to do your regular daily activities, other than work at a job.

	CIRCLE A NUMBER										
PROBLEM had no effect on my daily activities	0	1	2	3	4	5	6	7	8	9	10 PROBLEM completely prevented me from doing my daily activities

Methods

Study design and patient cohort

Data for this study were collected by means of a cross-sectional survey of SSc patients enrolled in the Australian Scleroderma Cohort Study (ASCS). The ASCS is an Australian multi-centre study of risk and prognostic factors for cardiopulmonary and other clinically important outcomes in SSc. Written consent is obtained from all patients at recruitment to ASCS and for the purposes of this study, consent was assumed if patients returned the completed survey questionnaires as approved by the human research ethics committees of each of the participating hospitals.

Inclusion and exclusion criteria

We included all consecutive English speaking adult (>18 years) SSc patients from four Australian States (Victoria, Tasmania, South Australia and Western Australia), recruited between December 2007 and January 2016. These states were chosen as they comprise the majority of patients in ASCS and have the most complete and up-to-date clinical data entered in the ASCS database. All patients fulfilled either the American College of Rheumatology or LeRoy and Medsger criteria for SSc (13, 14). After consultation with the treating physician, 800 patients were mailed the Workers Productivity and Activity Impairment Questionnaire: Specific Health Problem (WPAI:SHP) and a health related quality of life (HRQoL) questionnaire, the Patient-Reported Outcomes Measurement Information System 29 (PROMIS-29). A cover letter explaining the purpose of the study was included in the material mailed out to patients. Patients were asked to return the questionnaires within a six-week time frame, with a specified due date using a stamped addressed envelope supplied for this purpose.

WPAI:SHP

The WPAI:SHP (Appendix 1) measures the effect of health and symptom severity on work productivity and non-work activities (15). It consists of six questions addressing employment status, number of hours missed from work due to a health problem, hours missed from work for other reasons, hours actually worked, degree to which a health problem affected productivity while working, and the degree to which a health problem affected productivity in other non-paid activities outside work. From the responses to these questions, scores are generated for absenteeism (work time missed), presenteeism (impairment at work and or reduced on-the-job effectiveness), work productivity loss (overall work impairment which incorporates both absenteeism and presenteeism) and activity impairment (16). These four scores are each expressed as a percentage of impairment due to the health problem (0-100%) where a higher percentage indicates a greater reduction in productivity.

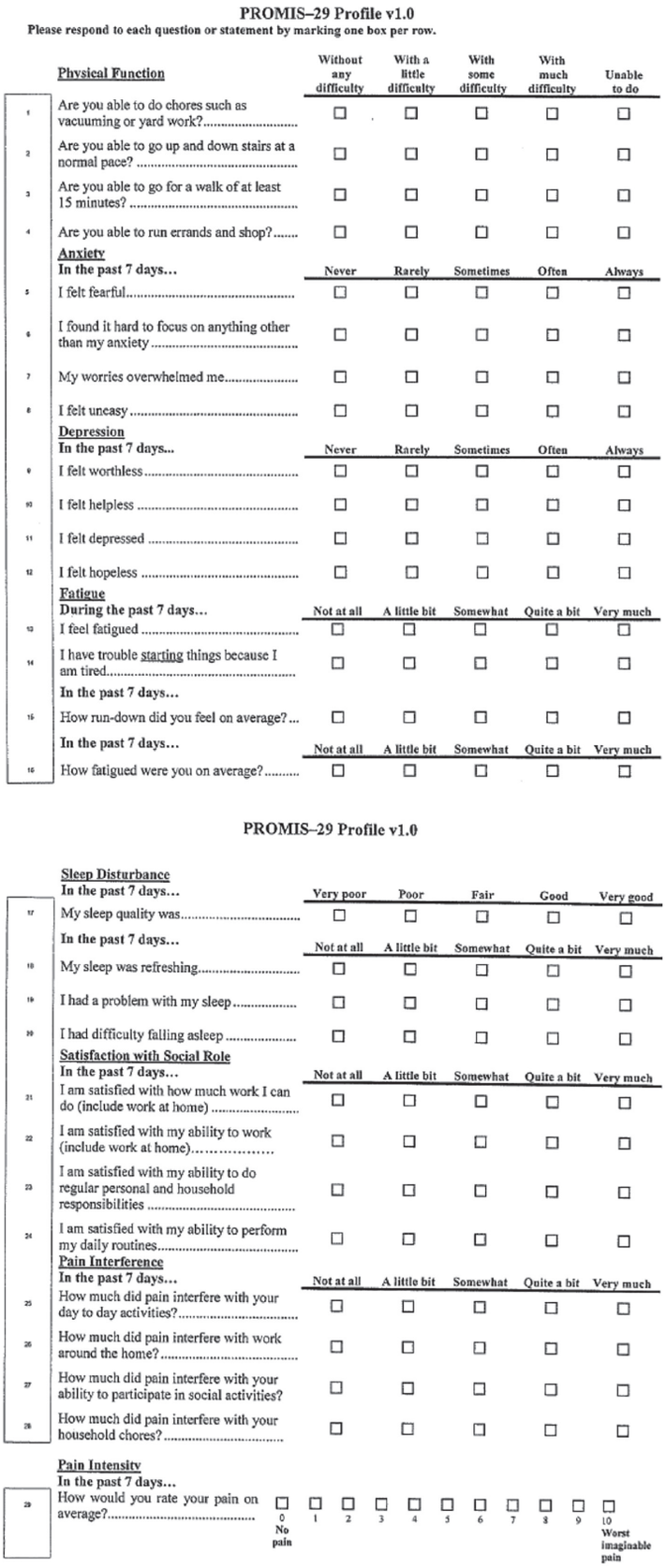
PROMIS-29

The PROMIS-29 (Appendix 2) is a validated instrument for measuring HRQoL in SSc (17). It consists of twenty-nine questions addressing physical function, anxiety, depression, fatigue, sleep disturbance, satisfaction with social role, pain interference and pain intensity. Each health domain has an individual score that is standardised against US normative population data. A score above 50 represents better than the average population, while a score below 50 is worse. Two exceptions are for anxiety and depression, where a score above 50 is worse than the general population and below 50 is better.

Data collection

Questionnaire results were entered into a spreadsheet and merged with data from the ASCS database that includes patient demographics, clinical variables including cardiac and pulmonary assessments and measures of health status (see below). Clinical manifestations and autoantibody status were defined as present, if present ever from SSc diagnosis. The physicians' classification of patients into limited (lcSSc) and diffuse cutaneous (dcSSc) subtypes was confirmed by reviewing their peak recorded modified Rodnan skin scores (mRSS). Pulmonary involvement was assessed by pulmonary

Appendix 2.
PROMIS-29



PROMIS–29 Profile v1.0

Please respond to each question or statement by marking one box per row.

	Physical Function	Without any difficulty	With a little difficulty	With some difficulty	With much difficulty	Unable to do
1	Are you able to do chores such as vacuuming or yard work?	☐	☐	☐	☐	☐
2	Are you able to go up and down stairs at a normal pace?	☐	☐	☐	☐	☐
3	Are you able to go for a walk of at least 15 minutes?	☐	☐	☐	☐	☐
4	Are you able to run errands and shop?	☐	☐	☐	☐	☐
	Anxiety					
	In the past 7 days…	**Never**	**Rarely**	**Sometimes**	**Often**	**Always**
5	I felt fearful	☐	☐	☐	☐	☐
6	I found it hard to focus on anything other than my anxiety	☐	☐	☐	☐	☐
7	My worries overwhelmed me	☐	☐	☐	☐	☐
8	I felt uneasy	☐	☐	☐	☐	☐
	Depression					
	In the past 7 days...	**Never**	**Rarely**	**Sometimes**	**Often**	**Always**
9	I felt worthless	☐	☐	☐	☐	☐
10	I felt helpless	☐	☐	☐	☐	☐
11	I felt depressed	☐	☐	☐	☐	☐
12	I felt hopeless	☐	☐	☐	☐	☐
	Fatigue					
	During the past 7 days…	**Not at all**	**A little bit**	**Somewhat**	**Quite a bit**	**Very much**
13	I feel fatigued	☐	☐	☐	☐	☐
14	I have trouble starting things because I am tired	☐	☐	☐	☐	☐
	In the past 7 days…					
15	How run-down did you feel on average?	☐	☐	☐	☐	☐
	In the past 7 days…	**Not at all**	**A little bit**	**Somewhat**	**Quite a bit**	**Very much**
16	How fatigued were you on average?	☐	☐	☐	☐	☐

PROMIS–29 Profile v1.0

	Sleep Disturbance	Very poor	Poor	Fair	Good	Very good
	In the past 7 days…					
17	My sleep quality was	☐	☐	☐	☐	☐
	In the past 7 days…	**Not at all**	**A little bit**	**Somewhat**	**Quite a bit**	**Very much**
18	My sleep was refreshing	☐	☐	☐	☐	☐
19	I had a problem with my sleep	☐	☐	☐	☐	☐
20	I had difficulty falling asleep	☐	☐	☐	☐	☐
	Satisfaction with Social Role					
	In the past 7 days…	**Not at all**	**A little bit**	**Somewhat**	**Quite a bit**	**Very much**
21	I am satisfied with how much work I can do (include work at home)	☐	☐	☐	☐	☐
22	I am satisfied with my ability to work (include work at home)	☐	☐	☐	☐	☐
23	I am satisfied with my ability to do regular personal and household responsibilities	☐	☐	☐	☐	☐
24	I am satisfied with my ability to perform my daily routines	☐	☐	☐	☐	☐
	Pain Interference					
	In the past 7 days…	**Not at all**	**A little bit**	**Somewhat**	**Quite a bit**	**Very much**
25	How much did pain interfere with your day to day activities?	☐	☐	☐	☐	☐
26	How much did pain interfere with work around the home?	☐	☐	☐	☐	☐
27	How much did pain interfere with your ability to participate in social activities?	☐	☐	☐	☐	☐
28	How much did pain interfere with your household chores?	☐	☐	☐	☐	☐
	Pain Intensity					
	In the past 7 days…					
29	How would you rate your pain on average?	☐ 0 No pain	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5	☐ 6	☐ 7	☐ 8	☐ 9	☐ 10 Worst imaginable pain

function tests (PFTs) and /or high resolution computed tomography (HRCT) of the chest. Interstitial lung disease was defined on HRCT appearances and consistent changes on the PFTs (reduced forced vital capacity and diffusion capacity for carbon monoxide (DLCO)). Cardiac involvement was assessed using transthoracic echocardiogram and pulmonary hypertension was diagnosed on right heart catheterisation (RHC) according to international criteria (18).

Construct validity: relationship to health status

In addition to the fatigue and pain health domains of the PROMIS-29, a number of indices of health status recorded as part of the ASCS protocol within twelve months of the mailed questionnaires were selected to assess construct validity of the WPAI:SHP. The health status indices are recorded on an annual basis at the ASCS visit. They were not collected as part of the questionnaire. These included the EUSTAR activity index, Health Assessment Questionnaire Disability Index (HAQ-DI), and physician-determined WHO Functional Class and Physician-Rated Global Disease Severity Assessment.

The EUSTAR activity index is a validated SSc disease activity instrument. It is used as a composite measure of global disease activity and predicts future damage (19). It consists of ten items including clinical manifestations such as mRSS, skin involvement, digital necrosis, vascular involvement and arthritis, serological markers such as titre of erythrocyte sedimentation rate (ESR) and hypocomplementaemia and PFT parameters including the DLCO. The index score ranges from 0-10, with 10 being maximum disease activity.

The HAQ-DI is a self-reported questionnaire assessing physical function validated for use in rheumatic diseases (20). The HAQ-DI score is measured from 0-3, with 0 being no functional limitation and 3 being severe functional limitation. WHO Functional Class is a measure of functional status limited by breathlessness, wherein Functional Class I is when patients can carry out their ordinary physical activity without undue breathlessness or fatigue and Class IV is when patients are unable to carry out any physical activity without symptoms of breathlessness or fatigue, and the Physician-Rated Global Disease Severity Assessment is a physician-rated estimate of organ damage caused by SSc rated on a scale from 0-10, with 0 being no damage and 10 being very severe damage (21).

Outcome variable and disease states

The outcome variable of interest was the WPAI:SHP which was treated as a continuous variable.

Analysis

We measured the extent to which the WPAI:SHP productivity scores correlated with health states as determined by the aforementioned indices recorded within twelve months of the cross-sectional survey. Construct validity was assessed using Spearman's correlation coefficient. We considered Spearman's correlation coefficient of 0-0.2 to indicate no or weak correlation, 0.21-0.40 to indicate moderate correlation and 0.4-1.0 to indicate strong correlation between the WPAI productivity outcome and health outcomes.

The ability of the WPAI:SHP to discriminate between health states was tested by dividing patients into two groups based on the median score for each health outcome including EUSTAR activity index, physical function, fatigue, pain and physician-rated disease severity assessment where better status was defined as less than or equal to the median outcome score and worse status was defined as more than the median outcome score. WHO Functional Class was dichotomised into better Functional Class (WHO Functional Class I and II) and worse Functional Class (WHO Functional Class III and IV). For between group comparison, the effect size and the standardised mean difference in outcome between the groups was calculated for all WPAI:SHP outcomes. An effect size of one indicates a change in magnitude equivalent to one standard deviation. According to Cohen (22), the absolute value of effect sizes can be categorised as small (d=0.2–0.5), medium (d=0.5-0.8) and large (d>0.8). A larger effect size indicates better discriminative ability. Student t-tests were used to identify whether WPAI outcomes differed between the two levels of health status.

Results

A total of 476 completed questionnaires was returned, equating to a response rate of 63.7%. There were no statistically significant differences in patient characteristics or disease manifestations between responders and non-responders (data not shown). Among those under sixty-five years of age, which is the standard retirement age in Australia, 155 patients (55.2%) were in paid employment. Employed patients had a mean (±SD) age of 56.5 (9.8) years and were predominantly female (87.3%) with limited disease subtype (75.6%) and long disease duration at the time the survey was completed (9.1±8.2 years). Employed patients had relatively mild disease activity as evidenced by mean (± SD) EUSTAR activity index of 2.9 (±1.9), mild physical dysfunction evidenced by mean (± SD) HAQ-DI score of 0.6 (±0.6), mild fatigue and pain measured by the PROMIS-29 (54.5±9.9 and 2.8±2.5, respectively), mild disease severity as evidenced by the Physician-Rated Disease Severity scale (3.6±1.8) and mild breathlessness with 83% of patients in either WHO Functional Class I or II. Patient characteristics and health outcome indices are summarised in Table I.

Among employed patients, 16.1% of patients reported missing work (absenteeism) and 63.3% reported impaired productivity while at work (presenteeism) in the last week due to their health. In addition, 70.3% of patients reported that their regular daily activities were impaired due to their health (Table II).

For the correlation analysis between the WPAI:SHP productivity outcomes and health status, all correlations were in the logical direction and were highly significant (Table III). There were moderate correlations between percent work-time missed due to health and disease activity, physical function, fatigue, pain, breathlessness and disease severity (0.24-0.38) and between per-

cent work-time impaired due to health and disease activity and breathlessness (0.34 and 0.39, respectively). Otherwise, there were strong correlations between the other three WPAI:SHP outcomes and all of the health states (Table III).

When patients were divided into two groups based on the median of each health outcome variable, each of the WPAI:SHP productivity outcomes was significantly lower among patients with better health status than patients with worse health status (Table IV). Patients with a EUSTAR disease activity score greater than the median for the cohort, compared with those with score less than the median, had higher absenteeism (23.0% *vs*.13.1%), presenteeism (32.2% *vs*. 16.4%), overall work impairment (36.3% *vs*. 16.7%) and higher social activity impairment (38.4% *vs*. 18.2%). Similarly, patients with a physical function score lower than the median score, compared with those above the median, had higher absenteeism (5.2% *vs*. 2.8%), presenteeism (31.6% *vs*. 12.8%), overall work impairment (34.6% *vs*. 13.7%) and higher social activity impairment (38.3% *vs*. 13.2%). This pattern was repeated across all the health outcome measures including fatigue, pain, breathlessness and disease severity.

According to the effect size, the four WPAI:SHP scores have a large discriminative ability (d=1.26-1.47) except for work-time missed (absenteeism) due to health, which had a small discriminative ability (d=0.37–0.42). Patients with a EUSTAR disease activity score greater than the median for the cohort had higher work-time impairment, overall work impairment and higher social activity impairment than those with an activity score below the median for the cohort.

Table I. Patient characteristics.

Variables (n=155)	% or mean (±SD)	Median (Q1-Q3)
Age at survey completion, years	56.5 ± 9.8	57.0 (50.2 - 62.8)
Female	138 (87.3%)	
Limited disease subtype	118 (75.6%)	
Disease duration, years*	9.1 ± 8.2	6.3 (2.7 - 13.4)
BMI	27.8 ± 5.7	26.8 (23.5 – 19.6)
Disease manifestations**		
Digital ulcers	47.8%	
Calcinosis	42.4%	
Synovitis	21.1%	
Joint contractures	37.3%	
Tendon friction rubs	8.9%	
ILD	23.4%	
PAH	0.6%	
Modified Rodnan Skin Score (mRSS)	11.0 ± 8.6	8.5 (4-17)
EUSTAR activity index (0-10)	2.9 ± 1.9	2.5 (1.5 – 4.5)
WHO Functional Class		
Class I	41.5%	
Class II	41.5%	
Class III	16.5%	
Class IV	0.7%	
Physical function (sHAQ_DI) (0-3)#	0.6 ± 0.6	0.4 (0-1)
Disease severity (0-10)##	3.6 ± 1.8	2 (2-5)
Fatigue (0-100)###	54.5 ± 9.9	55.1 (48.5 – 62.7)
Pain intensity global (NRS 0-10) ###	2.8 ± 2.5	2 (1-5)

*Disease duration from first non-Raynaud's clinical manifestation. **clinical manifestations defined as present if ever present from SSc diagnosis. #Health assessment questionnaire- Disability index (HAQ-DI) is a well-validated questionnaire measuring physical function in rheumatic conditions. ##disease severity as determined by the physician-rated global disease severity assessment scale on annual review. ###patient reported outcome on the PROMIS-29 form, which is a health related quality of life instrument. Pain measured on a numerical rating scale from 0-10, with 10 being the worst imaginable pain.

Table II. WPAI:SHP Outcomes in employed patients with systemic sclerosis.

WPAI Measurements	Patient number (n)	Mean (±SD)	Median (IQR)
Absenteeism			
overall % of work time missed due to SSc.	149	5.3 (17.3)	0 (0-0)
% of work time missed in those who missed work due to SSc.	24	32.9 (31.4)	25 (13.3-32.1)
Presenteeism			
overall % of work time impaired due to SSc.	150	22.2 (23.2)	20 (0-40)
% of work time impaired in those who experience work impairment	95	35.1 (19.9)	30 (20-50)
Overall work impairment			
% overall work impairment due to SSc.	143	24.4 (25.6)	20 (0-40)
% overall work impairment in those who experience absenteeism or presenteeism.	91	38.4 (22.2)	40 (20-50)
Overall activity limitation			
overall % activity limitation due to SSc.	155	26.5 (25.2)	20 (0-40)
% activity limitation due to SSc in those who experience limitation due to SSc.	109	37.7 (21.9)	30 (20-50)

Discussion

This is the first study to examine and support the construct validity of the WPAI:SHP in a large cohort of SSc patients. In our study, the WPAI:SHP showed construct validity as measured by moderate to strong correlations with health status as assessed by a range of health outcome measures including disease activity, disease severity, physical function, pain, fatigue and breathlessness. Additionally, the WPAI:SHP was able to discriminate between productivity levels across health states with worse productivity scores being associated with worse health status. Therefore, the WPAI:SHP productivity outcomes assess constructs that are relevant to the health status of patients living with SSc and to their physicians. Furthermore our study highlights the

Table III. Spearman correlations between WPAI:SHP scores and health status.

	EUSTAR activity index (n=125)	Function (n=116)	Fatigue (n=150)	Pain (n=150)	WHO functional class (n=145)	Disease severity (n=133)
% work time missed	0.24 (0.006)	0.27 (0.002)	0.38 (0.001)	0.34 (0.001)	0.29 (0.01)	0.29 (0.001)
% impairment while at work	0.34 (0.001)	0.55 (0.001)	0.63 (0.001)	0.68 (0.001)	0.39 (0.001)	0.55 (0.001)
% overall work impairment	0.39 (0.001)	0.59 (0.001)	0.62 (0.001)	0.68 (0.001)	0.38 (0.001)	0.56 (0.001)
% activity impairment	0.38 (0.001)	0.62 (0.001)	0.63 (0.001)	0.71 (0.001)	0.46 (0.001)	0.62 (0.001)

Disease activity score assessed by EUSTAR activity score; function assessed by HAQ-DI; fatigue and pain measured by the PROMIS-29; breathlessness measured by WHO functional class; physician-rated global disease severity assessment (physician determined score based on their assessment of extent of damage as a consequence of SSc). *p-value is denoted in bracket below the correlation coefficient.

Table IV. WPAI:SHP Outcomes between two patient groups defined by the median for each health status outcome.

	Patient group	EUSTAR activity index (n=132)	Function (n=121)	Fatigue (n=158)	Pain (n=158)	WHO functional class (n=152)	Disease severity (n=137)
% work time missed	Better	2.7 (13.1)	2.8 (14.5)	1.2 (6.1)	2.1 (11.9)	2.3 (7.6)	1.6 (8.1)
	Worse	9.6 (23.0)	5.2 (15.8)	10.8 (24.5)	8.9 (21.4)	23.0 (37.5)	8.3 (21.9)
	Effect size	0.38	0.38	0.40	0.40	0.42	0.37
	p-value	0.006	0.02	<0.001	<0.001	<0.001	0.01
% impairment while at work	Better	16.4 (20.3)	12.8 (18.2)	10.9 (14.4)	10.3 (15.6)	19.3 (21.7)	6.5 (11.2)
	Worse	32.2 (24.8)	31.6 (23.5)	37.8 (24.1)	37.0 (22.6)	40.5 (24.8)	31.7 (24.1)
	Effect size	1.28	1.26	1.34	1.34	1.35	1.29
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
% overall work impairment	Better	16.7 (21.6)	13.7 (21.2)	12.2 (16.2)	10.9 (16.9)	21.1 (23.5)	7.7 (14.6)
	Worse	36.3 (27.3)	34.6 (24.4)	41.4 (26.4)	40.2 (24.9)	46.8 (28.5)	35.3 (26.3)
	Effect size	1.29	1.27	1.36	1.36	1.36	1.31
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001
% activity impairment	Better	18.2 (19.9)	13.2 (17.1)	14.3 (17.1)	12.6 (17.5)	22.9 (23.4)	10.6 (13.6)
	Worse	38.4 (26.7)	38.3 (24.6)	43.0 (25.2)	43.4 (22.8)	47.8 (25.2)	35.9 (26.5)
	Effect size	1.4	1.38	1.47	1.47	1.47	1.41
	p-value	<0.001	<0.001	<0.001	<0.001	<0.001	<0.001

Disease activity score assessed by EUSTAR activity score; function assessed by HAQ-DI; fatigue and pain measured by the PROMIS-29; breathlessness measured by WHO functional class; physician-rated global disease severity assessment (physician determined score based on their assessment of extent of damage as a consequence of SSc).
Better health status is <= median value of each variable as shown in Table I, worse health status is > median value of each variable in Table I.
#The PROMIS-29 provides a score for each health domain that is standardised against US normative population data. A score about 50 represents better than the average population, while a score below 50 is worse. Two exceptions are for anxiety and depression, where a score above 50 is worse than the general population and below 50 is better.

higher unemployment rate among Australian SSc patients compared with the general Australian population aged 25–64 years (44.8% *vs*. 4.5% respectively) and the higher absenteeism rate in SSc patients compared with the general Australian population with and without a chronic disease (average of 0.48 days off work versus 0.25 days off respectively a fortnight (23).

Of note, absenteeism in SSc correlated less well with health outcomes than presenteeism, overall work impairment or activity impairment in our study. In SSc, there are many factors that influence a patient's ability to maintain paid employment including the physical nature of the job, diffuse skin disease, the presence of digital amputation, pulmonary arterial hypertension and sicca symptoms (3). Furthermore, higher rates of absenteeism in SSc patients were associated with lower education and those in a manual job compared to those in non-manual jobs (5). Therefore, missing days from work is likely to depend on more than just the health outcomes covered in this study. It may depend on the weather or air-conditioning in the workplace in those with severe Raynaud's phenomenon and digital ulceration, educational level or the physical nature of the job, and ability to modify work hours and/ or environment. The phenomenon of reduced correlation of absenteeism with health outcomes is not unique to SSc and has been noted previously in RA and ankylosing spondylitis (10, 11). This suggests that absenteeism encompasses other dimensions of the disease, which are not currently measured by these health outcome instruments.

In our cohort, pain was the patient-reported outcome most strongly correlated with WPAI:SHP scores including absenteeism, presenteeism, overall work productivity and activity impairment. Pain has also been reported to be associated with impaired productivity in RA (10, 24) and with work disability in SSc (25).

In addition, physical function, measured by the HAQ-DI, was strongly cor-

related with presenteeism, overall work impairment and activity impairment in our study. High HAQ-DI scores have previously been associated with work disability in both SSc and RA (26, 27). High HAQ-DI scores have also been shown to be associated with activity impairment in SSc with one study reporting a decrease in household productivity by one day for every increase in HAQ-DI score of 1.0 unit after controlling for covariates (5).

Fatigue also strongly correlated with presenteeism, overall work impairment and activity impairment in our study. Fatigue is a very burdensome symptom that is particularly difficult to treat and a risk factor for work disability across a wide range of rheumatic diseases including SSc (28-30). Disease severity was also strongly associated with work disability in these studies, as it was in our study (28, 29).

"The WPAI:SHP has an important role in the clinical management of SSc patients. It is a short questionnaire with only six questions that could be completed by patients while waiting for their clinic appointment. There is increasing evidence that SSc patients experience a high rate of unemployment and work disability early in their disease course (26) and that unemployed SSc patients report worse health related quality of life scores than employed SSc patients (3). Therefore work disability has clinical implications in relation to patient management and the treating physician should be aware of it. Information on employment and work productivity is particularly important in identifying risk factors that hinder employment and productivity. In turn, this is important for creating employment strategies and workplace modifications to enable patients' employment and productivity to be maintained for as long as practical."

One limitation of this study is that it was cross-sectional. There is the potential for non-response bias between patients who did and did not complete the questionnaire. However, there were no statistically significant differences in patient characteristics or disease manifestations between responders and non-responders. Additionally, we did not use an independent employment measure of missed or impaired work hours to validate the self-reported hours. Of note, however, it has been shown that 95% of self reported sick days match registered data when the recall period is less than 2–4 weeks (31). We did not test reliability of the WPAI:SHP in SSc patients in this study although the test-retest reliability has been well established in other rheumatic conditions including AS (11).

Conclusion

Our study indicates that the WPAI:SHP questionnaire has construct validity for use in SSc patients. Therefore, the WPAI:SHP could be used to quantify work productivity in SSc, identify factors impacting productivity and highlight potential workplace modifications for improving productivity.

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