Sensitivity to Change in Systemic Sclerosis of the McMaster-Toronto Arthritis Patient Preference Disability Questionnaire (MACTAR): Shift in Patient Priorities Over Time

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ABSTRACT. *Objective.* To assess the sensitivity to change of the McMaster-Toronto Arthritis Patient Preference Disability Questionnaire (MACTAR) in systemic sclerosis (SSc) and a shift in patient priorities over time.

Methods. We assessed 49 patients with SSc (8 men) using the MACTAR in a prospective longitudinal study twice or more during annual meetings of the French patient association from 2004 to 2007. Patient-perceived improvement or worsening regarding health status was recorded. Sensitivity to change was assessed by the effect size (ES) and the standardized response mean (SRM) of the MACTAR.

Results. The MACTAR global score was significantly increased at followup in the whole group of patients, and the ES and SRM values were –0.37 and –0.34, respectively. These values were similar to those observed for widely used outcome measures for SSc such as the Health Assessment Questionnaire. As defined by the International Classification of Functioning, Disability and Health, the 3 disability domains most often cited at baseline were mobility (7 activities, cited 17 times; 33.3% of patients), domestic life (4 activities, cited 17 times; 33.3% of patients), and community, social and civic life (3 activities, cited 10 times; 19.6% of patients). At followup, 40 patients had changed their first priority and 34 changed 3 priorities.

Conclusion. The evolution in MACTAR global score over time for patients with SSc reflects longterm general feelings of deterioration. However, shifts in patient priorities are common and may influence the sensitivity to change of the instrument. (J Rheumatol First Release Jan 15 2010; doi:10.3899/jrheum.090632)

Key Indexing Terms:

MCMASTER-TORONTO ARTHRITIS PATIENT PREFERENCE DISABILITY
QUESTIONNAIRE (MACTAR)
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Systemic sclerosis (SSc) is a connective tissue disease characterized by excessive collagen deposition, vascular hyper-reactivity, and obliterative microvascular phenomena[1,2]. Patients with SSc are classified according to the extent of skin involvement: limited SSc (lSSc), with no detectable skin involvement; limited cutaneous SSc (lcSSc), with skin involvement essentially limited to the hands and face and rare visceral involvement (with the exception of 8%–12% of patients in whom pulmonary arterial hypertension and/or interstitial lung disease and/or bowel involvement eventually develop)[3,4]; and diffuse cutaneous SSc (dcSSc), with proximal skin involvement and frequent visceral involvement, which is responsible for decreased survival[5-7]. In addition to diminishing life expectancy, SSc is responsible for skin, tendon, joint, and vessel damage, which leads to handicap[8]. Therefore, outcome measures with good metric

properties assessing handicap are needed to measure disease evolution and treatment efficacy in SSc.

Global disability in SSc is usually measured by the Health Assessment Questionnaire (HAQ) or the scleroderma HAQ[9]. More detailed evaluation of disability has involved the use of location-specific disability scales such as the Cochin Hand Function Scale (CHFS)[10,11] and the Mouth Handicap In Systemic Sclerosis (MHISS) scale[12]. Hand and mouth disabilities have been shown to contribute to 75%[11] and 36%[12] of the HAQ variance, respectively. However, these measures do not survey patient priorities. Taking into account such priorities may lead to better addressing the validity and responsiveness of instruments[13]. An example of a functional scale that surveys patient priorities is the McMaster-Toronto Arthritis Patient Preference Disability Questionnaire (MACTAR)[14], which has been validated in patients with rheumatoid arthritis (RA) in a multicenter randomized trial of RA[15]. For the MACTAR, an interviewer elicits from patients the impaired activities that are most important to the individual patient and probes whether these problems are due to the illness. Because of this format, the MACTAR concept of function may be more comprehensive than that of traditional fixed-item questionnaires and may reveal issues that really matter to the patient.

We recently provided evidence in patients with SSc that the MACTAR score is weakly correlated with other disability measures and associated with patients' own opinions of their health status. These findings suggest that this score adds nonredundant and pertinent information to other scales for the assessment of health status in SSc[16]. However, before considering the MACTAR as an outcome measure in future trials of SSc, its sensitivity to change should be established. Two previous studies have suggested that patients with RA and patients with chronic low back pain (CLBP) frequently shifted their priorities over time and that may alter the validity of the MACTAR in patient followup[15]. Therefore, we aimed to assess the sensitivity to change of the MACTAR in SSc, the frequency of shifts in patients' priorities over time, and the implication of these factors on the usefulness of the MACTAR for SSc followup.

MATERIALS AND METHODS

Study design. Members of the Association des Sclérodermiques de France (ASF), the French association for patients with SSc, were surveyed during their annual meetings in 2004, 2005, 2006, and 2007. To be eligible for the study, patients had to fulfill the American College of Rheumatology[17] and/or the Leroy and Medsger[18] criteria for SSc. All patients were assessed during at least 2 of 4 annual meetings of the ASF in the spring (temperature 20°C). To assess sensitivity to change, patients who attended only 1 meeting were excluded. A total of 15 (4 men), 28 (7 men), 46 (8 men), and 34 (5 men) patients attending the 2004, 2005, 2006, and 2007 meetings, respectively, agreed to participate. Among these patients, 5 attended 4 meetings, 13 attended 3, and 31 attended 2. Baseline measurements were recorded at the first meeting and followup measurements at the last one, if patients attended more than 2 meetings (n = 18). Overall, 49 patients were included. The mean time between baseline and followup evaluations was 1.7 ± 0.8 years. Measurements recorded were age; sex; ethnicity; body mass index; age at disease onset; disease duration; disease form (lSSc, lcSSc, or dcSSc); Karnofsky Performance Status (KPS) Scale score (range 0–100)[16]; mouth opening (interincisor distance in mm); skin involvement; Raynaud's phenomenon; pitting scars; digital ulcers; calcinosis; gastrointestinal tract, joint, and/or muscle involvement; dyspnea (assessed by the New York Heart Association 4-point scale); interstitial lung disease; pulmonary arterial hypertension (PAH); and renal crisis. Evidence of gastrointestinal tract, joint, and/or muscle involvement; interstitial lung disease; PAH; and scleroderma renal crisis was based on patient reports.

Handicap and disability assessment. Patient priorities in disability were assessed by use of a French version of the MACTAR[14]. At baseline evaluation, patients were first asked about activities impaired by SSc. To assist the patient, the interviewer read a series of probing questions adapted to the French way of life. The MACTAR questions are open-ended and cover broad areas of function such as domestic care, self-care, professional activities, leisure activities, social interaction, and roles. Patients were encouraged to add activities not already listed. Then patients were asked to rank these activities in order of importance by answering, "Which of these activities would you most likely be able to do?". We introduced 2 modifications to the original MACTAR format. First, we used a "3-item priority function" and asked patients to identify and rank 3 situations among activities of daily living that caused them maximal trouble. Second, in the original MACTAR, items were not scored, but patients were asked if they had noticed changes in the problem they had identified several weeks ago. In the validation study of MACTAR, a Likert scale was added to quantify changes[15]. Thus, to reflect the degree of difficulty in performing a priority activity, each item was scored on an 11-point semiquantitative scale (0–10), the global score ranging from 0 (no disability) to 30 (maximal disability), as was done in the previous survey assessing patient disability priorities in SSc[16] and CLBP[19]. At followup evaluation, patients were not reminded of the 3 priorities they identified at baseline but were asked once again to define and score 3 activities they considered impaired by SSc.

To classify the activities identified by patients, we used the domains of the International Classification of Functioning, Disability and Health (ICF)[20] with the 10 linking rules given by the World Health Assembly in May 2001. According to these rules, each item of an activity should be linked to the most precise ICF category, and if concepts refer to more than 1 ICF category, then all the ICF categories to which the concepts refer should be linked[21]. So, one activity may correspond to 2 domains. For example, running belongs to the mobility domain (D 4552 running) and the community, social, and civic life domain (D 9201 sports).

Global disability was assessed by use of the HAQ[22], the scale ranging from 0 (no disability) to 3 (maximal disability). The HAQ comprises 20 items divided into 8 domains.

Hand disability was assessed by use of the CHFS[23], a questionnaire administered by the physician that contains 18 items related to daily activities, each question scored on a scale of 0 (performed without difficulty) to 5 (impossible to do). The total score was obtained by adding the scores of all items (range 0–90). This questionnaire has been validated in SSc[11].

Mouth disability was assessed by use of the MHISS scale, a questionnaire with 12 items concerning daily activities, each question scored on a scale of 0 (never) to 4 (always), which is administered by the physician[12]. The total score was obtained by adding the scores of all items (range 0–48).

Anxiety and depression assessment. Anxiety and depression were assessed by use of the Hospital Anxiety and Depression scale (HADa and HADd)[24]. This scale has 7 questions for the anxiety dimension and 7 for the depression dimension. Each point is scored on a scale of 0 to 3, the total score ranging from 0 (no depression, no anxiety), to 21 (maximal depression, maximal anxiety).

Health status and aesthetic burden assessment. In 2006 and 2007, patients were asked whether their health status related to SSc had improved, stabilized, or worsened, and aesthetic burden was assessed on an 11-point semiquantitative scale.

Ethical considerations. This survey was conducted in compliance with the protocol Good Clinical Practices and Declaration of Helsinki principles. In accord with French law, a formal approval from an ethical committee is not required for this kind of project. Patients gave their consent to participate after being informed about the study protocol.

Statistical analysis. Data analysis involved use of Systat 9 (SPSS Inc., Chicago, IL, USA). Quantitative variables are described with means ± standard deviations (SD) and ranges, and qualitative variables with percentages. The paired Student's t test was used to compare outcome measures at baseline and followup and the Mann-Whitney test was used to compare changes in scores between patients who deteriorated and those who did not. A p value < 0.05 was considered statistically significant.

Responsiveness is considered an aspect of validity[25] and describes a scale's ability to detect a clinically meaningful change over time[26]. Two different statistical approaches were used to assess responsiveness[26,27]. First, standardized response mean (SRM) is defined as the mean change in scores between the baseline and followup evaluations divided by the SD of the individual changes in scores. A higher SRM indicates greater responsiveness. A negative value indicates that the mean score at baseline is smaller than the mean score at followup. Second, effect size (ES) is defined as the mean change in scores between the baseline and followup evaluations divided by the SD of the baseline score. A higher ES indicates greater responsiveness. A negative value indicates that the mean score at baseline is smaller than the mean score at followup. The ES is considered small if < 0.2, moderate if close to 0.5, and large if > 0.8.

RESULTS

Demographic and clinical data. Mean age at the time of evaluation was 57.5 ± 11.8 years; 41 patients (83.7%) were women, and all were Caucasians. Mean disease duration was 16.1 ± 11.6 years. Twenty-three patients (47%) had dcSSc, 25 (51%) lSSc, and 1 (2%) lcSSc. The mean KPS was 77.6 ± 9 (range 60–90). All patients underwent a second evaluation at 1.7 ± 0.8 years (range 1–3 yrs).

Priority disabilities. Priorities were individual and different for each participant. Only 3 patients maintained their 3 priorities as defined at baseline. A total of 46 patients changed at least 1 of the 3 priorities. Among them, 4 changed one priority, 8 two, and 34 three. The different activities were classified according to the ICF[20] (Table 1). Among the domains chosen as the 3 priority activities at baseline, patients cited 32 activities and 2 body functions, which matched 7 ICF domains. Twenty-nine activities or body functions were cited less than 5 times and 10 only once. Only 5 were cited more than 5 times. The domains cited were mobility (12 activities, cited 46 times; 34.3% of patients); domestic life (7 activities, cited 37 times; 27.6% of patients); community, social, and civic life (7 activities, cited 35 times; 26.1% of patients); self-care (3 activities, cited 8 times; 6% of patients); interpersonal interactions and relationships (2 activities, cited 4 times; 3% of patients); major life areas (1 activity, cited twice, 1.5% of patients); and mental functions (2 functions, cited twice; 1.5% of patients). Among the domains chosen as the first priority, the 3 domains most often identified by patients were mobility (7 activities, cited 17 times; 33.3% of patients); domestic life (4 activities, cited 17 times; 33.3% of patients); and community, social, and civic life (3 activities, cited 10 times, 19.6% of patients).

The 5 activities most often cited at baseline were walking (n = 21, 42.9% of patients), sports (n = 21, 42.9% of patients), cleaning living area (n = 18, 36.7% of patients), driving (n = 8, 16.3% of patients), and taking care of plants (n = 6, 12.2% of patients). Eighteen different activities were ranked number 1, and of these, the 3 activities most often identified by patients as the first priority were cleaning living area (n = 10, 20.4% of patients), walking (n = 9, 18.4% of patients), and sports (n = 7, 14.3%).

At followup evaluation, a shift in priorities modified the order of the ICF domains when patients considered the 3 priorities or the first priority cited. Considering the 3 priorities, the activities and their frequencies in each domain changed in order, although sports remained the activity most cited (55.1% of patients). The order of the ICF domains was modified as follows: community, social, and civic life (40% of patients); domestic life (31.7%); mobility (14.2%); major life areas (7.5%); self-care (6.7%); interpersonal interactions and relationships (0%); and mental functions (0%). Considering the first priority cited, only 9 patients maintained their first priority as defined at the baseline evaluation, whereas 40 patients shifted their priorities. Among these patients, 34 also shifted their second and third priorities. In addition, a shift in priorities was associated with a shift in ICF domains. The order of the ICF domains was modified in that domestic life became the first priority domain (3 activities, cited 22 times), followed by community, social, and civic life (4 activities, cited 8 times), and major life areas (1 activity, cited 6 times). Overall, for 29 patients (59.2%), a shift in the first priority induced a change of the corresponding ICF domain.

Sensitivity to change. The mean MACTAR total score for the 49 patients at baseline and at final evaluation was 14.7 ± 5.8 (4–25) and 16.8 ± 5.5 (5–26), respectively (Table 2). We observed statistically significant differences between baseline and followup evaluations for the HAQ (p = 0.01), MACTAR (p = 0.01), and HADa (p = 0.04), and a tendency toward significance for CHFS (p = 0.055).

The responsiveness of the measures evaluated by the SRM and ES for the MACTAR total score was among the highest of the measurements studied, with –0.34 for the SRM and –0.37 for the ES (Table 2). The SRM and ES values for the MACTAR score were similar to those observed for the HAQ (–0.41 and –0.38, for SRM and ES, respectively).

Health status and sensitivity to change. When considering patients' perceived improvement or worsening regarding their health status, 34 patients (6 men) considered themselves as deteriorated, 12 (2 men) as unchanged, and 3 (0 men) as improved. To assess the difference significance, we recoded the current health status in 2 groups: deteriorated (n = 34) or not deteriorated (n = 15). In the group of patients considering themselves as deteriorated, the MACTAR ES (–0.48) and SRM (–0.39) values were similar to

Table 1. MACTAR activities identified by the 49 patients with SSc at baseline and at followup evaluations and classified according to the ICF. In the patient columns, the domain percentages refer to 100% and the activity percentages to 49 patients. The total activities in each domain could be above 100. The total activity percentage is more than 100% and the total domain percentage is 100%.

Activities, Participation, and Body Functions	Baseline Evaluation				Followup Evaluation			
	Three Priority Activities		First Priority Activity		Three Priority Activities		First Priority Activity	
	No. of Times Cited	% of Patients	No. of Times Cited	% of Patients	No. of Times Cited	% of Patients	No. of Times Cited	% of Patients
Chapter 4: Mobility** (n = 12 activities)	46	34.3	17	33.3	17	14.2	2	4.8
4509 Walking unspecified	21	42.9	9	18.4	8	16.3	0	0
4751 Driving motorized vehicles	8	16.3	1	2	4	8.2	1	2
4602 Moving around outside the home and other buildings	4	8.2	1	2	2	4.1	0	0
4551 Climbing	3	6.1	0	0	1	2	1	2
4401 Grasping	2	4.1	2	4.1	0	0	0	0
4409 Fine hand use unspecified	2	4.1	2	4.1	0	0	0	0
4309 Lifting and carrying unspecified	1	2	0	0	0	0	0	0
4359 Moving objects with lower extremities unspecified	1	2	0	0	1	2	0	0
4453 Turning or twisting the hands or arms	1	2	1	2	0	0	0	0
4552 Running	1	2	1	2	0	0	0	0
4554 Swimming	1	2	0	0	0	0	0	0
4750 Driving human-powered transportation	1	2	0	0	1	2	0	0
Chapter 6: Domestic life** (n = 7 activities)	37	27.6	17	33.3	38	31.7	22	52.4
6402 Cleaning living area	18	36.7	10	20.4	18	36.7	14	28.6
6505 Taking care of plants, indoors and outdoors	6	12.2	2	4.1	5	10.2	0	0
609 Preparing meals unspecified	3	6.1	0	0	7	14.2	6	12.2
6200 Shopping	3	6.1	2	4.1	6	12.2	2	4.1
6409 Doing housework unspecified	3	6.1	3	6.1	0	0	0	0
649 Household tasks other specified and unspecified	3	6.1	0	0	0	0	0	0
6500 Making and repairing clothes	1	2	0	0	2	4.1	0	0
Chapter 9: Community, social and civic life** (n = 7 activities)	35	26.1	10	19.6	48	40	8	19
9201 Sports	21	42.9	7	14.3	27	55.1	5	10.2
9203 Crafts	4	8.2	2	4.1	2	4.1	1	2
9209 Recreation and leisure unspecified	4	8.2	1	2	6	12.2	0	0
999 Community, social and civic life unspecified	3	6.1	0	0	9	18.4	1	2
9205 Socializing	2	4.1	0	0	0	0	0	0
9204 Hobbies	1	2	0	0	2	4.1	0	0
9202 Arts and culture	0	0	0	0	2	4.1	1	2
Chapter 5: Self-care** (n = 3 activities)	8	6	6	11.7	8	6.7	4	9.5
550 Eating	4	8.2	4	8.2	4	8.2	3	6.1
5109 Washing oneself unspecified	2	4.1	1	2	0	0	0	0
5409 Dressing unspecified	2	4.1	1	2	4	8.2	1	2
Chapter 7: Interpersonal interactions and relationships** (n = 2 activities)	4	3	0	0	0	0	0	0
7500 Informal relationships with friends	2	4.1	0	0	0	0	0	0
7702 Sexual relationships	2	4.1	0	0	0	0	0	0
Chapter 8: Major life areas** (n = 1 activity)	2	1.5	0	0	9	7.5	6	14.3
850 Remunerative employment	2	4.1	0	0	9	18.4	6	12.2
Chapter 1: Mental functions* (n = 2 functions)	2	1.5	1	2	0	0	0	0
1449 Memory functions unspecified	1	2	1	2	0	0	0	0
1349 Sleep functions unspecified	1	2	0	0	0	0	0	0
Total	134		51		120		42	

* Domains of body functions. ** Domains of activities and participation. SSc: systemic sclerosis; MACTAR: McMaster-Toronto Arthritis Patient Preference Disability Questionnaire; ICF: International Classification of Functioning, Disability and Health.

those in the group of patients considering themselves as not deteriorated (ES –0.42 and SRM –0.37; Mann-Whitney test p value = 0.91).

DISCUSSION

Our finding of the evolution in the MACTAR global score over time in patients with SSc reflects the general feeling of deterioration among patients with SSc and provides qualitative information about their health status over time. Shifts in patient priorities are common and probably influence sensitivity to change of the instrument.

Among the outcome measures studied in this survey,

Table 2. HAQ, KPS, MHISS, MACTAR, and CHFS scores, as well as aesthetic burden, anxiety, and depression assessment at baseline visit, at followup visit and their differences, and responsiveness of the disability and handicap measures for the 49 patients with SSc.

Outcome Measures (range)	Baseline Evaluation				Followup Evaluation				Difference				ES	SRM	p*
	Mean	SD	Minimum	Maximum	Mean	SD	Minimum	Maximum	Mean	SD	Minimum	Maximum			
HAQ (0–3)	1.2	0.6	0	2.4	1.4	0.9	0	3	–0.2	0.6	–1.4	0.8	–0.38	–0.41	0.01**
KPS (0–100)	77.6	9	60	90	78	7.6	60	90	–0.7	8.3	–20	20	–0.04	–0.04	0.6
MHISS (0–48)	19.2	10.6	0	38	19.3	9.8	2	37	–2.3	8.2	–34	11	–0.01	–0.02	0.13
MACTAR (0–30)	14.7	5.8	4	25	16.8	5.5	5	26	–2.3	6.3	–14	14	–0.37	–0.34	0.01**
CHFS (0–90)	20.1	16.5	0	55	22.7	16.6	0	62	–2.6	9.3	–26	17	–0.16	–0.28	0.055
HADa (0–21)	10.4	4.5	1	19	9.5	4.3	1	19	1	3.3	–5	–6	0.20	0.27	0.04**
HADd (0–21)	7	3.9	0	15	6.6	3.6	1	16	0.4	2.8	–6	6	0.11	0.15	0.30
Aesthetic burden (0–10)	5.5	2.3	1	9	4.5	2.7	0	9	0.5	1.8	–2	5	0.46	0.60	0.28

* Comparisons were performed using paired samples t test. ** Difference is significant. SSc: systemic sclerosis; HAQ: Health Assessment Questionnaire; KPS: Karnofsky Performance Status Scale; MHISS: Mouth Handicap In Systemic Sclerosis Scale; MACTAR: McMaster-Toronto Arthritis Patient Preference Disability Questionnaire; CHFS: Cochin Hand Function Scale; HADa: Hospital Anxiety and Depression scale for Anxiety; HADd: Hospital Anxiety and Depression scale for Depression; ES: effect size; SRM: standardized response means. P value: comparison between outcome measures at baseline and at followup.

MACTAR scale and HAQ had the highest ES and SRM values. In addition, previous studies suggested that the HAQ has acceptable sensitivity to change in SSc[9,29], especially for clinical measures[30]. Altogether, these findings suggest that the MACTAR scale is a responsive outcome measure in patients with SSc.

Shifts in priorities occurred in almost all the patients with SSc (n = 46, 93.9%) and had several implications. They modified domains according to the ICF classification when considering the first or the 3 priorities cited at followup evaluation. There was also a shift when considering activities and their frequencies in each domain, suggesting that even in a chronic disabling disease condition, patients' expectations change over time. Thus, one advantage of using the MACTAR is that, eventually, it identifies well what is most important for a patient at one moment and shows how handicaps may change with time. In epidemiological surveys aimed at describing clinical situations and their evolution, taking into account patient shifts in priorities may add useful information about the evolution and the types of participation limitations over time. Our findings are consistent with those of another study of patients with RA, which demonstrated that two-thirds of prioritized impaired activities were new after 1 year of followup[15]. This observation could also help in developing more individual therapeutic strategies.

One limitation of our study was that we did not remind patients of their baseline priorities and therefore could not assess changes in restriction of participation over time for these priorities. Taking account of shifts in priorities for calculating MACTAR global score may lead to modifying its sensitivity to change. Because the aim is to reduce participation limitations in activities defined as priorities at baseline, it would be interesting, in addition to a shift in priorities, to consider baseline priorities to calculate the MAC-

TAR global score at followup by reminding patients of the activities and scores they cited at baseline. Another limitation is that, because of the way patients were recruited, reproducibility could not be assessed.

Even though the MACTAR approach closely reflects real-life participation limitations and may be helpful for clinical decisions, the tool has potential limitations for routine use. Use of the MACTAR requires trained interviewers and might not be an easy, cost-effective instrument to use routinely[15]. This point should be assessed in further studies. Whether the instrument would measure real changes rather than just unrealistic desires is also unclear. This latter limitation also applies to outcome measures with predefined items assessing participation limitations. These measures are widely used in the clinic for conditions such as SSc.

Another limitation of our study may be the procedure used to recruit patients. Since all patients belonged to the French association of patients, they may not be representative of the whole French SSc population. Thus, more than two-thirds of the patient members of this association self-reported worsening of their health status during followup, while only 6% felt improved, and their HAQ scores were quite high and comparable to those reported in a study conducted in a tertiary care setting[31]. In addition, patients had longstanding disease, which could imply more symptoms. Therefore, further evaluation in other cohorts of patients with SSc is necessary to confirm the sensitivity to change of the MACTAR in SSc.

The MACTAR provides a qualitative tool for identifying the deterioration in condition of patients with SSc. Recording shifts in priorities provides a qualitative analysis of participation limitations that guarantees more accurate information about what matters the most to patients. However, longer followup is needed to better characterize the changes in the patients' priorities, and clinical investiga-

tors must be aware that taking into account shifts in priorities may modify sensitivity to a change of the MACTAR scale.

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