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Contact. **The Managing Editor, The Journal of Rheumatology, 365 Bloor Street East, Suite 901, Toronto, ON CANADA M4W 3L4. Tel: 416-967-5155; Fax: 416-967-7556; E-mail:jrheum@jrheum.com Financial associations or other possible conflicts of interest should always be disclosed.**

The Medical Outcome Study Short Form 6D and Whiplash

To the Editor:

Faced with ever-increasing demands and costs for healthcare services, health authorities and public and private insurers have become interested in the effectiveness and cost-effectiveness of healthcare interventions. Patient-perceived health is an important healthcare outcome, relevant to patients and therefore relevant to those who evaluate intervention outcomes. Previously in *The Journal*, Russell, *et al* showed that the Medical Outcome Study Short Form 6D (SF-6D), a preference based measure for health related quality of life (HRQOL) derived from the Medical Outcomes Study Short Form-36 (SF-36), was sensitive to changes in health status of patients with rheumatoid arthritis (RA) treated with infliximab[1]. Thus, future studies may utilize measures like the SF-6D in evaluating the cost-effectiveness of interventions for RA and other musculoskeletal disorders.

Among musculoskeletal disorders, whiplash-associated disorders (WAD) have a significant economic and health importance[2]. The health burden of WAD is huge for both the individual and society, the latter in terms of direct costs of care. The societal costs of absenteeism from work may perhaps be second only to those of low back pain. Studies in Canada[3,4], Sweden[5], the United States[6], the United Kingdom[7,8], Ireland[9], and Norway[10] have shown that as many as 50% of victims of whiplash injury following a motor vehicle collision will still be experiencing chronic neck pain and disability 6 months after the collision. Similar results have been found in other countries. While litigation costs are significant, much of the cost of WAD can also be attributed to the investigation, assessment, and attempted treatment for chronic pain. It is known, for example, that while only 12.5% of whiplash patients in Quebec remained absent from work due to chronic pain 6 months after their collision, these 12.5% accounted for 46% of total costs of all whiplash injuries[2]. Approaches that prevent the progression from a transient, acute injury to chronic pain would be of major importance to the injured individual and to society. At the same time, however, many treatment approaches to WAD are proposed and are costly. We suggest that WAD interventions can and should be evaluated with similar HRQOL measures as many other illnesses. In that regard, the SF-6D may be useful.

The SF-6D was derived by Brazier, *et al* as a preference based single index from the SF-36[11,12]. The main approach in health economics has been to value health status in a single unit of measurement known as "quality adjusted life years" (QALY), or "well years." The index or "utility" scale is anchored on 0 (death) and 1 (full health), and is integrated with survival, so that it is not merely the number of years of life expectancy but also the quality of those years that is considered. Brazier, *et al* developed the SF-6D to reconcile the generic health status measure, the SF-36, with the QALY approach. The result of their work is the 6-dimensional health classification. Each dimension of the SF-6D has up to 6 ranked statements or levels, such as "you have no pain" through "you have pain that interferes with your normal work (both outside the home and housework) extremely." A health state is composed of statements from each of 6 dimensions, starting with physical functioning and ending with vitality. A total of 18,000 possible health states are defined this way. Various studies have begun to evaluate the usefulness, interchangeability (with other utilities), and responsiveness to change of the SF-6D in a number of illnesses including RA, irritable bowel syndrome, chronic lung disease, and a general population of patients with musculoskeletal disease[1,13-16].

A large epidemiological study of WAD outcomes in Saskatchewan has suggested that WAD is in many ways a profound, systemic-like illness for a significant number of afflicted individuals, with numerous symptoms having an effect measureable by the SF-36[3]. Taking a sample of tertiary referral WAD patients with chronic pain, we found that a comparable result can be seen with the SF-6D. We examined a group of 135 patients and asked them to complete the SF-36. The group consisted of 75% females, with a group mean age of 39.5 years (SD 13.0), and mean duration of illness was 9 months (SD 9 mo). The mean SF-6D index in this group was 0.57 (SD 0.09, range 0.37 to 0.89). There was no significant correlation between the SF-6D and duration of illness ($r = -0.08$).

An SF-6D scale score of 0.57 indicates significant effect of WAD on HRQOL. The clinical importance of this value can also be appreciated by comparison with 60 patients with severe, active RA immediately prior to receiving biological agents[1]. These patients were found to have an SF-6D score of 0.55. Clearly, the patients with RA have substantial pathology and the whiplash patients, all of whom were grade 1 and 2 WAD, have, by classification definition, no objective pathology[2]. It is difficult to accept that the SF-6D score is a measure of the direct effects of neck injury, but more likely it reflects that even "medically unexplained symptoms" have a significant impact on the HRQOL. In WAD, the environment in which symptoms exist (including litigation), the mixed messages from multiple therapists, the belief that one has significant and incurable damage, and that activity that hurts may be harmful could all contribute to the HRQOL. Studies in the analogous problem of low back pain demonstrated that the chronicity of low back pain can be mimimized by appropriate education with a brief but clear educational pamphlet explaining the importance of continuing normal activities[17]. And a recent study has shown that a "whiplash booklet" can alter the beliefs about whiplash injury among both injured and non-injured subjects[18]. We further know that the symptoms of daily life, of benign acute pain disorders, and even simple chest infections can be amplified through emotional distress and close attention to symptoms and thus lead to adverse health perceptions[19-23]. Given this and the knowledge that legislative changes can also affect WAD outcomes[3], it seems reasonable that evaluations of treatment interventions and legislative changes on the outcome of WAD could involve the use of SF-6D. As health authorities and governments deal with rising healthcare and insurance costs associated with WAD, the SF-6D is a convenient and simple tool to provide the utility measure that these bodies look to in evaluating the effectiveness and cost-effectiveness of proposed interventions.

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Clinical Characteristics of Juvenile Systemic Sclerosis in Japanese

To the Editor:

The severity of systemic sclerosis (SSc) is highly variable. Since disease onset before age 16 years is quite rare in patients with SSc[1], the clinical characteristics of juvenile SSc have not been fully clarified to date. We determined retrospectively the clinical features of Japanese SSc patients younger than age 16 years in comparison with adult Japanese patients with SSc. All 184 SSc patients referred to our hospital during a period of 11 years (1992–2003) were eligible for the study. All patients fulfilled the criteria for SSc proposed by the American College of Rheumatology[2]. Patients with localized scleroderma were excluded. The cases were grouped into diffuse cutaneous SSc (dSSc) or limited cutaneous SSc (lSSc) according to the classification system proposed by LeRoy, *et al*[3]. Complete histories, physical examinations, and laboratory tests including high resolution computed tomography (HRCT) of the chest were conducted for all patients at the first visit and during followup. We evaluated general health by blood sampling and urinalysis at every third month, and cardiopulmonary condition by chest radiograph semiannually, with more examinations if necessary. Organ system involvement was defined as described[4,5].

Among 184 Japanese SSc patients, 5 were classified as having juvenile SSc. Clinical profiles of these patients are summarized in Table 1. Concerning the individual profiles of these patients, a male patient (Patient 1) first presented at the age of 6 months, with skin sclerosis on the foot that was noted by his mother. At the first visit, at age 2 years, 11 months, he showed skin sclerosis of the whole body, with 24 points on the Modified Rodnan total skin thickness score (TSS). Assessment was the same as for an adult assessment and the TSS obtained was the average score of 2 physicians. Skin biopsy of the foot revealed an increase in thickened collagen fibers in the dermis. He had no internal involvement other than esophageal hypomotility. His skin sclerosis gradually improved to 15 TSS points, 9 years after starting treatment with prednisolone (PSL).

A 7-year-old girl (Patient 2) with disease onset at age 5 years was admitted to our hospital because of skin sclerosis on the extremities, with 27 TSS points. She had no internal involvement. One year after starting treatment with PSL, skin sclerosis improved to 17 TSS points.

A 13-year-old boy (Patient 3) who developed skin sclerosis on the hands at age 8 years was admitted to hospital with whole-body skin sclerosis, with 27 TSS points. Although treatment with PSL had been started at age 11 years at another hospital, skin sclerosis had not improved. HRCT showed pulmonary fibrosis that had developed slowly.

In a girl (Patient 4) who presented with Raynaud's phenomenon at age 9 years, skin sclerosis had rapidly developed on the upper arms within 6 months, resulting in 19 TSS points. Treatment with PSL was started and skin sclerosis improved to 15 points after 5.5 years' followup.

A boy (Patient 5) experienced Raynaud's phenomenon at age 13 years; skin sclerosis developed on the upper arms, but not the trunk, at age 17 years (TSS 11 points). He was treated with PSL, which appeared to be effective for his skin sclerosis (TSS 8 points after 8 years' PSL treatment).

The clinical and laboratory features of juvenile SSc were compared with those of adult SSc. The prevalence of dSSc in patients with juvenile SSc was significantly higher than in patients with adult SSc ($p < 0.05$; Table 2). Furthermore, TSS of those with juvenile SSc were significantly higher than those of adult SSc ($p < 0.05$). There was no difference in the prevalence of organ involvement between juvenile and adult SSc patients. Juvenile SSc exhibited an increased frequency of antitopoisomerase I (topo I) antibody positivity compared to adult SSc patients (100% vs 29%; p <

Table 1. Clinical course of 5 patients with juvenile SSc.

Patient	Sex	Age at Onset	Age at Presentation	Followup Period	TSS	Organ Involvement	Autoantibodies	PSL Therapy (first dose; mg/kg/day)	Outcome of Skin Sclerosis
1	M	6 mo	2 yrs, 11 mo	9 yrs	24	Esophagus	Topo I	0.36	Improvement
2	F	5 yrs	7 yrs	1 yr	27	—	Topo I	1.2	Improvement
3	M	8 yrs	13 yrs	2 yrs	27	Lung	Topo I	0.35	No change
4	F	9 yrs	9 yrs, 6 mo	5 yrs, 6 mo	19	Esophagus	Topo I	0.36	Improvement
5	M	13 yrs	17 yrs	8 yrs	11	Esophagus	Topo I	0.3	Improvement

Topo I: antitopoisomerase I antibodies, PSL: prednisolone, TSS: Total Skin Score.

Table 2. Clinical and laboratory features of juvenile SSc patients in comparison with adult SSc patients. Unless noted otherwise, values are percentages.

	Vesely[10] (n = 12)	Foeldvari[11] (n = 135)	Lababidi[12] (n = 5)	Suarez-Almazor[7] (n = 5)	Vancheeswaran[1] (n = 11)	Cassidy[13] (n = 15)	Martinez-Cordero[8] (n = 7)	Current Study Juvenile SSc (n = 5)	Current Study Adult SSc (n = 179)	Current Study Adult SSc with Anti-Topo I Antibodies (n = 52)
Country	Italy	Europe	Lebanon	Argentina	UK	USA	Mexico	Japan	Japan	Japan
Sex, male:female	4:8	35:100	1:4	1:4	4:7	0:15	1:6	2:3	24:155	8:44
Clinical features										
TSS points, mean ± SD	ND	ND	ND	ND	ND	ND	ND	21.6 ± 6.8*	10 ± 9.3	15.5 ± 10.5
dSSc	100	ND	ND	80	ND	ND	ND	100*	40	40
Raynaud's phenomenon	81	72	60	100	82	73	71	80	74	72
Pitting scar	ND	ND	60	100	ND	60	ND	100*	35	58
Contracture of phalanges	ND	ND	60	ND	ND	67	ND	60	45	65
Organ involvement								60	74	72
Lung	60	50	20	20	ND	73	100	20**	43	88
Heart	0	44	0	20	ND	13	0	20	16	26
Kidney	0	13	0	ND	ND	ND	0	0	1.7	6
Esophagus	45	47	40	40	ND	73	100	80	67	71
Joint	45	79	80	100	ND	60	71	20	26	29
Muscle	27	10	20	60	ND	27	29	20	14	14
Laboratory data										
Elevated CRP	ND	ND	ND	ND	0	ND	ND	0	15	24
Elevated ESR	ND	ND	40	ND	ND	ND	0	0**	31	40
Elevated IgG	ND	ND	100	ND	ND	ND	57	20	36	47
Positive ANA	ND	ND	20	100	45	57	43			
Positive anti-topo I antibodies	ND	50 (2/4)**	ND	40	27	ND	43	100*	29	100
Positive ACA	ND	ND	ND	0	0	ND	0	0	40	0

* $p < 0.05$ vs patients with adult SSc in the present study. ** $p < 0.05$ vs patients with adult SSc patients positive for anti-topo I antibodies in the present study. TSS: Modified Rodnan total skin thickness score; CRP: C-reactive protein; ESR: erythrocyte sedimentation rate; ANA: antinuclear antibody; ACA: anticentromere antibody; ND: not done.

0.05). When profiles of juvenile SSc patients were compared with adult SSc patients positive for anti-topo I antibodies, pulmonary fibrosis was present in only 20% of juvenile SSc patients, significantly lower than the frequency (88%) in adult patients ($p < 0.05$; Table 2).

Studies in other countries have reported that juvenile SSc patients had more severe skin sclerosis (Table 2)[6,7]. The frequency of dSSc (80–100%), Raynaud's phenomenon (60–100%), and contracture of phalanges was high. These observations were consistent with the result of our study. Previous studies reported low prevalence of scleroderma renal crisis[8]: scleroderma renal crisis was not detected in any patient in our study. Other studies have shown that anti-topo I antibodies are frequently detected (27–50%) in juvenile SSc, whereas anticentromere antibodies are rarely detected (Table 2). Although a previous study reported relatively higher mortality in patients with juvenile SSc[9], a recent study of 135 juvenile SSc patients showed a favorable outcome[11]. In our study, all juvenile SSc patients survived for the followup period of 9 years. Although racial differences may affect the mortality rate of juvenile SSc, larger longitudinal studies will be needed to confirm mortality rates.

Our study suggests that Japanese patients with juvenile SSc have more severe skin sclerosis than adults with SSc, although the frequency of internal organ involvement and the mortality rate is lower than with adult SSc.

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Corrections

Raskar JJ, Arnauts FLJ. The "New" International League for Rheumatology [editorial]. J Rheumatol 2005;32:1177–81. The title should correctly be, The "New" International League of Associations for Rheumatology. We regret the error.

Devauchelle Pensec V, Saraux A, Berthelot JM, et al. Ability of foot radiographs to predict rheumatoid arthritis in patients with early arthritis. J Rheumatol 2004;31:66-70. The name of the first author should correctly be spelled Valérie Devauchelle-Pensec. We regret the error.

Pease CT, Haugeberg G, Morgan AW, Montague B, Hensor EMA, Bhakta BB. Diagnosing late onset rheumatoid arthritis, polymyalgia rheumatica, and temporal arteritis in patients presenting with polymyalgic symptoms. A prospective longterm evaluation. J Rheumatol 2005;32:1043-6. Table 1: the line in the left column, "Arthritis at any time during followup," should give the data 30 (39) in the column for PMR. Figure 1 should appear as follows. We regret the errors.