Development of a Modified Hand Mobility in Scleroderma (HAMIS) Test and its Potential as an Outcome Measure in Systemic Sclerosis

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ABSTRACT. Objective. To modify the hand mobility in scleroderma (HAMIS) test by reducing the number of items and amount of equipment needed, and to evaluate the construct validity of this modified HAMIS (mHAMIS).

Methods. Our retrospective study is based on 266 patients previously examined using the original HAMIS test. Data were divided into 3 groups depending on disease duration after onset: (1) 0–3 years, (2) 3.1–5 years, and (3) 5.1–9 years. Disease variables included were skin involvement using the disease subset and the modified Rodnan skin score (mRSS), and digital lesions. Cronbach’s alpha coefficient was calculated separately for limited (lcSSc) and diffuse systemic sclerosis (dcSSc) for the right and left hand, and for the groups with different disease duration. The construct validity of the mHAMIS was assessed by searching for a correlation with hand skin score.

Results. An mHAMIS test consisting of finger flexion, finger extension, finger abduction, and dorsal extension was developed. The internal consistency of this test was 0.78, 0.83, and 0.73 in the 3 groups with different disease duration. In the whole study group, mHAMIS showed a significant correlation with mRSS and hand skin score (rs = 0.39 and 0.43, respectively), and was able to discriminate between lcSSc and dcSSc (p = 0.001), and between patients with and without ulcers (p = 0.015).

Conclusion. The mHAMIS involves 4 easily measurable items and has the potential to be a relevant clinical measure of outcome in the evaluation of fibrotic skin involvement in SSc. (First Release Oct 1 2014; J Rheumatol 2014;41:2186–92; doi:10.3899/jrheum.140286)

Key Indexing Terms: SYSTEMIC SCLEROSIS HAND MOBILITY IN SCLERODERMA MODIFIED HAND MOBILITY IN SCLERODERMA

Systemic sclerosis (SSc; scleroderma) is an autoimmune disease characterized by microvascular injury and excessive fibrosis of the skin and internal organs. Although there is considerable individual variation, skin involvement tends to reach a maximum within the first 3 years, after which the skin becomes thinner, although complete remission in the fingers and hands is less common. Skin thickening is a manifest consequence of SSc. Skin thickness is determined by using the modified Rodnan skin score (mRSS), the most commonly used and valid method of assessing skin involvement in SSc, and meets the requirements of an outcome measure according to the Outcome Measures in Rheumatology Clinical Trials (OMERACT) filter. However, the mRSS provides a measure of the skin involvement of the whole surface of the body, and not specifically the hands. The hands account for only a small fraction of the total skin area of the body, but are necessary for many important functions in daily life. It is, therefore, important to study hand involvement separately, especially in studies on the outcome of therapy.

Hand involvement in SSc includes skin thickening, presumably caused by inflammatory edema and fibrosis, as well as vascular injury and articular involvement. Individuals may perceive impairment in mobility, hand strength, and dexterity, having a significant influence on the activities of daily living. Hand involvement is one of the early signs of the disease, and there is thus a need for tests that can provide information on the efficacy of therapeutic interventions aimed at minimizing functional impairment. In addition, there is a need for feasible endpoints in routine followup and clinical trials. The guidelines of the OMERACT filter, and a combination of measures including body function, body structures, and activity and participation according to the different domains of the International Classification of Functioning (ICF) are useful tools in the selection of appropriate outcome measures in studies on the outcome of therapy.
measures. The Cochin Hand Function Scale (CHFS)\textsuperscript{12} is a self-reported disability questionnaire that provides a reliable and valid measure within the activity domain, with significant correlations with activities of daily living in SSC\textsuperscript{13,14}. On the body function level, the performance-based Arthritis Hand Function Test (AHFT), measuring hand strength and dexterity\textsuperscript{15}, and the Hand Mobility in Scleroderma (HAMIS) test\textsuperscript{16}, measuring hand mobility, have been used in the assessment of hand function in SSC\textsuperscript{16,17}. The AHFT measures different aspects of hand function, and has shown significant correlations with the performance of activities of daily living, but not with skin involvement\textsuperscript{17}.

The HAMIS test focuses on the mobility of the fingers and wrist, and comprises 9 items using differently sized grips and different movements\textsuperscript{16}. The HAMIS test usually takes 10 min to perform in a patient with SSC. Advantages of the HAMIS test are the feasibility, the high interobserver and intraobserver reliability ($\kappa = 0.82$ and $0.74$, respectively)$\textsuperscript{13}$, and the test-retest reliability (ICC = 0.99)$\textsuperscript{7}$. Further, the test has shown significant correlations with skin involvement in the hands in both cross-sectional and longitudinal followup studies, and with activities of daily living$\textsuperscript{7,9,18,19}$. HAMIS has also been shown to be sensitive to change, and to be a useful outcome measure in hand rehabilitation$\textsuperscript{20,21}$.

The HAMIS test clearly reflects the consequences of skin involvement on hand function. The test is not particularly time-consuming, but requires some equipment, which may be one reason why the test has not been used in clinical trials. To improve the feasibility of the HAMIS test in clinical trials, and to investigate its ability to measure the consequences of the fibrotic involvement of the hands, we decided to reevaluate the HAMIS test. The aims of our present study were thus to modify the original HAMIS test by reducing the number of items and the equipment needed (to provide a more feasible test for clinical trials), and to evaluate its construct validity in patients with different durations of SSC.

**MATERIALS AND METHODS**

**Study design.** Our retrospective cross-sectional study was conducted at 1 rheumatology department at a single center, and was based on measurements made with the HAMIS test between 1998 and 2011. Four experienced occupational therapists performed the assessments during this period. To elucidate possible differences during the edematous, indurative, and atrophic phases of the skin disease, the data were divided into 3 groups based on the date of assessment in relation to the duration of the disease: group (1) 0–3 yrs, (2) 3.1–5 yrs, and (3) 5.1–9 yrs after disease onset. Because the same patient may have been assessed during more of these periods, and may also have been assessed more than once in the same period, the patient’s first assessment in each period was used to avoid possible duplication of data.

**Ethical considerations.** Our study was conducted in accordance with the Declaration of Helsinki and was approved by the Regional Ethics Committee in Lund. The patients were given verbal information on the aim of our study, and written consent was obtained according to the Declaration of Helsinki.

Subjects. All patients who were assessed twice or more during the period between 1998 and 2011, within the context of our regular followup program, and who fulfilled the American College of Rheumatology criteria for SSC\textsuperscript{22} were included. Two hundred sixty-six patients were included: 218 with limited cutaneous SSC (lcSSc) and 48 with diffuse cutaneous SSC (dcSSc). Disease onset was defined as the time of the first non-Raynaud manifestation. The relevant clinical characteristics of the patients participating in our study are presented in Table 1.

**Instruments.** The patient’s skin involvement was characterized by subdivision into lcSSc or dcSSc, the measurement of the mRSS total skin score based on palpation of 17 anatomic sites, and a subscore measuring skin involvement on the fingers, the hands, and the forearms (here referred to as the hand skin score). Digital vascular lesions were defined as the presence or absence of digital tip ulceration or pitting scars. Hand mobility was assessed with the HAMIS test. Both hands were assessed separately, giving a score of 0 to 27 for each hand\textsuperscript{16}. Objective disease variables were assessed within the context of our regular followup program.

**Statistical analysis.** Clinical data are presented as the median and interquartile range (IQR), and as numbers and percentages. Cronbach’s alpha coefficient was used to estimate the internal consistency of the HAMIS test in the different groups. According to Bland and Altman\textsuperscript{23}, $\alpha$ values of 0.7 to 0.8 are considered acceptable for scales that are used to compare groups in research studies. To estimate the construct validity of modified HAMIS (mHAMIS), and the degree to which mHAMIS and the skin score are related, the convergent validity of the mHAMIS test was assessed by searching for a correlation with the hand skin score. Moderate to high correlations were expected, and $r_s$ values were interpreted as follows: $r_s < 0.25$ as little or no correlation, $r_s = 0.26–0.49$ as low correlation, $r_s = 0.50–0.69$ as moderate, $r_s = 0.70–0.89$ as high, and $r_s = 0.90–1.00$ as very high correlation\textsuperscript{24}.

The Mann-Whitney U test was used to assess the ability of the mHAMIS test to discriminate between the presence or not of ulcers or pitting scars, and between lcSSc and dcSSc.

**RESULTS**

**Hand mobility and skin involvement in groups with different disease duration.** Of the total number of 266 patients, 109 patients were categorized in Group 1 (0–3 yrs of disease duration), 118 in Group 2 (3.1–5 yrs of duration), and 143 in Group 3 (5.1–9 yrs of duration). All groups consisted mainly of women with lcSSc (Table 1). Hand mobility was 1 unit more impaired in the right hand in all groups ($p = 0.001$). The median HAMIS score in Group 1 was 7 points (4 points in the right hand and 3 points in the left), and 6 points in both Groups 2 and 3 (3 points in both the right and left hands in Group 2, and 3 points in the right hand and 2 points in the left hand in Group 3). Finger flexion, finger extension, and thumb abduction were more impaired in the right hand in all groups ($p < 0.05$). In Group 1, the skin scores for the fingers and the hands were higher on the right hand than on the left hand ($p = 0.004$ and $p = 0.035$, respectively). The distribution of the HAMIS scores in the 3 groups shows that finger flexion and extension were the most impaired functions in all groups, while pronation was impaired in only about 10% of the patients (Figure 1).

**Development of the mHAMIS test based on groups with different disease durations.** Cronbach’s alpha coefficient with item reduction was calculated for the right and left hands separately for each group. Because of the high
Table 1. Clinical characteristics of the patients according to duration of SSc.

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Group 1, 0–3 Yrs, n = 109</th>
<th>Group 2, 3.1–5 Yrs, n = 118</th>
<th>Group 3, 5.1–9 Yrs, n = 143</th>
</tr>
</thead>
<tbody>
<tr>
<td>Female, n (%)</td>
<td>86 (79)</td>
<td>94 (80)</td>
<td>120 (84)</td>
</tr>
<tr>
<td>lcSSc, n (%)</td>
<td>87 (80)</td>
<td>99 (84)</td>
<td>120 (84)</td>
</tr>
<tr>
<td>Ulcers, n (%)</td>
<td>13/109 (12)</td>
<td>12/108 (11)</td>
<td>13/131 (10)</td>
</tr>
<tr>
<td>Pitting scars, n (%)</td>
<td>36/108 (33)</td>
<td>47/110 (43)</td>
<td>52/134 (39)</td>
</tr>
<tr>
<td>Age, median (IQR)</td>
<td>54 (47–65)</td>
<td>55.5 (46–64)</td>
<td>55 (47–64)</td>
</tr>
<tr>
<td>mRSS, median (IQR)</td>
<td>9 (4–16.1), n = 106</td>
<td>6 (3.5–12), n = 113</td>
<td>6 (3–9), n = 133</td>
</tr>
<tr>
<td>Hand skin score, median (IQR)</td>
<td>6 (4–10), n = 104</td>
<td>6.0 (2–8), n = 116</td>
<td>5 (2–7), n = 129</td>
</tr>
<tr>
<td>HAMIS score, median (IQR)</td>
<td>7 (3.5–13.5)</td>
<td>6 (2–12)</td>
<td>6 (3–9)</td>
</tr>
</tbody>
</table>

SSc: systemic sclerosis; lcSSc: limited cutaneous SSc; mRSS: modified Rodnan skin score; HAMIS: hand mobility in scleroderma; IQR: interquartile range.

DISCUSSION

Skin fibrosis is a manifest consequence of SSc and impairs joint motion also in the absence of joint involvement. To evaluate the efficacy of specific therapies, there is a need for instruments that measure fibrotic skin involvement specifically. In our present study, we developed an mHAMIS test having only 4 items that showed the same associations with skin involvement as the original HAMIS test, which has 9 different items. We believe that the mHAMIS test can be valuable in assessing the outcome of therapy aimed at reducing fibrotic skin manifestations. The results regarding construct validity and discrimination indicate that the mHAMIS test fulfills the criteria of the OMERACT filter regarding truth and discrimination. The feasibility is better than that of the original HAMIS test because the equipment needed are only 3 cylinders (5 mm, 15 mm, and 30 mm in diameter), and the time required to perform the test is only a few minutes. The 5 mm cylinder can be an ordinary pencil, and the 15 mm and 30 mm cylinders can be made of doweling or plastic piping used for electricity or water installation.

An incidental finding in our study was that the mobility of the right hand was significantly more impaired than that of the left hand, and we therefore calculated Cronbach’s alpha coefficient separately for the right and left hands. Unfortunately, we did not record which hand was dominant. About 16% of the inhabitants of Sweden are left-handed, and it is reasonable to assume that the proportion was similar in our study. The finding that finger flexion, finger extension, and thumb abduction were more impaired in the right hand in Group 1 could be explained by the higher skin score in the right hand. Further, it is reasonable to assume that the skin involvement in the first stage of SSc results in skin tightness, especially on the fingers, affecting both finger flexion and extension. The mHAMIS test was developed to assess skin involvement as the original HAMIS test, which has 9 different items, but it was shown to be more discriminative and valid. Construct validity of the mHAMIS test

The correlations between the mHAMIS score and the hand skin score and total mRSS were moderate in Groups 1 and 2, and low in Group 3 (Table 3). The correlations between the original HAMIS test and the mHAMIS test ($r_c$) varied between 0.92 and 0.94. The mHAMIS test discriminated significantly between lcSSc and dcSSc in the total study group ($p = 0.001$), and in Group 1 ($p = 0.034$) and Group 2 ($p = 0.003$). The median mHAMIS score in the total study group was 3 (IQR 1–6) for the patients with lcSSc and 7 (IQR 2.5–12) for the patients with dcSSc. The median mHAMIS score in Group 1 was 4 (IQR 2–7) for the patients with lcSSc and 8 (IQR 5–12.5) for the patients with dcSSc. In Group 2, the median score was 3 (IQR 1–7) for the patients with lcSSc and 6 (IQR 1–11) for the patients with dcSSc. Further, the mHAMIS score differed between patients with and without ulcers in the total study group ($p = 0.015$). The median mHAMIS score for patients without ulcers was 3 (IQR 1–7) and for patients with ulcers was 5 (IQR 3–12.8). There was also a difference between patients with and without pitting scars in the total study group; the median mHAMIS score being 5 (IQR 2–8) for patients with pitting scars and 2 (IQR 0–6) for patients without pitting scars ($p = 0.001$).
based on Cronbach’s alpha coefficient, combined with clinical experience showing that skin tightness affects mobility. To make the test more feasible in clinical trials, our aim was also to minimize the amount of equipment required while retaining the items that involve movements related to the skin score measure area. Regarding the items for the wrist,
The Journal of Rheumatology 2014; 41:11; doi:10.3899/jrheum.140286

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Dorsal extension was retained, while supination was excluded because this movement is conducted both in the wrist and elbow, and could be the result of articular manifestations.

The mHAMIS test is shorter and easier to perform than the original HAMIS test, but the 2 are not completely interchangeable. The HAMIS test measures the mobility of the fingers and wrist based on the most common movements assessed in a range-of-motion test. This makes the HAMIS test useful when instructing patients in hand exercises and when monitoring changes in hand mobility. The mHAMIS test consists of a subset of items from the original HAMIS test, and is therefore useful as an instrument for screening hand mobility, but is not as valuable as the HAMIS test in instructing patients in hand exercises.

The moderate correlation between the mHAMIS score and mRSS and hand skin score, and its ability to discriminate between lcSSc and dcSSc in the first 3 years after disease onset (Group 1) demonstrate its usefulness in evaluating skin involvement in the early stages of the disease when information is needed concerning appropriate therapy. However, articular and vascular manifestations may also influence hand function and mobility. The mHAMIS test developed in our study discriminates between individuals with and without ulcers, and therefore we cannot exclude the possibility that vascular manifestations also affect the mHAMIS score. This is in line with studies by Mouthon, et al,25 who found that patients with ulcers had significantly greater impairment of hand mobility, and Del Rosso, et al,7, who reported that patients with SSc with arthritis or ulcers had higher HAMIS scores than did patients without arthritis or ulcers.

Our study had some potential limitations. Articular involvement was not recorded, and we cannot exclude the possibility that arthritis may have influenced the HAMIS score, and therefore also the mHAMIS score. Nevertheless, the convincing correlation between the mHAMIS score and skin involvement indicates that the mHAMIS test is a valid measure of skin involvement. In our study, we recorded the incidence of ulcers and pitting scars because those are features of SSc, and associations have been established between joint contracture and vascular involvement,26 and between vascular involvement and skin fibrosis.27 Our study was based on data collected during routine clinical care and includes a rather small number of patients with dcSSc.

Table 2. Cronbach’s alpha coefficient for the original HAMIS test and the 4 most representative items in the right and left hands in each group and in the dcSSc subgroups.

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Group 1, 0–3 Yrs</th>
<th>Group 2, 3.1–5 Yrs</th>
<th>Group 3, 5.1–9 Yrs</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cronbach’s alpha</td>
<td>Right hand</td>
<td>Left hand</td>
<td>Right hand</td>
</tr>
<tr>
<td>Item-reduced HAMIS</td>
<td>Finger flexion, finger extension, thumb abduction, finger abduction</td>
<td>0.62</td>
<td>0.61</td>
</tr>
<tr>
<td>Cronbach’s alpha</td>
<td>Right hand</td>
<td>Left hand</td>
<td>Right hand</td>
</tr>
<tr>
<td>Finger flexion, finger extension, pinch, finger abduction</td>
<td>0.882</td>
<td>0.878</td>
<td>0.773</td>
</tr>
<tr>
<td>Finger flexion, finger extension, pinch</td>
<td>0.914</td>
<td>0.934</td>
<td>0.843</td>
</tr>
</tbody>
</table>

HAMIS: hand mobility in scleroderma; dcSSc: diffuse cutaneous SSc; lcSSc: limited cutaneous SSc; SSc: systemic sclerosis.

Table 3. Correlations between the original HAMIS test, the mHAMIS test, and skin score in groups with different disease duration. All values are p = 0.001.

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Total Study</th>
<th>Group 1, 0–3 Yrs</th>
<th>Group 2, 3.1–5 Yrs</th>
<th>Group 3, 5.1–9 Yrs</th>
</tr>
</thead>
<tbody>
<tr>
<td>mRSS</td>
<td>0.42</td>
<td>0.39</td>
<td>0.60</td>
<td>0.59</td>
</tr>
<tr>
<td>Hand skin score</td>
<td>0.45</td>
<td>0.43</td>
<td>0.62</td>
<td>0.61</td>
</tr>
<tr>
<td>Skin score, fingers</td>
<td>0.43</td>
<td>0.43</td>
<td>0.48</td>
<td>0.48</td>
</tr>
<tr>
<td>Skin score, hand</td>
<td>0.35</td>
<td>0.31</td>
<td>0.59</td>
<td>0.59</td>
</tr>
<tr>
<td>Skin score, forearm</td>
<td>0.33</td>
<td>0.30</td>
<td>0.52</td>
<td>0.52</td>
</tr>
</tbody>
</table>

HAMIS: hand mobility in scleroderma; mHAMIS: modified hand mobility in scleroderma; mRSS: modified Rodnan skin score.
Therefore, Cronbach’s alpha coefficient was also calculated separately for patients with dcSSc.

To obtain as complete assessment of hand function and the efficacy of specific therapies as possible, attempts should be made to include measurements from the different domains of the ICF, specifically the scoring of hand function, as well as self-reported questionnaires that assess activity limitations. Although we have previously demonstrated a significant relationship between the original HAMIS and CHFS scores, these tests are not interchangeable, but rather complement each other. The mHAMIS test provides a measure of hand mobility assessed by health professionals, while the CHFS measures the patient’s perception of their ability to perform activities in daily living, which can be influenced by function and by other factors. Both measures are needed to evaluate disease severity and the efficacy of drugs and rehabilitation.

The hands account for only a small proportion of the total area of the body, but are important in many functions in daily life. Because of the correlation between the mHAMIS score and the mRSS score, the mHAMIS score may be a relevant outcome measure in evaluating the fibrotic process in the skin of patients with SSc. However, interventional studies are needed to further investigate the responsiveness of the mHAMIS test.

REFERENCES

**APPENDIX 1. Modified hand mobility in scleroderma (mHAMIS).**

The test equipment consists of standardised cylinders for assessment of finger flexion (5, 15, 30 mm diameter) and finger extension (5, 15 mm diameter).

**Finger flexion**
- All fingers must be tight to the object.
- Can bend fingers 2-5 around a pencil (5 mm diam). 0
- Can bend fingers 2-5 around a piece of cutlery (15 mm diam). 1
- Can bend fingers 2-5 around handlebar (30 mm diam). 2
- Cannot manage the previous item. 3

**Finger extension**
- All fingers must be tight to the object.
- Can feel the table completely with digit 2-5. 0
- Can feel the pencil (5 mm diam) with digit 2-5. 1
- Can feel the piece of cutlery (15 mm diam) with digit 2-5. 2
- Cannot manage the previous item. 3

**Finger abduction**
- Can spread the fingers and then fold the hands together to the bottom of the fingers. 0
- Can spread the fingers and then fold the hands together to the first phalange. 1
- Can spread the fingers and then fold the hands together to the second phalange. 2
- Cannot manage the previous item. 3

**Dorsal extension**
- Can hold the palms together and put the wrists against the stomach. 0
- Can hold the palms together and put the thumbs against the throat. 1
- Can hold the palms together and put the thumbs up to the mouth. 2
- Cannot manage the previous item. 3