Evaluating the cutaneous involvement in scleroderma: torsional stiffness revisited

L. R. Knight, J. E. Smeathers¹, A. H. Isdale and P. S. Helliwell

Rheumatology and Rehabilitation Research Unit, University of Leeds, Leeds, UK and ¹School of Human Movement Studies, Queensland University of Technology, Brisbane, Australia

Abstract

Objective. The pace, progression and extent of the skin lesions in scleroderma may parallel the risk of new internal organ involvement and the progression of existing internal lesions. Accurate assessment of cutaneous change permits an evaluation of patient prognosis and the response to therapy. The aim of this study was to assess a simple device for measuring skin stiffness for its ability to measure sclerodermatous skin in a quantitative and reproducible manner.

Materials and methods. Torsional skin stiffness was measured in 36 normal subjects and 42 scleroderma patients (31 of whom had the limited form and nine the diffuse form, and two had mixed connective tissue disease). Data for the scleroderma patients were compared with data obtained by the use of the modified Rodnan clinical skin scoring technique. Intraclass correlation coefficients (ICCs) were calculated as a measure of intraobserver and interobserver variability.

Results. For the left and right hands respectively, the ICCs for intraobserver variability were 0.908 and 0.906 and those for interobserver variability were 0.871 and 0.628. There was a significant difference in mean angular rotation obtained by normal subjects compared with scleroderma patients (15.1 vs 11.3°, P < 0.001). There was a significant difference in the angular rotation with increasing severity of skin involvement (skin score 0, median rotation 16.3°; score 1, 10.5°; score 2, 8.5°; score 3, 8.0°; P < 0.00001).

Conclusions. The measurements obtained with the skin stiffness device are highly reproducible and are consistent with the current clinical method of assessment of skin involvement. The significant difference in angular rotation obtained by normal subjects and scleroderma patients indicates that the device can distinguish normal from sclerodermatous skin. The torsional stiffness measurements derived from the device may also be useful in longitudinal studies.

Key words: Torsional stiffness, Skin, Scleroderma.

Scleroderma is a multisystem connective tissue disorder causing fibrosis of the visceral organs and skin. In sclerodermatous skin there is excessive accumulation of extracellular matrix in the dermis, predominantly of collagen types I and III, leading to clinically taut skin. There are also associated deposits of connective tissue components, (glycosaminoglycans, fibronectin and tenasin) [1]. Tethering (the fixation of the epidermis and dermis to underlying structures such as the subcutaneous fat) prevents free movement of the skin at the surface. Together, these changes result in increased stiffness in sclerodermatous skin.

Biomechanically, the skin is a non-linear, non-homogeneous, viscoelastic material consisting of two main layers. The epidermis is the external layer and is made up of stratified epithelium. The dermis provides the skin with its elastic and mechanical properties [2]. When an extension load is suddenly applied to the skin, there is an initial and sudden local increase in skin length as the collagen fibres straighten and orientate, an effect dependent on the direction of the applied force due to anisotropy of the skin. Tethering of the skin to the underlying structures, as occurs in scleroderma, is likely to reduce this initial displacement. The initial change is followed, if the force is maintained, by further time-dependent extension due to creep, the extent and rate of which depend on the viscoelastic properties of the dermis. In scleroderma a reduction in the rate of creep deformation would be expected.

Clinically, cutaneous lesions may range from extensive skin involvement to mild sclerodactyly. Clinical observation of changes in the skin allows ready appreciation of the course of the skin disease. Additionally, the importance of the skin lesions lies in the understanding that the progression and extent of the lesions may parallel the risk of occurrence of new internal organ...
involvement and of the spreading of existing disease, particularly in the early years of diffuse scleroderma [3]. By accurately assessing skin involvement, not only can the patient’s prognosis be evaluated but also the patient’s response to therapy can be assessed [4, 5]. Additionally, one of the most compelling requirements for the competent care of scleroderma patients is that of relevant and reproducible measurement of disease status [4, 6].

So far, a variety of techniques have been employed to provide an objective measure of the cutaneous changes. Such techniques have included radiography, ultrasound, skin biopsy weight, palmprint areas and the measurement of cutaneous hypoxia [5]. Most of these techniques have not gained widespread acceptance. A clinical method of scoring sclerodermatous skin based on palpation has been favoured by most clinics. However, its role in the monitoring of sclerodermatous skin remains controversial since it is semiquantitative and subject to observer bias [4–6].

A number of mechanical methods have been used to measure the properties of skin, mostly as a research tool or commercially to assess the effects of emollients. Such methods include extensometry, which measures the force required to extend the skin; indentation and suction techniques; pliometry; and torsional tests, in which a torsional force is applied to the skin surface with measurement of the resulting deformation [7]. This project aimed to evaluate a simpler version of a device for measuring torsional stiffness for use in scleroderma so as to allow patient monitoring and provide a potential outcome measure for clinical trials of new therapies.

Materials and methods

Skin stiffness device

The device was designed as a lightweight, portable, easy-to-assemble apparatus requiring negligible maintenance. Further requirements were that the procedure should be quick and simple to perform and should readily provide data on torsional stiffness. The principle of the device is the application of a torsional force (5 mNm) via a shaft applied directly to the surface of the skin, using an adhesive pad (Fig. 1). The resulting displacement of the skin is a function of the torsional stiffness of the skin at that point. Application of a torsional force overcomes the problem of anisotropy noted above. Typical curves for angular rotation against time are shown in Fig. 2. The 15-s cut-off was chosen as a practical and clinically relevant end-point at which the curve begins to flatten, although creep deformation is clearly still occurring at this point. The effect of shearing at the interface between shaft and skin was negligible, as were the effects of skin temperature, diurnal variation and hand position. From start to finish the whole procedure was usually completed in 5 min.

The reproducibility of the results given by the skin stiffness device was evaluated from intraobserver variability (nine normal subjects were measured on three separate occasions by the same observer using the same protocol) and interobserver variability (11 normal subjects were measured by two examiners within 7 days) using the intraclass correlation coefficient (ICC).

Subjects

The device was used to provide normal data on skin stiffness for 56 subjects, mostly members of the University of Leeds. None of the normal subjects had a skin condition, and people with diabetes and rheumatological disorders were excluded. Forty-two patients with scleroderma were measured. All the patients fulfilled the American Rheumatism Association preliminary clinical criteria for systemic scleroderma [8].

![The skin stiffness device](image-url)
Modified Rodnan scores were obtained by clinical palpation of 17 anatomical areas: the face, anterior chest, abdomen and, right and left separately, upper arms, forearms, hands, fingers, thighs, legs and feet. The rating scale consisted of 0 = normal; 1 = thickened; 2 = thickened, unable to pinch; 3 = thickened, unable to move.

Results

Reproducibility of the device
Acceptable intraobserver variability was found: the ICC for the left hand was 0.908 and that for the right hand was 0.906. For the interobserver variability, the ICCs were 0.871 and 0.628 for the left and right hands respectively.

Skin stiffness of normal subjects and scleroderma patients
The ages of the normal subjects (33 females and 23 males) ranged from 22 to 82 yr, and those of the patients (36 females and six males) ranged from 30 to 82 yr. Nine patients had diffuse systemic scleroderma, 31 had the limited form and two had mixed connective tissue disease. The median disease duration for these 42 patients was 4 yr. Lung, cardiovascular and renal involvement was found in 41, 30 and 3%, respectively.

Age (20–30, 31–40, 41–50, 51–60 and > 60 yr), gender and body mass index (two cohorts with a cut-off of 25) had no significant effect on the mean angular rotations obtained for either the normal subjects or the scleroderma patients. Furthermore, since there was no significant difference in angular rotation obtained for the left and right hands, mean angular rotation was used.

Summary statistics for mean angular rotation for the normal subjects and the patients with scleroderma are shown in Fig. 3. The median value for the angular rotation in the normal subjects was 15.1° (95% confidence interval 12–17°) and that for the scleroderma patients was 11.3° (8.5–12.3°) (Mann–Whitney U-test, Z = −4.5, P < 0.001).

Relationship between angular rotation and clinical skin scoring technique
For all scleroderma patients, the median modified Rodnan skin score was 9 (95% confidence interval 7–12). Comparing the limited form of systemic sclerosis with the diffuse form, the median score for patients with the limited type was 8 (6–11) while the median skin score for patients with the diffuse form was 18 (9–24). Figure 4 provides summary statistics for the relationship between angular rotation and skin score. Significant
differences between the four grades of skin score were found: skin score 0, median angular rotation 16.3°; score 1, 10.5°; score 2, 8.5°; score 3, 8.0° (Kruskal–Wallis test, $\chi^2 = 26.0$, $P < 0.00001$).

**Discussion**

The skin stiffness device is simple to use, quick and reproducible. The measurements appear to be valid in that the device can discriminate between normal and diseased skin. Furthermore, there is an inverse relationship between angular rotation and local clinical skin score, the current standard. It may be postulated, therefore, that the data obtained from the skin stiffness device may be used as an outcome measure of skin involvement in scleroderma. However, further validation work is required. It would be valuable to be able to obtain concurrent histological and biochemical data from skin biopsy. Furthermore, the ability of the device to measure changes in response to therapeutic interventions and the measurement of biomechanical properties longitudinally (in parallel with cutaneous and internal organ involvement) are desirable.

Previous attempts to measure the torsional stiffness of the skin have used a slightly different technique. The method, originally described by de Rigal and Leveque [9], imparted a torsional load of between 4 and 57 mNm to the skin via an in-line motor, with continuous measurement of rotational displacement. The main differences between the de Rigal technique and the device used in the present work are (i) that the torque is now applied via a weight which is allowed to drop freely, and (ii) the relatively crude level of measurement with the present method. However, this lack of sophistication is compensated for by the simplicity, portability and lack of dependence on an electrical power source. The other important difference between the devices lies in the way the rotational displacement is expressed. With the present device, the total angular displacement at 15 s is recorded, but this figure conceals a number of different phases in the torque/displacement curve (Fig. 2). In contrast, de Rigal and colleagues chose to record only the initial instantaneous displacement [9]. Nevertheless, measuring sclerodermatous skin, the de Rigal group found results similar to those of the present study, with decreases in rotational displacement in diseased skin compared with normal subjects [10]. In fact, the ability of the device to record the free movement of skin over the underlying tissues greatly facilitates the distinction between normal and sclerodermatous skin as it probably reflects the effect of tethering.

There is generalized thinning and elastic degeneration of skin with increasing age [11]. It has been demonstrated that there is a progressive rise in the modulus of elasticity of skin with increasing age [12], and this stiffening process is attributed to a decrease in the total amount of soluble collagen together with an increase in the number of stable crosslinks and the proportion of insoluble collagen [13]. It has also been shown that females have a higher modulus of elasticity compared with males [12], but in more recent studies there was no significant difference between the sexes [14]. In this limited study, we found no relationship between skin measurements and age or sex and, this discrepancy may be a result of the difference in techniques. Using the current device, the greatest proportion of the angular rotation is obtained immediately after release of the weight: it is likely that this deformation reflects the gross realignment of the collagen fibres in skin that is relatively free to move over the underlying tissues. The viscoelastic properties of skin are reflected by the deformation occurring immediately after realignment; any changes due to ageing may therefore be masked by the initial deformation. Using their rotational device, Leveque et al. [15] found a U-shaped relationship between initial deformation and age, but were able to convert this to a negative correlation by normalizing the measurements for skin thickness.

Currently, skin involvement in scleroderma is assessed by the semiquantitative modified Rodnan skin scoring method derived from clinical palpation of 17 anatomical areas. In comparing results obtained from the skin stiffness device with local skin scores, an appreciable decline in angular rotation was seen with increasing severity of skin involvement. Both clinical palpation and the skin stiffness device can distinguish an increase in skin stiffness. However, clinical palpation can only detect skin changes numerically with scores of 0, 1, 2 or 3; these scores may not be equidistant or absolute, in that score 2 for one patient may not be equivalent to score 2 for another. Furthermore, observers do not always agree on the score [4–6]. The new skin stiffness device is advantageous over clinical palpation since it has good inter- and intraobserver reproducibility and does not restrict the observer to a relatively coarse four-point scale. The advantage for intervention studies is obvious: having a continuous outcome measure with an interval level scale can increase the power of an intervention study to detect significant differences.

In summary, the results given by the skin stiffness device are highly reproducible and correlate well with results obtained with the current method of skin assessment. The presence of scleroderma has a significant effect on torsional stiffness data. Although the degree of variation between individuals may be great, studies beyond this preliminary investigation should probably be directed at investigating intra-individual variation longitudinally and the effects of treatment. The skin stiffness device could be adapted for use at sites other than the dorsum of the hands.

**Acknowledgements**

We wish to thank Dr R. Melsom and Dr M. Goodfield for permission to approach their patients. This work formed part of the requirements for an intercalated BSc in clinical sciences and was funded by an Arthritis Research Campaign Studentship (number E0525).
References