A Multi-Centre Qualitative Study Exploring the Patient Experience of Digital Ulcers in Systemic Sclerosis

Original article

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Short title: The patient experience of SSc-DUs

Key Indexing Terms: Systemic Sclerosis, Scleroderma, Digital Ulcers, Qualitative Research, Patient Participation, Patient Reported Outcomes

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Sources of support: Scleroderma Clinical Trials Consortium.

Word Count: 3851

Abstract

Objectives:

Digital ulcers (DUs) are a major cause of disease-related morbidity and difficult to treat vascular complication of systemic sclerosis (SSc). Demonstrating treatment efficacy has traditionally focussed upon clinician assessment of DUs alone. No existing patient reported outcome (PRO) instrument captures the multi-faceted impact of SSc-DU. We report the findings of a multi-centre qualitative research study exploring the patient experience of SSc-DU.

Methods:

Patient focus groups (FGs) were conducted across 3 scleroderma units, following a topic guide devised by SSc patients, experts and experienced qualitative researchers. A purposive sampling framework ensured the experiences of a diverse group of patients were captured. FGs were audio recorded, transcribed, anonymised, and analysed using inductive thematic analysis. We continued FGs until thematic saturation was achieved.

Results:

Twenty-nine SSc patients with a history of DU disease participated in 4 FGs across the UK (Bath, Manchester and London). Five major inter-related themes (and sub-themes) were identified which encompass the patient experience of SSc-DUs: 'Disabling pain and hypersensitivity', 'Deep and broad-ranging emotional impact', 'Impairment of physical and social activity', 'Factors aggravating occurrence, duration and impact' and 'Mitigating, managing and adapting'.

Conclusion:

The patient experience of SSc-DU is multi-faceted and comprises a complex interplay of experiences associated with significant pain and morbidity. Patient experiences of SSc-DU are not captured using existing SSc-DU outcomes. Our findings shall inform the development of a novel PRO instrument to assess the severity and impact of SSc-DUs for use in future SSc-DU clinical trials.

Significance and Innovations:

- Existing SSc-DU outcome measures do not capture the complete patient experience of SSc-DU.
- The patient experience of SSc-DU is comprised of interrelated factors which contribute to the significant morbidity of SSc-DUs.
- Five major inter-related themes were identified: ‘Disabling pain and hypersensitivity’, ‘Deep and broad-ranging emotional impact’, ‘Impairment of physical and social activity’, ‘Factors aggravating occurrence, duration and impact’ and ‘Mitigating, managing and adapting’.
- The interplay between the themes suggest that the presence of SSc-DUs can have a considerable impact on patients’ physical and psychological wellbeing, impairing physical and social activities, and that patients expend great effort in remaining vigilant and managing their condition, often in innovative ways.
- Our findings can be used to inform the development of a novel patient reported outcome instrument to assess the severity and impact of SSc-DU.

Introduction

Digital ulcers (DUs) are a major cause of pain and disability in people living with systemic sclerosis (SSc) (1). DUs are common with around half of patients reporting a history of ulceration and 5-10% of people with SSc at any time have a current ulcer (2,3). DUs have a major impact on quality of life and hand function including occupation (4). Although we have a number of treatments (5) available to both prevent and heal SSc-DUs (e.g. phosphodiesterase type-5 inhibitors and intravenous prostanoids) (6–8); despite such interventions, a third of patients with SSc are affected by refractory DU disease (9).

In general, demonstrating treatment efficacy in previous clinical trials has been based upon clinician assessment of ulcer healing and/or new ulcer occurrence alone (1). However, the agreement amongst clinicians with an interest in SSc to classify SSc-DU is poor to moderate at best (10–12). Inter-rater agreement is not improved with the provision of clinical ('real world') contextual information (e.g. the severity of pain and duration of the lesion) (11). Recent negative clinical trials of promising therapies for SSc-DUs (13,14) have led to calls for a fresh approach to establishing treatment efficacy in SSc-DU (15–17).

No studies have specifically explored the patient experience of SSc-DU, although studies examining broader symptom burden in scleroderma have identified the major impact that SSc-DU can have for patients as the following quotation attests:

"..the pain that you felt in your fingers as they were dying was so excruciating that you almost begged to say please cut it off" (reproduced from[18])

Previous attempts to quantify the impact of SSc-DUs have used legacy patient reported outcome (PRO) instruments to assess broader aspects of SSc disease severity and function (19). There was limited or no SSc patient participation in the development of many of these instruments (20). The patient perspective captured by PRO instruments provides insight into the patient experience of disease that can not be assessed using clinician-reported instruments (21). Regulatory bodies, such as the FDA, seek target patient population involvement in PRO instrument development to ensure instruments fully capture the way patients 'feel' and 'function' (22). A thorough understanding of the patient experience of SSc-

DU is necessary to ensure a future PRO instrument captures the multi-faceted impact of SSc-DU.

Against this background, the aim of this present study was to comprehensively explore the experiences, attitudes and perspectives of patients with SSc-DUs. A further aim was to inform the development of a future SSc-DU PRO instrument.

Patients and methods

Study management

The development and conduct of the study were overseen by a dedicated steering committee which comprised of experts in SSc (MH, JDP, CPD, RTD, TMF, ALH, DK, MMC and LAS), 2 patient research partners (LM and JW), and a team of experienced qualitative methodologists (YA, RGH and AM). The study was approved by the East Midlands – Nottingham 1 research Ethics Committee (REC reference – 18/EM/0018) and all participants provided written, informed consent.

Study design

A multi-centre qualitative research study comprising patient focus groups (FGs) was undertaken at scleroderma centres across the United Kingdom (Bath, Manchester and London). Patient FGs create an open environment in which a broad range of experiences can be expressed and explored; and can often enable some (but not necessarily all) sensitive issues to be discussed more freely than in a one-to-one interview setting (23).

Participants

Adult SSc patients (>18 years) with a history of SSc-DU, fulfilling the 2013 American College of Rheumatology/European League Against Rheumatism classification criteria for SSc (24) with sufficient language skills to participate in a FG discussion were enrolled at each site. A purposive sampling framework ensured the enrolment of a diverse cohort comprising a 60:40 split between limited and diffuse cutaneous SSc (25), early and established disease (≤3 and >3 years since first non-Raynaud's phenomenon symptom, respectively), a spectrum of history of DU disease, sex (aiming for 5:1 female predominance) and ethnicity (e.g. with Caucasian and Black British). The FGs sought to include 6-10 participants to enable open discussion whilst ensuring that each participant had the opportunity to express their personal experiences, interact and offer alternative opinions should they wish. A minimum of two to three FGs was expected to be necessary in order to achieve 'thematic saturation' but the intention was to continue enrolment until there was consensus that no meaningful new experiences were being shared by participants or warranted further exploration by the investigators (26).

Data collection

Each FG lasted approximately one hour in duration and were all facilitated by MH, with the first focus group also facilitated by JP and AM to ensure that there were no issues including a need to revise the topic guide (which was not the case). FGs were facilitated by rheumatologists (MH and JP) with experience in the clinical heterogeneity and management of patients with SSc. FGs were held within hospitals but outside of clinical areas in a quiet ambient environment without external distraction. The FG lead facilitator (MH) is rheumatologist with an interest in SSc and was not directly involved in the clinical care of the participants. JP is a rheumatologist with an interest in SSc and AM is an experienced qualitative researcher/methodologist. A relaxed environment in which each participant's views were sought, valued and respected enabled individuals to share experiences of SSc-DU and allow others to express similar or opposing views. The FGs were audio-recorded and subsequently transcribed verbatim, with all the context anonymised. A topic guide was developed with input from the study steering committee (see supplementary material). Each FG started with broad open questions asking participants to describe their experience of their disease and DU history. FGs adopt an adaptive study design enabling incompletely explored or newly emerging themes to be investigated to ensure thematic saturation was achieved.

Data analysis

Qualitative analysis was conducted by JJ and AM, both experienced qualitative methodologists, with further input from the wider team (MH, JP and RGH and patient partners).

NVivo 11 software was used to manage and interrogate the data. Transcribed data were analysed using thematic analysis (26). First, JJ read and re-read transcripts to ensure familiarity with the content. Information relevant to patients' experience and understanding of digital ulcers was then coded using descriptive labels. Codes that occurred repeatedly, or that shared conceptual similarities were then grouped together to form initial categories. The initial set of codes and categories were then discussed with the wider team (AM, MH, JP, RGH) to ensure it captured all elements from the focus group. The coding framework was then applied to subsequent transcripts and any newly identified codes added as appropriate. The FG facilitators decided when data saturation had been reached (27). Codes were collated and grouped into themes and sub-themes. Coded data within each theme was checked to ensure internal coherence (fit within the pattern of the theme) and external representativeness (fit within the whole data set). JJ and AM regularly discussed the conceptual development of the themes and subthemes and an analysis de-briefing meeting was convened involving (JJ, AM, RGH & JP) to discuss the final theme groupings and the conceptual map describing the interrelationship of the respective themes.

Our approach was both deductive, in the sense that the research team examined pre-conceived considerations on the impact of DUs (derived from an earlier comprehensive literature review (19)) and how participants understood and managed them, for the purposes of developing a PRO instrument, and inductive in the sense there was no pre-existing coding frame and the developing codes were derived from and grounded in the data themselves (28).

Results

Twenty-nine patients with SSc participated in 4 FGs conducted in Bath (n=8), Manchester (n=7) and two FGs in London (n=6 & 8). Our *a priori* purposive sampling framework ensured that we studied a broad study population of patients with SSc and DU disease (Table 1). Thematic saturation was felt to have been achieved after 4 FGs.

Five major themes emerged, that together constitute the patient experience of SSc-DUs: (i) disabling pain and hypersensitivity, (ii) deep and broad-ranging emotional impact, (iii) impairment of physical and social activity, (iv) factors aggravating occurrence, duration and impact, and (v) mitigating, managing and adapting to ulcers. The 5 constituent themes (and subthemes) can be arranged within a conceptual map of the patient experience of SSc-DUs (Figure 1).

Theme 1: Disabling pain and hypersensitivity (Table 2)

Our study found that pain is a cardinal symptom of SSc-DUs and is often very severe (Q1-4). Participants used a wide range of words and phrases to describe the severity of pain such as: 'excruciating', 'pain that could reduce you to tears', 'agonising' and 'unbearable'. Participants often described the pain as pulsatile or throbbing in nature (Q5-6), including a pressure like effect (Q7). Not all participants used the word 'pain' to describe the physical discomfort of SSc-DUs; other expressions included 'soreness', 'tenderness' or 'discomfort'. The level of reported pain was often considered as being disproportionate to the size of the DU (Q8). DU pain can radiate to the other digits and proximally (Q9-10). Co-existent infection of the ulcer increases DU pain (Q11) and some participants reported that changes in temperature can worsen DU pain (Q12-13). Many participants described pain in the areas where previous ulcers had occurred, whereas, others said the area was tender, sore or sensitive, and could be aggravated by touch or exposure to cold (Q13-15). Other sensations in areas of previous ulcers included tingling nerve-like sensations and partial or complete numbness (Q16-17). One participant said, "it's never the same again" (P6 M1) when talking about the area where previous ulceration had occurred. Due to the severity of DU pain, some participants suggested that invasive procedures (including digital amputation) may be both necessary and appropriate to relieve symptoms (Q1, Q4, Q5, Q7, Q18, Q24). Across all the FGs, participants talked about the need to validate the pain they experienced with friends, family and colleagues (Q19-22). Participants described the severity of their ulcers in different ways. These included the need for hospitalisation, the time to heal, changes in their life (e.g. giving up work or hobbies) due to ulcers, and previous/risk of amputation (Q23-24). There was a wide variety in the reported location (fingertips, over the small joints, under the nails and on the sides of the fingers) of DUs amongst participants. Some experienced ulcers in different locations on the hands, whereas, others tended to only get ulcers in one area.

Theme 2: Deep and broad-ranging emotional impact (Table 3)

Related to the severity of pain, most participants shared a constant fear of the development of new DUs (Q25) and many considered it inevitable that further lesions would develop (Q26-

27). Participants experienced anxiety/uncertainty around how severe each ulcer would be, whether they were treating the ulcer correctly, and how long it would take the ulcer to heal (Q28). Although most participants did not explicitly say that ulcers caused them depression (Q29), they mentioned many associated emotions (in addition to anxiety and embarrassment) including uncertainty/fear for the future and anger (Q29-32). Participants described the need for a constant level of vigilance to prevent the development of new DUs and infection of intercurrent ulcers (Q33-34). Participants described many different emotions associated with the ulcers from panic, anxiety, fear and irritability to anger (Q35). Participants did not forget about the past impact of the ulcers and some described frightening times (with current ulcers) when they were perhaps unsure whether they would need to have part of their finger amputated (Q24). Patients also experienced embarrassment and distress due to the physical appearance of SSc-DUs and took a range of actions to hide DUs from others (Q36-40).

Theme 3: Impairment of physical and social activity (Table 4)

The physical and psychological impact of SSc-DUs was closely related to impact on physical and social functioning. Patients interactions with the world and other people were characterised by an avoidance of pain, and a constant vigilance during physical and social interaction. Participants reported about how DUs impacted on their ability to use their hands during activities of daily living (Q41-46), including self-care/grooming (Q38, Q47-48), hobbies (Q49) and domestic activities (e.g. cooking and household chores) (Q13, Q50-51). Taken for granted activities of daily living became foregrounded, such as their ability to reach their hands into pockets, a bag or a purse (Q45, Q52-53), difficulty driving (Q30, Q54), sleeping (Q55) and challenges when shopping (Q12, Q56-57). Impact of DUs on work varied between the participants. For some participants, ulcers had not severely impacted on their work, whereas, others had to change roles in the organisation or even change jobs completely (58-59). Some participants described financial concerns from the impact of DUs on their work (Q60). DUs impact on social participation and participants reported taking measures to conceal ulcers with bandages or gloves to both avoid others seeing them and to reduce the risk of infection (Q61-62). A number of participants described difficulties undertaking caring roles within the family; for example, avoiding taking their children outside to play due to the cold weather (Q63) as the cold both exacerbated the pain, but could also aggravate the healing of ulcers, or provoke their onset.

Theme 4: Factors aggravating occurrence, duration and impact (Table 5)

There were a number of factors that aggravated the occurrence and duration and impact of ulcers. There was variation in the number of ulcers experienced by participants; ranging from experiences of solitary DU to recurrent episodes of refractory digital ulceration (Q24, Q26, Q64-66). There was variation amongst participants on the time to DU healing (weeks, months or even years). The length of time to heal was often related to the season and treatment. Most participants reported that over the winter it took longer for ulcers to heal (Q67), or they did not heal at all until the summertime (Q68). Most participants seemed to be able to identify where previous ulcers had occurred either based on how they looked or how they felt or both (Q13-16).

Theme 5: Mitigating, managing and adapting to ulcers (Table 5)

Participants used a variety of ways to describe whether a treatment had been effective or not. This included: whether and how quickly the ulcer had healed; if there had been a reduced rate of recurrence of the ulcer; how the appearance of the ulcer had changed; whether the level of pain was reduced; positive impact on other activities such as sleeping and whether the participant thought circulation had improved; if the wound dressing had been effective in protecting the ulcer, and whether the risk of amputation was reduced (Q69-73). As well as the effectiveness of treatment, participants also alluded to the burden of treatment. This could mean the need for hospitalisation or the burden of medication, the duration (time) of receiving treatments or the severity of associated side effects, and the time and ease of putting on bandages (Q74-75). Participants discussed a range of coping strategies to manage DUs including different ways in which they had adapted or used support in order to cope with their ulcers. This included using a device or aid to help manage ulcers (Q76-77), strategies to avoid causing pain or preventing a new ulcer developing (Q78-79) and getting help or support (paid or unpaid) from others (Q80). Several participants talked about how their children have adapted to the condition and help them cope with limited function (Q80). However, some noted that it was not possible to avoid all activities which may aggravate the ulcer especially if they have young children (Q81). Participants described a variety of techniques they used to manage their ulcers, from the earliest stages of development to when the ulcer is visible and active. These included using ‘home remedies’ and alternative treatments (Q82), wound care

(Q83), the vigilance associated with self-management (Q84-Q85) and avoiding behaviours (e.g. cold exposure) that they consider can cause ulcers (Q86).

Discussion

The present study is the first to specifically explore the multi-faceted patient experience of SSc-DU. We have identified 5 major inter-related themes (and subthemes) which constitute the patient experience SSc-DUs that we have organised within a conceptual map of SSc-DU. The major themes comprised 'Disabling pain and hypersensitivity', 'Deep and broad-ranging emotional impact', 'Impairment of physical and social activity', 'Factors aggravating occurrence, duration and impact' and 'Mitigating, managing and adapting to SSc-DUs'.

The multi-centre study design and purposive sampling framework ensured we captured the experiences from a broad cohort of SSc patients and the whole spectrum of SSc-DU disease (from solitary DUs to recurrent refractory disease). Thematic analysis of the FG transcripts was conducted by experienced qualitative researchers without direct experience in the management of SSc-DUs, avoiding the potential bias that pre-conceptions held by scleroderma clinicians might have introduced. The study benefited from a broad international steering committee of experts in SSc, qualitative researchers and patient research partners.

Painful physical symptoms and signs were the most important experiences of SSc-DU. Pain is the cardinal symptom of SSc-DUs and is often very severe. Patients often consider the severity of pain disproportionate to the physical size of DUs. Infection and changes in temperature can worsen DU pain. The physical symptoms of DUs results in considerable psychological distress, and impaired hand function impacts on all the activities of daily living including occupation and social interactions. Many patients describe a constant state of vigilance both during and between episodes of ulceration. There are a number of aggravating factors including the number and severity of DUs. Of interest, participants reported that the ulcers took longer to heal during the winter, and residual symptoms at sites of previous DUs. In particular, dysesthesias and paraesthesias could suggest potential persistent nerve damage from tissue ulceration. Patients with SSc make considerable efforts to both prevent and manage DUs (e.g. avoiding trauma and preventing infection) and describe a wide range of coping strategies and adaptations. This mirrors the patient experience of SSc-Raynaud's phenomenon, in which patients report the need for constant vigilance & self-management (29). Overall, our themes show similarities to those reported by Nakayama et al who conducted a systematic review and thematic analysis of 26 studies with 463 patients' to explore patients perspectives and experiences living with SSc (29). The 6 key themes were: 'distressing appearance transformation', 'palpable physical limitations', 'social impairment', 'navigating uncertainty', 'alone and understood', and 'gradual acceptance and relative optimism' (29). Furthermore, DUs (along with Raynaud's phenomenon and calcinosis) were described as 'being intensely painful by some patients', was 'emotionally distressing', and 'limited patients' ability to work, go outdoors', 'or even walk' (29).

As previously described, previous clinical trials of SSc-DUs, primary assessment of treatment efficacy has focussed on clinician assessment of DU presence alone (occurrence and persistence) and have largely overlooked the patient experience of SSc-DUs. Legacy PRO instruments assessing function and interference capture patient experiences relevant to SSc-DUs, but are limited by the inclusion of redundant items which are less relevant to SSc-DUs (e.g. the inclusion of non-hand domains of the HAQ-DI). The recent development of a SSc-specific PRO instrument: the Hand Disability in SSc DUs (HDISS-DU) was developed through modification of the Cochin Hand Function Scale including qualitative patient interviews to assess the impact of DUs on hand function in patients with SSc (30). However, to date, other important experiences of SSc-DUs (e.g. psychological impacts and social participation) have been comparatively overlooked. The development of a novel PRO instrument that captures the broader patient experience of SSc-DU (e.g. pain, social participation, relationships, body image dissatisfaction etc.) would be valuable for assessing interventions in the clinical trial setting, but also in routine clinical practice, where there is a dearth of practice-based evidence examining the comparative efficacy of different pharmacological, surgical and wound care protocols. Furthermore, even after ulcer healing, patients can still suffer from significant residual pain and anxiety of future DUs. Therefore, effective ulcer treatments (and PRO instruments) should also modify future patient (negative) experiences of DU disease even after ulcer healing.

Our analysis has not addressed potential differences in experiences relating to DUs occurring at different locations on the hands (e.g. fingertip vs extensor). The aetiopathogenesis (and patient experience) of different types of DU may differ, although it is generally accepted that all types of DU have an ischaemic contribution (31,32). Therefore, future efforts to develop a dedicated PRO instrument for assessing SSc-DU should explore different experiences (including treatment effects) at different ulcer locations. We also highlight that we only recruited a relatively small number of patients with ‘early’ disease. This is likely due to the need in our study to include a large majority of patients with a significant burden (history) of digital vascular disease, which usually takes time (years) to accrue. There were differences observed in the clinical and demographic characteristics (e.g. age and gender) of participants who participated in the four FGs. For example, the majority of patients in Bath and Manchester had the limited subset of the disease, whereas, approximately equal numbers of patients had diffuse disease in the two London FGs. We did not entirely achieve our intended purposive sampling framework but we were satisfied that we had captured the experiences of a broad spectrum of patients and did not feel this was a barrier to achieving the study’s aims. Due to the rarity and heterogeneity of the disease, it is not always possible to identify and enrol patients with specific phenotypes to studies of this nature. We also excluded participants that could not speak English. Although our FGs were conducted only in the UK, previous studies (including multi-national recruiting clinical trials) have demonstrated no important differences in the spectrum of DU disease between countries. In our study we captured limited information on the impact of SSc-DU on intimate relationships (18). It is likely that if the data had been collected during one-to-one interviews, then comments on the impact of DUs on intimate relationships would have arisen and should be considered in the design of related future research. We shall explore such themes in a 1:1 setting during future cognitive de-briefing of a provisional item-bank for the proposed novel PRO instrument.

It should be highlighted that treating clinicians (MH and JP) facilitated the FGs which could have impacted on the reflexivity of the research and introduced potential bias, for example, by shaping the discussion and/or limiting patients’ willingness to discuss certain aspects of their experience. However, mitigating factors include the study topic guide which was developed with support from patient insight partners and used to inform the structure of the FGs. Patients were only known to one individual clinician at one geographical location. Furthermore, while background clinical knowledge of SSc was essential to successfully facilitate the FGs, the analysis of data was led by two independent researchers (JJ and AM), to mitigate this potential source of bias.

In conclusion, ours is the first study to examine the multi-faceted patient experience of SSc-DUs. Traditional clinical trial end-points are not currently designed to capture the patient experience of SSc-DUs, which should be a key priority for demonstrating meaningful treatment benefit. The resultant themes and subthemes from our study provide a unique insight into the patient experience of SSc-DUs. This work could form the basis of a novel SSc-DU PRO to assess the impact and severity of SSc-DUs to support much needed new treatment approaches for SSc-DUs.

Acknowledgements

We wish to thank Ms Jane Withey (JW) for her contribution to the project steering committee as a patient research partner. This research was supported by the NIHR Manchester Biomedical Research Centre.

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Table 1: Demographics and clinical phenotype of enrolled participants according to purposive sampling framework.

DcSSc, Diffuse cutaneous systemic sclerosis; DU, digital ulcer; LcSSc, limited cutaneous systemic sclerosis; RP, Raynaud's Phenomenon.

Demographics/clinical phenotype		**Bath**	**Manchester**	**London (1)**	**London (2)**	**Overall**
Number of participants, n		8	7	6	8	29
Age in years (mean, SD)		66.1 (12.6)	61.6 (12.2)	50.4 (12.4)	59.5 (12.8)	59.9 (13.3)
Sex (F:M), n		7:1	7:0	3:3	3:5	20:9
Disease subtype, n	LcSSc	8	6	2	4	20
	DcSSc	0	1	4	4	9
RP duration (mean, SD) (years)		20.7 (19.9)	17.9 (15.9)	23.1 (22.1)	13.6 (9.5)	18.5 (16.6)
Disease duration* (mean, SD) (years)		14.3 (11.2)	10.9 (7.3)	13.9 (12.6)	13.2 (12.2)	12.8 (9.7)
Early vs established disease^, n	Early	0	1	1	0	2
	Established	8	6	5	8	27
History of DU, n	1 previous DU	1	1	0	1	3
	2-4 previous DU	3	3	2	1	9
	≥5 previous DU	4	3	4	6	17
Ethnicity, n	White/ Caucasian	7	6	5	5	23
	Black British	0	1	1	2	4
	Asian	1	0	0	1	2
Vasodilator medication used‡, n	None	1	2	1	2	6
	Calcium channel blocker	5	2	1	2	10
	Phosphodiesterase type-5 inhibitor	5	4	4	5	18
	Endothelin receptor antagonist	3	2	2	2	9

* Since first non-Raynaud's symptom. ^ Early and established disease (≤3 and >3 years since first non-Raynaud's phenomenon symptom, respectively). ‡ Indication not specified and includes SSc-RP, SSc-DU, SSc-pulmonary artery hypertension and/or systemic hypertension/cardiovascular risk.

Table 2: Quotes supporting the "Disabling pain and hypersensitivity" theme of the patient experience of SSc-DU.

Q refers to the numbered quote cited in the text. B1 denotes Bath group; M1 denotes Manchester group; L1, L2 denotes London groups; S # denotes subject (participant) number within each focus group.

Subtheme	*Q*	*Subject*	*Quotation*
Disabling pain and hypersensitivity			
Pain	1	P1 M1:	The pain is just unbearable, in fact you just want to chop your finger off don't you? You think well I'd rather have my finger chopped off than have that pain. I have, I've got to the point where I think just take it off. I can't stand it
	2	P8 B1:	When the pain is really bad you, you just rock back and forward like this
	3	P1 M1:	the pain, I just wanted to sit on the floor and cry … the pain is the worst thing I've had
	4	P6 M1:	I just want it off. It needs to go, it gets that bad. You think, sorry, you feel like you want to bang your head to refer the pain somewhere else, just to relieve it
Pulsatile/ throbbing	5	P7 L2:	You just want to take your finger off, that's how bad it is. The pulsating pain
	6	P7 L2:	Like someone's getting a nail and hammering a nail right through the tip …. And keep going and going, because it just keeps going through the finger
Pressure	7	P2 B1:	If I could have taken my nail off just to release the pressure I would have done
Pain disproportionate to ulcer size	8	P5 M1:	It's quite incongruous the amount of pain from the minimal amount of disruption to your thumb
Radiation	9	P2 B1:	The pain started actually in the finger bed, and I could feel it tracking along the finger and it dipped down into the first joint, so I could actually feel the pain in between the two joints
	10	P7 B1:	So the ulcer is in the middle but I'll still get pain in the index and ring finger which is equivalent to the ulcer pain but there's nothing there
Infection	11	P5 L1:	I try not to get mine infected because then the pain level goes up
Temperature	12	P2 L1:	It's almost impossible to go in, in the summer when they've got the air conditioning on, it's not just the frozen aisles, it's the whole supermarket…. if you've got an ulcer, the change in temperature will make the ulcer sensitive like a nerve, you can really feel it
	13	P1 B1:	I don't go near the freezer for that reason, but even a cold bottle of milk in the winter, if you take it out of the fridge that's enough to set things off. …where I've had the ulcers, particularly that one it, it becomes painful
	14	P3 M2:	It's really tender if I just catch me finger now, but I, luckily I haven't had any more since then, it just left a lot of tenderness on, on the tips of me fingers… it's just the pain where I had the ulcer that's where it's straight away, the cold, as soon as I go out
Pain/ sensitivity at sites of past ulcers	15	P1 L1:	It's the very end of the fingers, it's extremely, erm, sensitive and it doesn't matter if it looks like an ulcer sort of wound, or it might be completely healed up it can still be extremely sensitive to touch
	16	P3 B1:	Just a slight tingly nerve sensation now, no pain
Numbness at sites of past ulcers	17	P1 B1:	It's a bit numb
Considered need for	18	P7 M1:	It's just so painful that the idea of cutting my finger open to take it out seems better than having that pain all the time

invasive procedures			
Need to validate pain	19	P4 B1:	Going back to what you said just now about people seeing it, sometimes you almost want to show, because you can’t explain the pain you get with them, you almost want to show people this is what it’s causing. My family’s seen them obviously but I couldn’t get it across
	20	P1 B1:	Seems a bit feeble ringing and saying I can’t come to work ‘cause my finger’s hurting doesn’t it?”
	21	P2 L1:	Some people don’t understand the pain we’re going through
	22	P1 M1:	But you could cry with them, it is, you could sit down and cry and you can’t explain to anyone in your family how bad the pain is
Description of severity	23	L1 P5:	It was very difficult to be an electrician. Erm, I think, er, the difficulty is the severity of the winter, as you get the ulcer appear during the winter and then it’s the amount of time after the winter they take to heal up
	24	P5 B1:	I’ve had the two digital ulcers, erm, touch wood that’s healed up. I thought I was going to lose this finger at one stage

Table 3: Quotes supporting the “Deep and broad-ranging emotional impact” theme of the patient experience of SSc-DU.

Q refers to the numbered quote cited in the text. B1 denotes Bath group; M1 denotes Manchester group; L1, L2 denotes London groups; S # denotes subject (participant) number within each focus group.

Subtheme	*Q*	*Subject*	*Quotation*
Deep and broad-ranging emotional impact			
Fear	25	P1 M1:	I don’t particularly want to go out when I’ve got one because I’m so frightened of getting in the car and banging or, you know, picking my keys up and banging it
Unavoidable recurrence of DUs	26	P5 L1:	I don’t know if it’s a good or bad thing but I’ve got used to having them so it becomes a way of life …. when I used to maybe have one a year, I used to think it was quite a big deal but then since getting five or six a year, it doesn’t become a big deal any more [laughs], you just get used to it
	27	P4 L2:	Because you can probably guarantee you are going to get another one sometime… I don’t see how you can prevent it, if it’s going to happen, it’s going to happen. I don’t see how it could be…
Anxiety/ uncertainty	28	P7 M1:	I’m still learning about the whole thing so it changes every day, I call it the Hunger Games, when something starts to get better something else happens and you don’t know what is happening so the answer is I don’t know what brings them, I don’t know what I do wrong or not wrong…. it’s one of the worst things about the disease because it makes you scared and it makes you nervous, irritable
Depression / anger / uncertainty about the future	29	P2 L2:	It affected me quite a bit, yes… it really depressed me at that time
	30	P1 M2:	It’s like a black cloud isn’t it? …. It doesn’t tend to go away does it? … Some days you just think well I’m not thinking about it and then other days it …. It gets you down a bit don’t it?”
	31	P7 M1:	Just angry all the time because you have to be conscious and you can’t relax
	32	P6 L2:	It really, it ruins the day, it changes your life
Constant vigilance	33	P3 M1:	you do feel very cautious, if you, if you do have a bang then you’re more, erm, aware that you’re not to do things for the next few days in case it, it goes really bad
	34	P1 M1:	Well I’ve got to be particularly careful now if me nails grow, especially at the side I’ve got to try and cut them… and then of course you’re worried when you cut them that you’re not going to do any damage as well, so it’s a bit difficult really
Anger	35	P7 M2:	Just angry all the time because you have to be conscious and you can’t relax… And it affects you, yes, it affects you and it affects the kids, it affects everything around you. You have to tell yourself all the time, you’ve got this, you have to, you have to remember your hand all the time
Embarrassment/ hiding/ protecting ulcers	36	P8 B1:	I used to hide mine under the table cloth at a function…. Embarrassment, probably
	37	P4 B1:	You don’t want other people be distressed at seeing them, also it’s protection against infection
	38	P2 B1:	So I kept them covered up and I’ve got photographs in my bag that I took for my own record really, you know, and my son said last time ‘don’t you let me see those, I don’t want to see them…’ but even the doctors never looked at my fingers when I had the ulcers
	39	P7 L2:	Sometimes it looks awful, all the skin peeled back and it’s all exposed, yeah, you just hide it… I just don’t want people to look at it as well, I feel conscious sometimes
	40	P4 B1:	If I was going out to a social function or meeting friends or something I would put plasters on, because it’s better for someone to see plasters than, you know, and your friends get used to the fact of, how’s your hands, you know

Table 4: Quotes supporting the “Impairment of physical and social activity theme of the patient experience of SSc-DU.

Q refers to the numbered quote cited in the text. B1 denotes Bath group; M1 denotes Manchester group; L1, L2 denotes London groups; S # denotes subject (participant) number within each focus group.

Subtheme	*Q*	*Subject*	*Quotation*
Functional Impacts of SSc-DUs			
Hand function and activities of daily living	41	P1 L1:	Just trying to handle things with your fingers, you just have to careful you don’t drop a tea cup, your dexterity goes
	42	P5 L1:	Where the ulcers were, sort of like stop the movement in your hands so I wasn’t able to do these things that I needed to do
	43	P4 L4:	To actually bend the fingers where your ulcers are actually on top of the knuckles is practically impossible
	44	P4 B1:	That’s the thing, that’s what I say, I can get things done, but I cannot do it at the speed that I used to before
	45	P5 L1:	Putting things in bags, lifting stuff, you can’t actually grip stuff so I just feel really clumsy
	46	P6 M1:	It’s like opening a bag of crisps if you’re out for a drink, I can’t open the crisps
Self-care/ grooming	47	P1 M1:	I’m frightened of catching it. You don’t want to get dressed in case you’ve got to zip something up and you catch it
	48	P5 L1:	Even just going and brushing our teeth it’s painful when our hands are sore and ulcerated
Hobbies	49	P2 B1:	I’ve had to stop doing things like knitting….Because they flare up straight away and open and it doesn’t matter whether I use natural fibres it’s just the irritation of my skin so I had to give up knitting…. I have to be very careful gardening
Domestic activities	50	P6 L1:	Two years ago I can do nothing really, so I needed help my daughter, husband, everyone doing something at home. I could do nothing, cooking
	51	P8 L2:	It’s impossible to make the bed, I can’t put my hand, I can’t put the sheet under
Putting hands in pockets/ bags/ purse	52	P4 M1:	When it starts to crust over that, that’s when I can’t go in me bag, you know, and you just tip everything out to find what you want and then scoop everything back up
	53	P1 L1:	Putting your hand in your pocket can be horrendous if you hit a key or something like that
Difficulty driving	54	P4 B1:	Things to try and protect it, ‘cause you’re guaranteed knocks on every single day, you carefully put the ignition key in the car, you still knock this one on the steering wheel and things like that
Sleep disturbance	55	P1 M1:	It’s like somebody’s sticking a needle in your finger when you’re trying to go to sleep, you could hold your hand in the air
Shopping	56	P4 L1:	Going to supermarkets I can’t go up and down the fridge aisle. I have to stand there and wait and think about do I need anything down there, but even just going into a supermarket, it’s just too cold… Because you have to balance your bags so that you can carry them, if they’re rushing you, you’re just dropping everything in and it’s all falling out and it just becomes a disaster
	57	P3 L1:	There’s always that doubt in the checkouts, you know, they’re not, not only are they not hassling me, but I’m sort of thinking I’m holding the queue up and I suddenly hear this voice behind me saying, ‘you don’t have to rush you know’ [laughing], people are nice I find
Change in working/ occupation	58	P3 L1:	Well I was a programmer so it wasn’t a difficult job to carry on doing
	59	P7 B1:	Obviously it’s affected a lot of the work that I do as well. There’s only 50% of the work that I used to do that I can continue to do now, with the digital ulcers, but it’s just knowing what you can and can’t get away with anymore
Financial concerns	60	P7 B1:	Most people have said you need to change your job, but once you’re set up and you’re established and you’ve got a wife, kids, a mortgage and bills to pay, it’s impossible to go back and start as tea boy again somewhere else, so you carry on but you’ve got to try and adjust what you do to maintain your income, that’s the biggest difficulty I’ve had so far
Concealing ulcers	61	P4 B1:	You don’t want other people be distressed at seeing them, also it’s protection against infection and also, you know, if you’re going out to any social function I will bandage…. I did go to my daughter’s wedding which was in all of this, and so I did wear my black gloves all through the wedding
	62	P5 L1:	I don’t know if it’s them or myself thinking, oh are they thinking I’m contagious or that kind of thing, because they look horrible when they’re at their worst, but now I’ll try to keep them, I’ll keep them covered if they’re… I wouldn’t go out anywhere without them being covered but still when you’re covered in a million plasters, that doesn’t look nice either

Change in caring roles within the family	63	P7 B1:	It changes the way you have to think of it, everything that you do. I mean the wife says to me, do you want to take the kids down the fair, and the first thing I have to do is check the temperature outside, you know. If it's 20° or less, I'll bail out, I wouldn't bother going, but it's not nice because you miss out on a lot of life experiences with your family

Table 5: Quotes supporting the "Factors aggravating occurrence, duration and impact" and "Mitigating, managing and adapting" themes of the patient experience of SSc-DU. Q refers to the numbered quote cited in the text. B1 denotes Bath group; M1 denotes Manchester group; L1, L2 denotes London groups; S # denotes subject (participant) number within each focus group.

Subtheme	*Q*	*Subject*	*Quotation*
Factors aggravating occurrence, duration and impact			
Number of ulcers	64	P3 M1:	I have only had one ulcer, erm, which was really quite bad. Erm, and it, I was put on a drip in hospital with, is it Epoprostenol, twice to see if that would help, erm, but it didn't and they ended up going to theatre to have it cleaned out, erm, and it's just, and that's the only ulcer I've ever had
	65	P4 B1:	I've had, erm, I was diagnosed with limited scleroderma approximately 28 years ago, erm, which started with an ulcer in one finger and just gradually got worse over the years with anything up to four or five ulcers every winter, which sometimes cleared up in the summer, yeah, so on-going
	66	P2 L1:	When I was first diagnosed ulcers weren't really a problem. I might have one a year but as the scleroderma has progressed I have had up to 10 ulcers at a time on my hands, erm, in different degrees of severity
Ulcers heal slower in the winter	67	P4 M1:	They get easier in the summer, they heal better
	68	P4 L1:	They hey just would erupt through the whole winter and then I've got to wait till the middle to the end of the summer, then I get a short respite
Mitigating, managing and adapting to SSc-DUs			
Indication that treatment is effective	69	P1 B1:	Being able to sleep during the night with the bearable pain would be an absolutely added bonus
	70	P7 B1:	Well within five days that finger healed up more than it did in three months so the minute I came in on the Iloprost, …. certainly the five days I spent here last week, I wouldn't be as healed up as I am now, and able to work again
	71	P7 L2:	If the pain stops
	72	P4 L2:	it helps it, calm it down, to stop being hurting
	73	P5 L1:	I just find it keeps them at bay. I worry that if I was to lengthen it again it would just be worse, erm, so yeah, it sort of helped the aggression that you say, the inflammation and things
Burden of treatments	74	P4 B1:	I think I would definitely say it's helped a lot and it's kept me out of hospital. I've managed, the ulcers are still taking several weeks, if not months to heal but they do heal without the need to intervene with Iloprost on top and a stay in hospital, presumably that's an extra cost to the NHS and it's better for me 'cause I'm not in hospital
	75	P3 L1:	It takes forever to get them on and get them off and then you realise that the reason they're hurting more than usual is you made a complete mess of putting on last time and you've got to start again
Coping strategies/ aids and devices	76	P8 B1:	I keep a pair of gloves up on top of the fridge freezer to do just that, you know, to take anything out from the freezer
	77	P4 L1:	I've got things that help me grip jars
	78	P1 L2:	I also wear gloves, 'cause every time you hit it on something it flares more, that is a big problem I've got no matter, if you touch it, or anything you touch, once you hit it, it flares up again
	79	P8 L2:	The other thing that I've done for the last 18 months, I never, ever, wet them, as least as possible to get them wet, so in the shower I've got rubber gloves
Support from others	80	P4 L1:	They've adapted, my children have, I mean they're grown up now, but they just know I'll just call, they walk in, open a bottle, if I'm cooking and if I look, they know which one, which saucepan to get out, they just know, like in and out of cars, and they just know now, and so do my friends. They just know

	81	P7 M1:	I have three children, erm, and I live alone and it’s not easy because you have to do everything, so you have to cook, you have to touch water and that is something that terrifies you,… It is very difficult but the way to cope about it, I think it is just to explain to them …. and they will know that they have to step up to do something of the things so they, they understand that part, but the other part that you have to live, you have to do it, you have to bath them, you have to do everything else, and you know that you’ll be in pain for that time, all the time. You know it’s going to happen whether you like it or not
Adaptations/ self-management	82	P1 L4:	It’s really good, the pumice stone really helps peel it down
	83	P7 L2:	I think the hardest thing is trying to treat it, and put bandages on it because it’s such awkward positions, you can’t keep the bandage on there and do other things
	84	P2 L1:	It’s just a lot of care that I have to take, and just move very, very slowly, be very aware of your space around you, with my ulcers
	85	P3 M1:	You do feel very cautious, if you, if you do have a bang then you’re more, erm, aware that you’re not to do things for the next few days in case it, it goes really bad.”
	86	P1 B1:	When you put it in hot water or cold water, moving from one room to another it would just set the pain off again.”

Figure 1. A conceptual map comprising the five major inter-related themes that constitute the patient experience of SSc-DUs. The manifestation of pain that is often unbearable affects both the day-to-day functioning of the individual and their psychological well-being. For example, an inability to physically manipulate the world through their hands, can lead to avoidance of activities or social interaction and subsequently cause low mood. This can be supported through the use of aids and devices, such as gloves, or help from other people.