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Exploring the nature of pain related to digital ulceration in systemic sclerosis through focus groups

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Title: Exploring the nature of pain related to digital ulceration in systemic sclerosis through focus groups

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Abstract

Objectives:

Digital ulcers (DUs) are a common complication in systemic sclerosis (SSc). No existing studies have specifically reported on the qualitative patient experience of DU pain and our current patient reported outcome (PRO) instrument does not capture the multi-faceted painful experience of SSc-DU. Our aim was to examine the patient experience of SSc-DU pain.

Design:

Focus groups with people diagnosed with SSc who had experienced DUs were conducted using a topic guide developed by people with SSc, experts in SSc and experienced qualitative researchers. Focus groups were continued until data saturation had been reached. The focus groups were audio recorded, transcribed verbatim, anonymised, and analysed using inductive thematic analysis. Our current study is an integration of the data from these focus groups to specifically examine the patient experience of DU pain.

Setting: Three specialist scleroderma units across the UK (Bath, Manchester and London).

Participants:

Four focus groups were undertaken; 29 adults (20 women, 9 men) with SSc and a spectrum of historical DUs participated. We included participants with a diverse demographic (including ethnic) background and disease-related characteristics.

Results:

Five narrative devices were identified which encompass how people describe the pain from SSc-DUs: ‘Words to express DU associated pain’, ‘Descriptions of physical and psychological reactions to pain’, ‘Comparisons with other painful events’, ‘Descriptions of factors that exacerbate pain’ and ‘Descriptions of strategies for coping with the pain’.

Conclusion:

The experience of SSc-DU pain leads to the use of graphic language and rich description by participants in the focus group setting. Existing SSc-DU outcomes do not adequately capture the patient experiences of SSc-DU pain. Our findings further highlight the multi-faceted nature of SSc-DUs and will hopefully support the development of a novel PRO instrument to assess the severity and impact of SSc-DUs.

Article summary – Strengths and limitations of this study

- First qualitative study to use focus groups to look in-depth at the patient experience of SSc-DU pain.
- Two people diagnosed with SSc-DUs were involved in the development of the topic guide.
- Focus groups facilitated the discussion of participants to deeply understand the patient experience of SSc-DU pain.
- Findings were discussed with a patient partner with DU disease which helped to shape the analysis.
- Sample included only those with a competent level of English language

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Competing interests statement:

Dr. Pauling reports personal fees from Boehringer Ingelheim; grants, personal fees and non-financial support from Actelion Pharmaceuticals and personal fees from Sojournix Pharma, outside the submitted work.

Michael Hughes - has received speaker honoraria (<\$10,000) from Actelion Pharmaceuticals.

JJ, RGH and AM do not report any competing interests relevant to the content of this work.

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Introduction

Studies of the lived experience of people with systemic sclerosis (SSc) indicate that pain is one of the most challenging symptoms of this rare autoimmune connective tissue disease⁽¹⁻³⁾. SSc is characterised by widespread vasculopathy, tissue fibrosis and immune system activation⁽⁴⁾. Around half of people with SSc experience Digital Ulcers (DUs) at some point in their disease course⁽⁵⁾. DUs often recur⁽⁵⁻⁹⁾ and the pain from ulcers limit daily activities and impacts on social, family and work life^(3, 10, 11). A recent study by Hughes and colleagues found five themes which described patient experience of those with 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’*”⁽³⁾. Some people with DUs experience such difficult symptoms that they express a desire to have their fingers amputated^(1, 3). People with SSc-DUs report higher levels of pain than those without DUs^(10, 12, 13).

Pain has been described as “always subjective”⁽¹⁴⁾; the problem of pain and its subjectivity is compounded by a lack of consideration of the first person perspective in guidelines on how to assess pain^(15, 16). For example, the McGill Pain Questionnaire, a multidimensional scaling device for assessing pain includes three measures; a pain rating index, a count of pain descriptors used, and present pain intensity⁽¹⁷⁾, but like most measures it lacks provision for capturing the lived experience of pain. Previous qualitative work has alluded to the pain associated with DUs^(1, 18, 19). However, we have limited information on the lived experience of those with SSc-DUs and how people express specific aspects of their pain⁽¹¹⁾. Previous qualitative research has investigated more deeply how pain is experienced and expressed and how it interferes with one’s life ⁽²⁰⁻³⁴⁾. One of the problems inherent in how we respond clinically to pain is that when pain is assessed it is often measured, with a focus on pain intensity using quantitative scales⁽²³⁾. However, there is an indication that patients use different approaches when rating pain intensity on scales such as the Brief Pain Inventory (BPI) which includes considering how pain fluctuates as well as the location and duration of the pain⁽²⁰⁾. Dannecker et al’s study also found that activity items measured on the Western Ontario and MacMaster Osteoarthritis Index (WOMAC) helped provide context for patients to remember pain experience⁽²⁰⁾.

The Multimodal Assessment Model of Pain (MAP) distinguishes between qualitative (words and behaviours) and quantitative (self and non-self-reported measures) assessments of pain *“and regards the qualitative pain narrative as the best available root proxy for inferring pain in others.”*⁽³⁵⁾. Wideman et al. describe ‘pain expression’ as *“the broad collection of qualitative words and behaviours that communicate pain”*. They divide pain expression into two components: ‘pain narrative’, comprising

words used to describe pain; and ‘pain behaviour’, which comprises non-verbal and para-verbal pain-related behaviours⁽³⁵⁾. Bostick et al found that the ‘pain narrative’ is important for capturing the experience of women who live with chronic pain conditions and suggest that it should be included alongside quantitative pain assessment measures⁽³⁶⁾. Pain is an important symptom of many health conditions, and some conditions have bespoke measurement scales to capture the severity and impact of pain such as WOMAC for osteoarthritis pain⁽³⁷⁾.

The aim of the present study was to explore the nature of pain related to SSc-DUs using data obtained from a multicentre qualitative research study examining the patient experience of SSc-DUs which had identified pain as a key and central focus defining the patient experience⁽³⁾. The specific objectives of this analysis were to firstly capture ‘pain expression’ and the pain narrative of people with SSc-DUs and secondly, to understand how the language and expressions relating to DUs could inform the narrative of clinical consultations.

Methods

Four focus groups were conducted. Participants were recruited from three UK SSc specialist centres who care for patients with DU disease. The focus group topic guide was developed by the steering committee which included people diagnosed with SSc with a history of DUs (appendix 1). In brief, focus groups were chosen for data collection because they allow interaction between participants and the opportunity for any new topics to be discussed that may not have emerged in 1:1 interview settings. Focus groups (FG) continued until thematic saturation had been reached. Participants were adults aged over 18 (range 33-87) with sufficient English language skills to be able to take part in a focus group. Potential participants were identified at routine clinic assessment or through a database of patients who had given prior consent to be considered for future research studies. Focus groups were facilitated by MH, sometimes in conjunction with other SSc specialists and qualitative researchers from the project team⁽³⁾. Ethical approval was provided by the East Midlands – Nottingham 1 research Ethics Committee (REC reference – 18/EM/0018) and all participants provided informed written consent before taking part. We used the SRQR checklist when writing our report⁽³⁸⁾.

Thematic analysis of the transcripts was conducted by JJ and AM with further input from the wider team including MH, JP and RGH and patient partners⁽³⁹⁾. The data were imported into NVivo 11 software to manage and facilitate its interrogation. Our approach was both deductive and inductive. Deductive in the sense that the research team examined pre-conceived considerations of the impact

1 of DUs and how they were understood and managed based on an earlier comprehensive literature
2 review⁽¹¹⁾. The approach was inductive in that there was no pre-existing coding frame and the
3 developing codes were derived from and grounded in the data⁽⁴⁰⁾. Pain was a major element discussed
4 within the focus groups, although other topics discussed included the impact of the SSc-DUs on hand
5 functionality, social functioning and psychological impacts⁽³⁾. This analysis explores the theme of pain
6 in greater depth, for the purposes of developing a future PROM, and to inform discussions about pain
7 between healthcare practitioners and people diagnosed with SSc experiencing DUs.
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15 *Patient and public involvement*

16 People diagnosed with SSc with a history of DUs helped develop the topic guide as part of the
17 steering committee and also contributed to the analysis process by commenting on the first thematic
18 analysis outputs.
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26 Results

27 The study cohort comprised 29 patients with SSc and a history of DUs. A summary of the main
28 patient characteristics is presented in table 1 and has been described previously⁽³⁾.
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32 Participants in the focus groups talked about pain in different ways using a variety of descriptive
33 tools such as metaphor, simile, adjectives and graphic description (see Figure 1).
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37 Five narrative devices were identified which will be expanded upon below:

- 38
- 39 • Words to express Digital Ulcer associated pain
 - 40 • Descriptions of physical and psychological reactions to pain
 - 41 • Comparisons with other painful events
 - 42 • Description of factors that exacerbate pain
 - 43 • Descriptions of strategies for coping with the pain
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48 ***Words to express Digital Ulcer associated pain***

49 Participants used a variety of words to describe the quality of pain associated with their DUs including
50 soreness, tenderness or discomfort. Often words used to describe the intensity of pain were extreme
51 (Table 2).
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55 One participant suggested the words that would best describe pain were all “four letter words”
56 implying that profanity reflected perhaps a mixture of emotions including severity, and frustration
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(FG1, participant (par) 7). There was acknowledgement amongst the participants that the amount of pain experienced was often disproportionate to the size of the ulcer which was causing the pain.

“it’s quite incongruous the amount of pain from the minimal amount of disruption to your thumb” (FG2, par5)

The location of the pain was usually mentioned only if it was beyond the ulcer itself; for example, the pain was described as sometimes appearing in adjacent fingers that were ulcer free or described as shooting up the arm. One participant described how the pain seemed to hurt their whole body if the ulcer was touched.

Participants described great variation in how long the pain from their ulcer lasted with one participant saying they were in pain for half an hour after knocking their ulcer whilst another described how pain was severe for three to four days after an ulcer emerged. Some participants commented on the build-up of a painful internal pressure in the area where an ulcer was about to emerge.

“I feel mine before I see them, I can feel that pain.” (FG3, par4)

“With me, it was a build-up of pressure on my index finger, the actual pad itself,” (FG1, par2)

Descriptions of physical and psychological reactions to pain

Some participants described how they reacted to the pain, for example wanting to cry, roll around on the floor or rock backwards and forwards. One participant spoke of wanting to defer the pain to another part of the body.

“When the pain is really bad you, you just rock back and forward like this” (FG1, par8)

“You feel like you want to bang your head to refer the pain somewhere else, just to relieve it” (FG2, par6)

“The pain, I just wanted to sit on the floor and cry ... the pain is the worst thing I’ve had” (FG2, par1)

“They’re painful if I knock them, I’m on the floor rolling” (FG3, par2)

Participants also described how they reacted psychologically to the pain, expressing feelings of anger, fear, frustration and depression. The state of constant vigilance employed by some participants, added to the psychological burden they experienced.

“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” (FG2, par7)

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Comparisons with other painful events

Some respondents compared the pain of an SSc-DU to other experiences or imagined scenarios. Table 3 summarises a range of similes used (all the respondents used the phrase ‘like’ or ‘as if’). One respondent started with a volcano metaphor which then turned into a simile:

*“You know it’s going to erupt into a volcano, and it starts all rough and there’s a point where it hurts a lot and it just keep on coming out. It does feel **like** a volcano in a weird way. It starts from here and it starts pushing out and it’s that you can’t stop it, it keep’s coming, coming... getting bigger like this, but inside the finger tip.” (FG4, par7)*

In trying to convey how unbearable the pain was, some participants described how they imagined extreme solutions to their pain.

“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.” (FG2, par1)

Description of factors that exacerbate pain

Participants described a variety of ways in which the ulcer could cause pain and how some of their activities were limited as a consequence thus impacting on their social, family and working lives. The main cause of pain was any direct contact with the ulcer such as knocking or catching the ulcer on something. Sometimes the contact did not have to be very strong; one participant described holding a newspaper as being painful. Another participant found that if the ulcer became infected it was more painful and therefore they avoided situations where the risk of infection was higher.

“I try not to get mine infected because then the pain level goes up....even just going and brushing our teeth it’s painful when our hands are sore and ulcerated.” (FG3, par5)

A change of air or water temperature also triggered pain. Some participants described the cold air-conditioned section of supermarkets as being painful. Participants also had to avoid direct contact with cold objects, such as foods in the freezer or fridge as it could be painful, even after ulcers had healed. Having a protective dressing on the ulcer was not enough to prevent pain when the area was touched, or in changing temperatures.

“When you put it in hot water or cold water, moving from one room to another it would just set the pain off again.” (FG1, par2)

Participants accepted that pain would occur when ulcers were present. Most participants described knocking or catching their DU on objects leading to extreme pain, despite their best attempts at protecting the DU to prevent contact. This could impact on social interaction, such as difficulty avoiding

shaking hands, despite knowledge that this could be very painful for them. Another participant observed that having young children meant some activities could not be avoided and so endured pain to enable her to fulfil their parental role.

Description of strategies for coping with the pain

Pain caused by the ulcer determined the extent to which people could or could not participate in daily activities. For some this meant social isolation and reduced work participation. Some continued working in the same job without taking time off, but tried to avoid knocking their ulcers; whereas others had to change role within the organisation, whilst some had either changed their job or had stopped working completely.

Participants described several different methods of coping with the pain. The main method was avoiding situations where the ulcer could get knocked or exposure to temperature changes, or risk of infection, or activities such as gardening or cooking where the hands could potentially be damaged or exposed to irritants (such as infection). Many gave examples of activities they could still do in their family, social or work life but also admitted that there were some activities which they had given up either temporarily or permanently. This was often with much regret but they felt they were controlling the ulcers by doing so.

"I find when you're trying to wash up or anything as well, you know the washing up liquid and stuff like that, you're frightened of even getting that on your hands when you've got an ulcer.... 'Cause I have touched on the odd occasion, you've just touched something that you use every day and you think, oh, has that made it worse?" (FG2, par1)

As well as receiving support from friends and family, participants also used adaptive devices to help avoid painful situations - some of these were bought whilst others were home-made. Participants mentioned tools which helped to open jars and bottles and one participant mentioned having a device which helped with putting on the seat belt otherwise they would have found driving extremely challenging. The most commonly mentioned 'device' was gloves which were used to protect the hands (from knocks and temperature changes). Gloves had the added advantage that in social situations others may avoid shaking hands if they suspected that something was wrong with the gloved hands. See table 4 for coping strategies.

Supermarkets in particular seemed to be a location where participants either avoided or adapted to their situation. One participant said she used a pair of pliers to remove the token from the trolley at

1 the supermarket. Several mentioned that they simply gave the cashier their purse for them to get the
2 money out as it was too painful for them to do so themselves. Home-made devices were often those
3 that helped participants pick up small objects as the ulcer restricted their ability to perform pinching
4 movements in the hand as well as being painful. A few participants also described wound dressings
5 which they had devised which helped protect the ulcer.
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14 Discussion

15 To our knowledge, ours is the first study to comprehensively explore qualitatively the patient
16 experience of SSc-DU pain. The key finding of our study is that people with SSc-DUs express their pain
17 in multiple ways using a rich variety of descriptive tools. We have identified five narrative devices:
18 describing pain sensations; describing the physical and psychological reaction to the pain; comparisons
19 with other painful events or imagined extreme solutions to unbearable pain; descriptions of factors
20 that exacerbate pain and limits participation in the world; and describing the strategies for coping with
21 the pain. These narrative devices resonate with the lived experience of pain described by people with
22 SSc^(1, 11), Raynaud's Phenomenon^(41, 42) and other conditions (e.g osteoarthritis⁽²⁰⁾, pancreatic cancer⁽²¹⁾,
23 dysmenorrhea⁽⁴³⁾, necrotising fasciitis⁽⁴⁴⁾, acute intermittent porphyria⁽²²⁾).
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32 The participants' pain narratives exhibit a diverse use of language; both elements of Wideman's⁽³⁵⁾
33 pain expression (pain narrative and pain behaviour) are evident in the transcripts. Rather than being
34 observed, 'pain behaviour' was described by participants (such as rocking backwards and forwards).
35 This has not been noted in SSc studies before⁽¹¹⁾ but has been observed in studies of other conditions
36 describing pain (i.e. fall to the floor, curl up in a ball, curl up in the fetal position and cry)^(43, 45, 46).
37 Psychological reactions to pain (anger, fear, frustration, depression) were also described in our study
38 as well as previous studies as people talked about how pain overwhelmed or frightened them or
39 negatively impacted their mood leaving them frustrated, aggravated, depressed or hypervigilant⁽²⁵⁻²⁸⁾.
40 This suggests that people are able to convey how they physically and psychologically react to pain
41 without the need for an independent observer to be present in the moment pain is being experienced.
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51 Many studies (including one on Raynaud's Phenomenon⁽⁴¹⁾) have described the language and
52 descriptions used to convey the pain experience of participants in terms of the type of pain
53 experienced (i.e. grinding, cramping, stabbing, burning)^(20-22, 41), and the intensity of the pain (i.e.
54 excruciating, unbearable, worst pain in the world, cannot be controlled by pain medication). Many but
55 not all of these terms used by people in these studies can be found in the McGill Pain Questionnaire
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(MPQ)⁽¹⁷⁾. In our study, those with SSc-DUs used descriptive tools which were beyond the original MPQ list of words suggesting that single words or adjectives cannot fully convey the lived experience of pain. In studies of other conditions, pain intensity could also be described by what relieved the pain in terms of medication⁽²³⁾ or in other cases how medication was not able to alleviate the pain⁽¹⁵⁾. The participants in the SSc-DUs focus groups did not tend to describe their pain in terms of relief through pain medication but instead described strategies they used for coping with the pain while some indicated the extreme scenario of wanting the finger to be amputated/removed to relieve the pain; a similar finding was described by Suarez-Almazor and colleagues⁽¹⁾ when investigating SSc.

Various studies, including those on SSc, have shown that people employ graphic descriptions to convey their experience of pain, making use of analogy, simile and metaphor. These make pain more visual and visceral to the listener^(1, 15, 22-26, 28-30, 43, 44, 47-50). Participants in our focus groups used strong imagery to describe their pain to others in the group; they did not tend to say that they were lost for words to describe their pain. We believe that the willingness to use such imagery to describe pain was in part because focus groups enabled all participants to share their pain narrative. It seems likely that if a clinical consultation similarly enabled participants to describe their pain then they would also provide detailed descriptions of their pain, including simile and other figures of speech.

The patient experience of SSc has a significant impact on people's ability to function⁽¹¹⁾. In our study, ulcer pain was described to limit functionality (social, domestic and work related) within the participant group. The focus group participants described many ways in which they avoided potential painful situations or used aids/devices to help with their day to day living; similar to how people with other painful conditions have described strategies for dealing with their pain^(15, 28). To understand the lived experience of pain better, it is necessary to capture how people employ strategies to live with the pain; this knowledge may help in developing pain management strategies.

People with SSc-DUs talk about pain in many different ways. Single, unidimensional scales such as Visual Analog scales (VAS) that only assess the intensity of the pain experienced exclude the multi-faceted nature of DU pain. Multi-dimensional scales can potentially address this but are still limited in their ability to capture the context and lived experience of pain. People need to express their pain otherwise there is the risk that they become "*isolated in their pain*"⁽⁴⁶⁾, especially if scars have healed and there is the expectation that there should no longer be pain. This is especially pertinent for those with SSc-DUs as participants talked about pain from 'healed' ulcers as well as active ulcers⁽⁵¹⁾.

1 Describing their extreme pain using graphic language seemed to come quite easily to the participants
2 in the focus group setting. However, hearing these pain narratives in a clinical environment may be
3 challenging for some healthcare professionals *“who need the understanding, the will and the humanity*
4 *to hear beyond the words and see behind the picture.”*⁽⁴⁷⁾. As Pither⁽⁵²⁾ says *“Listening and*
5 *acknowledgement are fundamental.”* The need to be believed by both friends and family and especially
6 healthcare professionals came through strongly in studies of other conditions with the
7 recommendation that pain is *“perhaps better captured as a dialogue between HCP and patient rather*
8 *than by the standardized rating scales alone.”*⁽¹⁵⁾. Our study adds to the wealth of studies which
9 recommend listening to the patient narrative alongside capturing a mark on a pain scale^(15, 16, 53). The
10 extreme pain experienced and the specific effect of certain activities on those with DUs highlights the
11 potential value of a specific PRO instrument for DUs that fully captures how patients ‘feel’ and
12 ‘function’. Pain would likely be a central feature. Such a PRO instrument would need to capture the
13 extreme nature of the pain, its duration, when pain happens, and what activities are avoided due to
14 the fear of pain.
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30 Conclusion

31 It is possible there are other descriptive tools which people with SSc-DUs use to describe their pain
32 experience that were not captured in this study. Data were collected from 29 participants across four
33 focus groups who were all able to converse in English so we have no information on how those who
34 express themselves in other languages may speak about pain. Participants in this study were
35 predominantly women. This is a weakness and further research could ensure that more men are
36 included, this is particularly advisable given that previous research has shown gender differences in
37 pain language^(24, 53).
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45 This study was based on focus groups that were designed to understand the patient experience of SSc-
46 DUs including to inform the development of a new PROM for adults with DUs. Pain was not the only
47 element of their experience that was elicited; however, we found that it was a major one. Participants
48 were able to speak freely during the focus groups about their DU experiences and a novel, rich data
49 set has been collected and robustly analysed.
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55 Patients’ experiences of SSc-DU pain are multi-dimensional; and understanding the complexity of SSc-
56 DU pain may support the clinical assessment and the development of novel ulcer outcome measures.
57 These could assess different elements of pain: nature and intensity, physical and psychological
58 reaction; comparing with other known pain events; how pain limits functionality, and how pain is
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managed. Alongside this, it could be valuable for future patient-reported outcomes to include items that elicit and record 'pain expression' qualitatively⁽³⁶⁾, potentially including descriptions of 'pain behaviours', as well as the impact of pain on mood.

Furthermore, in clinical practice decision-making might be enhanced when patients are enabled to speak about their pain in narrative form. Clinicians enabling and acknowledging patients' lived experience of pain may help to legitimise the patient's pain, thereby removing a number of potential barriers to shared decision-making about ulcer pain management. Furthermore, greater understanding of how those with SSc experience DU pain could help to inform the development of novel approaches to the treatment of SSc-DUs.

Author Statement: MH, JP, RGH and AM were on the steering committee which designed the study and developed the topic guide (other members of the steering committee included: experts in SSc; Christopher P Denton, Robyn T Domsic, Tracy M Frech, Ariane L Herrick, Dinesh Khanna, Marco Matucci-Cerinic and Lesley Ann Sakettkoo and two patient research partners; Lorraine McKenzie and Jane Withey). MH undertook the focus groups. JP and AM observed one of the focus groups. JJ and AM initially analysed the data, all authors discussed the analysis and contributed towards the ongoing analysis of the dataset. JJ wrote the first draft of the paper, all authors commented and edited drafts of the paper.

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Data sharing statement

The dataset used during this study is not available for further analysis as consent was only obtained for the data to be used for this study.

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Table 1 Demographics and clinical phenotype of enrolled participants (adapted with permission from Hughes et al⁽³⁾)

Demographics/clinical phenotype		Bath	Manchester	London (1)	London (2)	Overall
Number of participants, n		8	7	6	8	29
Mean age in years, (range)		66.1 (44.0-87.6)	61.6 (41.9-73.3)	50.4 (33.7-67.7)	59.5 (36.7-72.7)	59.9 (33.7-87.6)
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
Mean RP duration in years (range)		20.7 (5.1-54.3)	17.9 (2.0-48.3)	23.1 (2.7-59.7)	13.6 (3.7-32.7)	18.5 (2.0-59.7)
Mean disease duration* in years (range)		14.3 (3.3-33.3)	10.9 (1.7-23.8)	13.9 (1.7-37.7)	13.2 (4.7-31.7)	12.8 (1.7-37.7)
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

DcSSc - Diffuse cutaneous systemic sclerosis;

DU - digital ulcer;

LcSSc, limited cutaneous systemic sclerosis;⁽⁵⁴⁾

RP, Raynaud's Phenomenon.

* Since first non-Raynaud's symptom.

[^] Early and established disease (defined as ≤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 Words to describe intensity and nature of Digital Ulcer pain

'unbelievable'	'agonising'	'really hurt'	'throb'	'constant pain'	'excruciating'
'stabbing feeling'	'absolutely horrible'	'properly painful'	'absolutely agony'	'pulsating pain'	'very painful'
'unbearable'	'severe'	'stingy'	'agony'	'intense pain'	

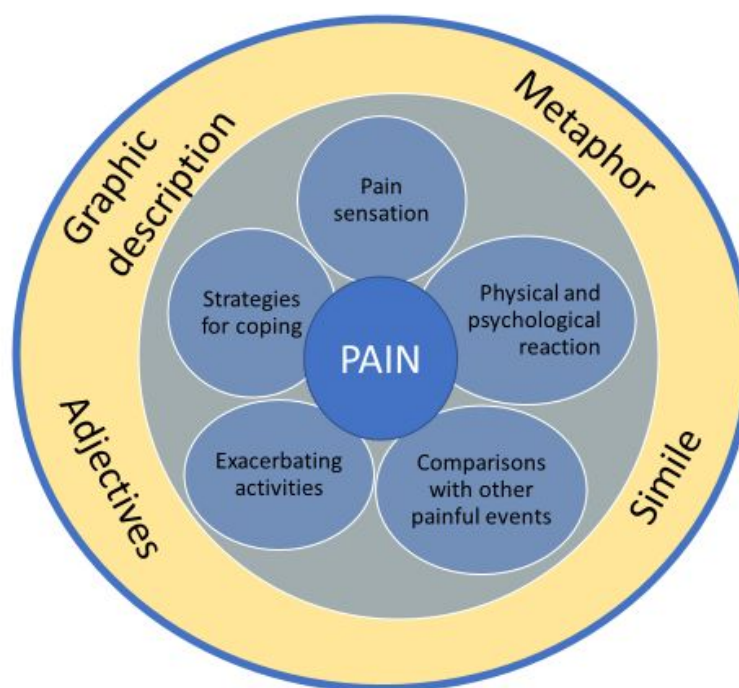
Table 3 Digital Ulcer Pain Similes

<p><i>"To me it's a bit like when you're a child and you fall over and graze your knee and it's open but the, the, you just don't see anything there, there's a slight redness but it almost feels as if you've taken the skin off with a cheese grater and it's exposed."</i> (FG1, par4)</p>
<p><i>"I find the smaller one are like rose thorns stuck in your finger that you can't get out and, it's like three kinds of pain really, another one is where I feel as though I've scalded me finger and then another one is like I just slammed the car door on it."</i> (FG2, par6)</p>
<p><i>"For me, this finger here it's as if somebody's got a needle, a very fine needle and they've just slipped it in and they're just wiggling it, like there, sometimes I sit there and I search to see is there a needle in there?"</i> (FG3, par4)</p>
<p><i>"It feels like your finger, someone's closed the door on your finger and they're holding the door tight and your finger is in the hinge of the door"</i> (FG4, par8)</p>
<p><i>"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"</i> (FG4, par7)</p>
<p><i>"It's like somebody's sticking a needle in your finger when you're trying to go to sleep, you hold your hand up in the air"</i> (FG2, par1)</p>

Table 4 Coping strategies for Digital Ulcers

<i>"Well I've got this thing for my seat belt which is invaluable." (FG2, par 6)</i>
<i>"I wear gloves all the time, in fact I even wear gloves in bed because it keeps your hands warm and that does help me, in fact it helps the ulcers because your hands are warm." (FG4, par 1)</i>
<i>"I've got things that help me grip jars" (FG3, par 4)</i>
<i>"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." (FG1, par 8)</i>
<i>"Avoid cold weather.... Rethinking strategies and things like that.... I have disposable gloves at home for doing things 'cause I've often got dressings on, so I can't wash my hands after cooking or something so I wear rubber gloves to do things." (FG3, par 2)</i>
<i>"One good thing is if you wear gloves people are sort of won't touch your hands, like if they shake hands, if you have gloves on all the time, they sort of think if you're wearing gloves there must be something wrong with you so they won't put their hand</i>
<i>"You can't get your money out of your purse or anything, I have to just tip it on the counter" (FG2, par1)</i>
<i>"They are changing the way supermarket trolleys are now, but I used to carry pliers around with me to get my pound coin" (FG2, par 6)</i>
<i>"I've got little pieces of blue tack around the place so that if things get dropped then I just tap it with a big of blue tack and then I know I've got." (FG1, par 1)</i>
<i>"The other thing I've tried to make is a stand-off dressing. You know you get the thing you put on your finger to put your bandage on, I've used one of those and taped it to the finger to create an extension.... So that I've got something hard, but trying to find something off the shelf that's purpose made for that, I couldn't find anything so I used to try and make my own up, but obviously your finger was then stiff, because it was like having a splint on." (FG1, par7)</i>

Figure 1 Pain elements and descriptive tools



Supplementary material. Appendix 1 The topic guide used for each focus group.

Development of a conceptual framework for a novel patient-reported outcome measure for the assessment of digital ulcers in systemic sclerosis

Focus Group Topic Guide

Preparation as patients arrive:

- Welcome patients, introduce ourselves, explain our role, offer refreshments
- Explain the reason for the focus group and the proposed duration (approximately 1.5 hrs)
- Explain that the focus group discussion will be audio-recorded, transcribed, and anonymised (by removing references to the names of people and places)
- Obtain written consent and offer travel expenses, give large name labels
- Ask participants to complete the Scleroderma Health Assessment Questionnaire (SHAQ)
- Remember to put audiorecorder on a cushioned surface to reduce background noise or on a separate table.

Introduction (5 minutes)

- Explain the 'ground rules' of respecting people's views, allowing others to speak, and preserving confidentiality of what is said during the focus group discussion
- Explain that systemic sclerosis (SSc) and digital ulcers is highly variable and individuals can be affected in very different ways and treatments have evolved in recent years. Therefore not to be alarmed by anything unexpected they hear, but to speak to us afterwards should they have any concerns

Discussion (50 minutes) TURN ON THE RECORDER NOW

1) Ask everyone to introduce themselves and say how long they have had SSc and a history of digital ulcers

2) Ask participants to describe how they would define a digital ulcer?

- What does that term mean to you personally?

3) What happens during as an ulcer is developing?

- Prompts if needed:
Physical signs? Visual signs? Duration? What do they think are the triggers? Do they develop at certain times of the year (e.g. more in the winter?)

4) How do digital ulcers affect you?

- Prompts if needed:
Pain? Quality of life? Psycho-social? Work? Leisure activities? Family roles? Body image dissatisfaction? Uncertainty?

5) How can you manage digital ulcers?

- Prompts if needed:
How do you manage them? Can you prevent them/what provokes them? Can you help to shorten them or reduce the severity (including drug treatments)?

6) How would you know a treatment for digital ulcers was working?

General prompts: Can you give me an example? What did that feel like? What did you do?

7) If you would want clinicians to focus on anything in particular about helping you to care for your ulcers what would it be?

Prompt: how could care be better for you? In an ideal world...

8) What are the most important research questions for you?

9) Are there any questions that we haven't asked or anything that you think is important that we haven't yet discussed in relation to caring for your ulcers?

Closing the group (5 minutes)

- Summarise the focus group discussion (based on notes) and ask participants to confirm whether it is an accurate reflection of the main points that emerged
- Thank participants for their time and input to the discussion

Reporting checklist for qualitative study.

Based on the SRQR guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the SRQR reporting guidelines, and cite them as:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. Standards for reporting qualitative research: a synthesis of recommendations. Acad Med. 2014;89(9):1245-1251.

	Reporting Item	Page Number
Title		
#1	Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	1

Abstract

[#2](#) Summary of the key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results and conclusions 2

Introduction

[#3](#) Problem formulation Description and significance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement 4

[#4](#) Purpose or research question Purpose of the study and specific objectives or questions 5

Methods

[#5](#) Qualitative approach and research paradigm Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. 5-6

1			As appropriate the rationale for several items might be	
2			discussed together.	
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6	Researcher	#6	Researchers' characteristics that may influence the	5-6
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8	characteristics and		research, including personal attributes, qualifications /	
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10	reflexivity		experience, relationship with participants, assumptions	
11			and / or presuppositions; potential or actual interaction	
12			between researchers' characteristics and the research	
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14			transferability	
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22	Context	#7	Setting / site and salient contextual factors; rationale	5-6
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25	Sampling strategy	#8	How and why research participants, documents, or	5
26			events were selected; criteria for deciding when no	
27			further sampling was necessary (e.g. sampling	
28			saturation); rationale	
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35	Ethical issues pertaining	#9	Documentation of approval by an appropriate ethics	5
36			review board and participant consent, or explanation	
37	to human subjects		for lack thereof; other confidentiality and data security	
38			issues	
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45	Data collection methods	#10	Types of data collected; details of data collection	5
46			procedures including (as appropriate) start and stop	
47			dates of data collection and analysis, iterative process,	
48			triangulation of sources / methods, and modification of	
49			procedures in response to evolving study findings;	
50			rationale	
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Data collection instruments and technologies	#11	Description of instruments (e.g. interview guides, questionnaires) and devices (e.g. audio recorders) used for data collection; if / how the instruments(s) changed over the course of the study	5
Units of study	#12	Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	6
Data processing	#13	Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymisation / deidentification of excerpts	5-6
Data analysis	#14	Process by which inferences, themes, etc. were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale	5-6
Techniques to enhance trustworthiness	#15	Techniques to enhance trustworthiness and credibility of data analysis (e.g. member checking, audit trail, triangulation); rationale	5-6
Results/findings			
Syntheses and interpretation	#16	Main findings (e.g. interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	6-10

1	Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts,	6-10
2			photographs) to substantiate analytic findings	
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6	Discussion			
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10	Intergration with prior	#18	Short summary of main findings; explanation of how	10-12
11	work, implications,		findings and conclusions connect to, support, elaborate	
12			on, or challenge conclusions of earlier scholarship;	
13	transferability and		discussion of scope of application / generalizability;	
14	contribution(s) to the field		identification of unique contributions(s) to scholarship	
15			in a discipline or field	
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24	Limitations	#19	Trustworthiness and limitations of findings	12
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27	Other			
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30	Conflicts of interest	#20	Potential sources of influence of perceived influence on	3
31			study conduct and conclusions; how these were	
32			managed	
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38	Funding	#21	Sources of funding and other support; role of funders in	3
39			data collection, interpretation and reporting	
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43 The SRQR checklist is distributed with permission of Wolters Kluwer © 2014 by the Association of
44 American Medical Colleges. This checklist was completed on 07. February 2020 using
45 <https://www.goodreports.org/>, a tool made by the [EQUATOR Network](#) in collaboration with
46 [Penelope.ai](#)
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BMJ Open

What narrative devices do people with systemic sclerosis use to describe the experience of pain from digital ulcers; a multicentre focus group study at UK scleroderma centres

Journal:	<i>BMJ Open</i>
Manuscript ID	bmjopen-2020-037568.R1
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What narrative devices do people with systemic sclerosis use to describe the experience of pain from digital ulcers; a multicentre focus group study at UK scleroderma centres

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Abstract

Objectives:

Digital ulcers (DUs) are a common complication in systemic sclerosis (SSc). No existing studies have specifically reported on the qualitative patient experience of DU pain and our current patient reported outcome measure (PROM) does not capture the multi-faceted painful experience of SSc-DU. Our aim was to examine the patient experience of SSc-DU pain.

Design:

Focus groups with people diagnosed with SSc who had experienced DUs were conducted using a topic guide developed by people with SSc, experts in SSc and experienced qualitative researchers. Focus groups were continued until data saturation had been reached. The focus groups were audio recorded, transcribed verbatim, anonymised, and analysed using inductive thematic analysis. Our current study is an integration of the data from these focus groups to specifically examine the patient experience of DU pain.

Setting: Three specialist scleroderma units across the UK (Bath, Manchester and London).

Participants:

Four focus groups were undertaken; 29 adults (20 women, 9 men) with SSc and a spectrum of historical DUs participated. We included participants with a diverse demographic (including ethnic) background and disease-related characteristics.

Results:

Five narrative devices were identified which encompass how people describe the pain from SSc-DUs: ‘Words to express DU associated pain’, ‘Descriptions of physical and psychological reactions to pain’, ‘Comparisons with other painful events’, ‘Descriptions of factors that exacerbate pain’ and ‘Descriptions of strategies for coping with the pain’.

Conclusion:

The experience of SSc-DU pain leads to the use of graphic language and rich description by participants in the focus group setting. Existing SSc-DU outcomes do not adequately capture the patient experiences of SSc-DU pain. Our findings further highlight the multi-faceted nature of SSc-DUs and will hopefully support the development of a novel PROM to assess the severity and impact of SSc-DUs.

Article summary – Strengths and limitations of this study

- Two people diagnosed with SSc-DUs were involved in the development of the topic guide.
- Focus groups facilitated the discussion of participants to deeply understand the patient experience of SSc-DU pain.
- Findings were discussed with a patient partner with DU disease which helped to shape the analysis.
- Data from all focus groups were analysed and quotes were selected to be illustrative
- Sample included only those with a competent level of English language

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Competing interests statement:

Dr. Pauling reports personal fees from Boehringer Ingelheim; grants, personal fees and non-financial support from Actelion Pharmaceuticals and personal fees from Sojournix Pharma, outside the submitted work.

Michael Hughes - has received speaker honoraria (<\$10,000) from Actelion Pharmaceuticals.

JJ, RGH and AM do not report any competing interests relevant to the content of this work.

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Introduction

Studies of the lived experience of people with systemic sclerosis (SSc) indicate that pain is one of the most challenging symptoms of this rare autoimmune connective tissue disease⁽¹⁻³⁾. SSc is characterised by widespread vasculopathy, tissue fibrosis and immune system activation⁽⁴⁾. Around half of people with SSc experience Digital Ulcers (DUs) at some point in their disease course⁽⁵⁾. DUs often recur⁽⁵⁻⁹⁾ and the pain from ulcers limit daily activities and impacts on social, family and work life^(3, 10, 11). A recent study by Hughes and colleagues found five themes which described patient experience of those with 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’*”⁽³⁾. Some people with DUs experience such difficult symptoms that they express a desire to have their fingers amputated^(1, 3). People with SSc-DUs report higher levels of pain than those with SSc without DUs^(10, 12, 13).

Pain has been described as “always subjective”⁽¹⁴⁾; the problem of pain and its subjectivity is compounded by a lack of consideration of the first person perspective in guidelines on how to assess pain^(15, 16). For example, the McGill Pain Questionnaire, a multidimensional scaling device for assessing pain includes three measures; a pain rating index, a count of pain descriptors used, and present pain intensity⁽¹⁷⁾, but like most measures it lacks provision for capturing the lived experience of pain. Previous qualitative work has alluded to the pain associated with DUs^(1, 18, 19). However, we have limited information on the lived experience of those with SSc-DUs and how people express specific aspects of their pain⁽¹¹⁾. Previous qualitative research in other diagnoses has investigated more deeply how pain is experienced and expressed and how it interferes with one’s life⁽²⁰⁻³⁴⁾. One of the problems inherent in how we respond clinically to pain is that when pain is assessed it is often measured, with a focus on pain intensity using quantitative scales⁽²³⁾. However, there is an indication that patients use different approaches when rating pain intensity on scales such as the Brief Pain Inventory (BPI) which includes considering how pain fluctuates as well as the location and duration of the pain⁽²⁰⁾. Dannecker et al’s study also found that activity items measured on the Western Ontario and MacMaster Osteoarthritis Index (WOMAC) helped provide context for patients to remember pain experience⁽²⁰⁾.

The Multimodal Assessment Model of Pain (MAP) distinguishes between qualitative (words and behaviours) and quantitative (self and non-self-reported measures) assessments of pain *“and regards the qualitative pain narrative as the best available root proxy for inferring pain in others.”*⁽³⁵⁾. Wideman et al. describe ‘pain expression’ as *“the broad collection of qualitative words and behaviours that communicate pain”*. They divide pain expression into two components: ‘pain narrative’, comprising

words used to describe pain; and ‘pain behaviour’, which comprises non-verbal and para-verbal pain-related behaviours⁽³⁵⁾. Bostick et al found that the ‘pain narrative’ is important for capturing the experience of women who live with chronic pain conditions and suggest that it should be included alongside quantitative pain assessment measures⁽³⁶⁾. Pain is an important symptom of many health conditions, and some conditions have bespoke measurement scales to capture the severity and impact of pain such as WOMAC for osteoarthritis pain⁽³⁷⁾.

The aim of the present study was to explore the nature of pain related to SSc-DUs using data obtained from a multicentre qualitative research study examining the patient experience of SSc-DUs which had identified pain as a key and central focus defining the patient experience⁽³⁾. The specific objectives of this analysis were to firstly capture ‘pain expression’ and the pain narrative of people with SSc-DUs and secondly, to understand how the language and expressions relating to DUs could inform the narrative of clinical consultations.

Methods

Four focus groups were conducted. Participants were recruited from three UK SSc specialist centres who care for patients with DU complications. The focus group topic guide was developed by the steering committee which included people diagnosed with SSc with a history of DUs (appendix 1). In brief, focus groups were chosen for data collection because they allow interaction between participants and the opportunity for any new topics to be discussed that may not have emerged in 1:1 interview settings. Focus groups (FG) continued until thematic saturation had been reached. Participants were adults aged over 18 (range 33-87) with sufficient English language skills to be able to take part in a focus group. Potential participants were identified at routine clinic assessment or through a database of patients who had given prior consent to be considered for future research studies. Focus groups were held in informal settings away from the clinical areas and were all facilitated by MH (a consultant rheumatologist with special interest in SSc), sometimes in conjunction with another consultant rheumatologist and SSc specialist (JP) and a qualitative researcher (AM) from the project team⁽³⁾. Focus groups (FG) continued until thematic saturation had been reached, such that no new codes or themes were developing by the fourth focus group⁽³⁸⁾. Ethical approval was provided by the East Midlands – Nottingham 1 research Ethics Committee (REC reference – 18/EM/0018) and all participants provided informed written consent before taking part. We used the SRQR checklist when writing our report⁽³⁹⁾.

1
2 Thematic analysis of the transcripts was conducted by JJ and AM (both experienced qualitative
3 researchers) with further input from the wider team including MH, JP and RGH and patient
4 partners⁽⁴⁰⁾. The data were imported into NVivo 11 software to manage and facilitate its
5 interrogation. Our approach was both deductive and inductive. Deductive in the sense that the
6 research team examined pre-conceived considerations of the impact of DUs and how they were
7 understood and managed based on an earlier comprehensive literature review⁽¹¹⁾. The approach was
8 inductive in that there was no pre-existing coding frame and the developing codes were derived from
9 and grounded in the data⁽⁴¹⁾. Previously reported findings from this study included the range and
10 nature of the impact of SSC-DUs on hand functionality, social functioning, psychological impacts and
11 pain ⁽³⁾. As pain was a major element we felt that a more in-depth exploration was warranted. This
12 analysis explores the theme of pain in greater depth, for the purposes of developing a future Patient
13 reported outcome measure (PROM), and to inform discussions about pain between healthcare
14 practitioners and people diagnosed with SSc experiencing DUs.

25
26 *Patient and public involvement*

27
28 People diagnosed with SSc with a history of DUs helped develop the topic guide as part of the
29 steering committee and also contributed to the analysis process by commenting on the first thematic
30 analysis outputs.

35
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37 Results

38
39 The study cohort comprised 29 patients with SSc and either a history of DUs or current DUs. A
40 summary of the main patient characteristics is presented in table 1 and has been described
41 previously⁽³⁾.

Table 1 Demographics and clinical phenotype of enrolled participants (adapted with permission from Hughes et al ⁽³⁾)

Demographics/clinical phenotype		Bath	Manchester	London (1)	London (2)	Overall
Number of participants, n		8	7	6	8	29
Mean age in years, (range)		66.1 (44.0-87.6)	61.6 (41.9-73.3)	50.4 (33.7-67.7)	59.5 (36.7-72.7)	59.9 (33.7-87.6)
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
Mean RP duration in years (range)		20.7 (5.1-54.3)	17.9 (2.0-48.3)	23.1 (2.7-59.7)	13.6 (3.7-32.7)	18.5 (2.0-59.7)
Mean disease duration* in years (range)		14.3 (3.3-33.3)	10.9 (1.7-23.8)	13.9 (1.7-37.7)	13.2 (4.7-31.7)	12.8 (1.7-37.7)
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

DcSSc - Diffuse cutaneous systemic sclerosis;

DU - digital ulcer;

LcSSc - Limited cutaneous systemic sclerosis; ⁽⁴²⁾

RP - Raynaud's Phenomenon.

* Since first non-Raynaud's symptom.

[^] Early and established disease (defined as ≤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.

1
2 Participants in the focus groups talked about pain using a variety of descriptive tools such as
3 metaphor, simile, adjectives and graphic description (see Figure 1). The participants were found to
4 use these descriptive tools to talk about pain in different ways which we have called ‘narrative
5 devices’. The narrative devices all served the purpose of underlining how severe the pain was and
6 how that was articulated by individuals.

11 Five narrative devices were identified which will be expanded upon below:

- 14 • Words to express Digital Ulcer associated pain
- 15 • Descriptions of physical and psychological reactions to pain
- 16 • Comparisons with other painful events
- 17 • Description of factors that exacerbate pain
- 18 • Descriptions of strategies for coping with the pain

23 **Words to express Digital Ulcer associated pain**

25 Participants used a variety of words to describe the quality of pain associated with their DUs including
26 soreness, tenderness or discomfort. Often words used to describe the intensity of pain were extreme
28 (Table 2).

31 Table 2 Words to describe intensity and nature of Digital Ulcer pain

33 ‘unbelievable’	34 ‘agonising’	35 ‘really hurt’	36 ‘throb’	37 ‘constant pain’	38 ‘excruciating’
39 ‘stabbing feeling’	40 ‘absolutely horrible’	41 ‘properly painful’	42 ‘absolutely agony’	43 ‘pulsating pain’	44 ‘very painful’
45 ‘unbearable’	46 ‘severe’	47 ‘stingy’	48 ‘agony’	49 ‘intense pain’	

51 One participant suggested the words that would best describe pain were all “four letter words”
52 implying that profanity reflected perhaps a mixture of emotions including severity, and frustration
53 (FG1, participant (par) 7). There was acknowledgement amongst the participants that the amount of
54 pain experienced was often disproportionate to the size of the ulcer which was causing the pain.

55 *“it’s quite incongruous the amount of pain from the minimal amount of disruption to your thumb” (FG2, par5)*

56 The location of the pain was usually mentioned only if it was beyond the ulcer itself; for example, the
57 pain was described as sometimes appearing in adjacent fingers that were ulcer free or described as

shooting up the arm. One participant described how the pain seemed to hurt their whole body if the ulcer was touched.

Participants described great variation in how long the pain from their ulcer lasted with one participant saying they were in pain for half an hour after knocking their ulcer whilst another described how pain was severe for three to four days after an ulcer emerged. Some participants commented on the build-up of a painful internal pressure in the area where an ulcer was about to emerge.

"I feel mine before I see them, I can feel that pain." (FG3, par4)

"With me, it was a build-up of pressure on my index finger, the actual pad itself," (FG1, par2)

Descriptions of physical and psychological reactions to pain

Some participants described how they reacted to the pain, for example wanting to cry, roll around on the floor or rock backwards and forwards. One participant spoke of wanting to defer the pain to another part of the body.

"When the pain is really bad you, you just rock back and forward like this" (FG1, par8)

"You feel like you want to bang your head to refer the pain somewhere else, just to relieve it" (FG2, par6)

"The pain, I just wanted to sit on the floor and cry ... the pain is the worst thing I've had" (FG2, par1)

"They're painful if I knock them, I'm on the floor rolling" (FG3, par2)

Participants also described how they reacted psychologically to the pain, expressing feelings of anger, fear, frustration and depression. The state of constant vigilance employed by some participants, added to the psychological burden they experienced.

"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" (FG2, par7)

Comparisons with other painful events

Some respondents compared the pain of an SSc-DU to other experiences or imagined scenarios.

Table 3 summarises a range of similes used (all the respondents used the phrase 'like' or 'as if'). One respondent started with a volcano metaphor which then turned into a simile:

*"You know it's going to erupt into a volcano, and it starts all rough and there's a point where it hurts a lot and it just keep on coming out. It does feel **like** a volcano in a weird way. It starts from here and it starts pushing out and it's that you can't stop it, it keep's coming, coming... getting bigger like this, but inside the finger tip." (FG4, par7)*

Table 3 Digital Ulcer Pain Similes

<i>“To me it’s a bit like when you’re a child and you fall over and graze your knee and it’s open but the, the, you just don’t see anything there, there’s a slight redness but it almost feels as if you’ve taken the skin off with a cheese grater and it’s exposed.” (FG1, par4)</i>
<i>“I find the smaller one are like rose thorns stuck in your finger that you can’t get out and, it’s like three kinds of pain really, another one is where I feel as though I’ve scalded me finger and then another one is like I just slammed the car door on it.” (FG2, par6)</i>
<i>“For me, this finger here it’s as if somebody’s got a needle, a very fine needle and they’ve just slipped it in and they’re just wiggling it, like there, sometimes I sit there and I search to see is there a needle in there?” (FG3, par4)</i>
<i>“It feels like your finger, someone’s closed the door on your finger and they’re holding the door tight and your finger is in the hinge of the door” (FG4, par8)</i>
<i>“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” (FG4, par7)</i>
<i>“It’s like somebody’s sticking a needle in your finger when you’re trying to go to sleep, you hold your hand up in the air” (FG2, par1)</i>

In trying to convey how unbearable the pain was, more than one participant indicated the extreme scenario of wanting to cut the finger open or to have part of the finger or the nail amputated/removed to relieve the pain.

“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.” (FG2, par7)

Description of factors that exacerbate pain

Participants described a variety of ways in which the ulcer could cause pain and how some of their activities were limited as a consequence thus impacting on their social, family and working lives. The main cause of pain was any direct contact with the ulcer such as knocking or catching the ulcer on something. Sometimes the contact did not have to be very strong; one participant described holding a newspaper as being painful. Another participant found that if the ulcer became infected it was more painful and therefore they avoided situations where the risk of infection was higher.

“I try not to get mine infected because then the pain level goes up....even just going and brushing our teeth it’s painful when our hands are sore and ulcerated.” (FG3, par5)

A change of air or water temperature also triggered pain. Some participants described the cold air-conditioned section of supermarkets as being painful. Participants also had to avoid direct contact with cold objects, such as foods in the freezer or fridge as it could be painful, even after ulcers had healed.

Having a protective dressing on the ulcer was not enough to prevent pain when the area was touched, or in changing temperatures.

“When you put it in hot water or cold water, moving from one room to another it would just set the pain off again.” (FG1, par2)

Participants accepted that pain would occur when ulcers were present. Most participants described knocking or catching their DU on objects leading to extreme pain, despite their best attempts at protecting the DU to prevent contact. This could impact on social interaction, such as difficulty avoiding shaking hands, despite knowledge that this could be very painful for them. Another participant observed that having young children meant some activities could not be avoided and so endured pain to enable her to fulfil their parental role.

Description of strategies for coping with the pain

Pain caused by the ulcer determined the extent to which people could or could not participate in daily activities. For some this meant social isolation and reduced work participation. Some continued working in the same job without taking time off, but tried to avoid knocking their ulcers; whereas others had to change role within the organisation, whilst some had either changed their job or had stopped working completely. For some participants it was not solely the pain from their ulcers which had led to occupational changes but also the impact of other SSc issues such as reduced hand mobility.

Participants described several different methods of coping with the pain. The main method was avoiding situations where the ulcer could get knocked or exposure to temperature changes, or risk of infection, or activities such as gardening or cooking where the hands could potentially be damaged or exposed to irritants (such as infection). Many gave examples of activities they could still do in their family, social or work life but also admitted that there were some activities which they had given up either temporarily or permanently. This was often with much regret but they felt they were controlling the ulcers by doing so.

“I find when you’re trying to wash up or anything as well, you know the washing up liquid and stuff like that, you’re frightened of even getting that on your hands when you’ve got an ulcer.... ‘Cause I have touched on the odd occasion, you’ve just touched something that you use every day and you think, oh, has that made it worse?” (FG2, par1)

As well as receiving support from friends and family, participants also used adaptive devices to help avoid painful situations - some of these were bought whilst others were home-made. Participants

mentioned tools which helped to open jars and bottles and one participant mentioned having a device which helped with putting on the seat belt otherwise they would have found driving extremely challenging. The most commonly mentioned ‘device’ was gloves which were used to protect the hands (from knocks and temperature changes). Gloves had the added advantage that in social situations others may avoid shaking hands if they suspected that something was wrong with the gloved hands. See table 4 for coping strategies.

Table 4 Coping strategies for Digital Ulcers

<i>“Well I’ve got this thing for my seat belt which is invaluable.” (FG2, par 6)</i>
<i>“I wear gloves all the time, in fact I even wear gloves in bed because it keeps your hands warm and that does help me, in fact it helps the ulcers because your hands are warm.” (FG4, par 1)</i>
<i>“I’ve got things that help me grip jars” (FG3, par 4)</i>
<i>“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.” (FG1, par 8)</i>
<i>“Avoid cold weather.... Rethinking strategies and things like that.... I have disposable gloves at home for doing things ‘cause I’ve often got dressings on, so I can’t wash my hands after cooking or something so I wear rubber gloves to do things.” (FG3, par 2)</i>
<i>“One good thing is if you wear gloves people are sort of won’t touch your hands, like if they shake hands, if you have gloves on all the time, they sort of think if you’re wearing gloves there must be something wrong with you so they won’t put their hand out.” (FG4, par1)</i>
<i>“They are changing the way supermarket trolleys are now, but I used to carry pliers around with me to get my pound coin” (FG2, par 6)</i>
<i>“I’ve got little pieces of blue tack around the place so that if things get dropped then I just tap it with a big of blue tack and then I know I’ve got.” (FG1, par 1)</i>
<i>“The other thing I’ve tried to make is a stand-off dressing. You know you get the thing you put on your finger to put your bandage on, I’ve used one of those and taped it to the finger to create an extension.... So that I’ve got something hard, but trying to find something off the shelf that’s purpose made for that, I couldn’t find anything so I used to try and make my own up, but obviously your finger was then stiff, because it was like having a splint on.” (FG1, par7)</i>

Supermarkets in particular seemed to be a location where participants either avoided or adapted to their situation. One participant said she used a pair of pliers to remove the token from the trolley at the supermarket. Several mentioned that they simply gave the cashier their purse for them to get the money out as it was too painful for them to do so themselves. Home-made devices were often those that helped participants pick up small objects as the ulcer restricted their ability to perform pinching movements in the hand as well as being painful. A few participants also described wound dressings which they had devised which helped protect the ulcer.

Discussion

To our knowledge, ours is the first study to comprehensively explore qualitatively the patient experience of SSc-DU pain. The key finding of our study is that people with SSc-DUs express their pain in multiple ways using a rich variety of descriptive tools. We have identified five narrative devices: describing pain sensations; describing the physical and psychological reaction to the pain; comparisons with other painful events or imagined extreme solutions to unbearable pain; descriptions of factors that exacerbate pain and limits participation in the world; and describing the strategies for coping with the pain. These narrative devices resonate with the lived experience of pain described by people with SSc^(1, 11), Raynaud's Phenomenon^(43, 44) and other conditions (e.g osteoarthritis⁽²⁰⁾, pancreatic cancer⁽²¹⁾, dysmenorrhea⁽⁴⁵⁾, necrotising fasciitis⁽⁴⁶⁾, acute intermittent porphyria⁽²²⁾).

The participants' pain narratives exhibit a diverse use of language; both elements of Wideman's⁽³⁵⁾ pain expression (pain narrative and pain behaviour) are evident in the transcripts. Rather than being observed, 'pain behaviour' was described by participants (such as rocking backwards and forwards). This has not been noted in SSc studies before⁽¹¹⁾ but has been observed in studies of other conditions describing pain (i.e. fall to the floor, curl up in a ball, curl up in the fetal position and cry)^(45, 47, 48). Psychological reactions to pain (anger, fear, frustration, depression) were also described in our study as well as previous studies as people talked about how pain overwhelmed or frightened them or negatively impacted their mood leaving them frustrated, aggravated, depressed or hypervigilant⁽²⁵⁻²⁸⁾. This suggests that people are able to convey how they physically and psychologically react to pain without the need for an independent observer to be present in the moment pain is being experienced.

Many studies (including one on Raynaud's Phenomenon⁽⁴³⁾) have described the language and descriptions used to convey the pain experience of participants in terms of the type of pain experienced (i.e. grinding, cramping, stabbing, burning)^(20-22, 43), and the intensity of the pain (i.e. excruciating, unbearable, worst pain in the world, cannot be controlled by pain medication). Many but not all of these terms used by people in these studies can be found in the McGill Pain Questionnaire (MPQ)⁽¹⁷⁾. In our study, those with SSc-DUs used descriptive tools which were beyond the original MPQ list of words suggesting that single words or adjectives cannot fully convey the lived experience of pain. In studies of other conditions, pain intensity could also be described by what relieved the pain in terms of medication⁽²³⁾ or in other cases how medication was not able to alleviate the pain⁽¹⁵⁾. The participants in the SSc-DUs focus groups did not tend to describe their pain in terms of relief through pain medication but instead described strategies they used for coping with the pain while some

indicated the extreme scenario of wanting the finger to be amputated/removed to relieve the pain; a similar finding was described by Suarez-Almazor and colleagues⁽¹⁾ when investigating SSc.

Various studies, including those on SSc, have shown that people employ graphic descriptions to convey their experience of pain, making use of analogy, simile and metaphor. These make pain more visual and visceral to the listener^(1, 15, 22-26, 28-30, 45, 46, 49-52). Participants in our focus groups used strong imagery to describe their pain to others in the group; they did not tend to say that they were lost for words to describe their pain. We believe that the willingness to use such imagery to describe pain was in part because focus groups enabled all participants to share their pain narrative. It seems likely that if a clinical consultation similarly enabled participants to describe their pain then they would also provide detailed descriptions of their pain, including simile and other figures of speech.

The patient experience of SSc-DU has a significant impact on people's ability to function⁽¹¹⁾. In our study, ulcer pain was described to limit functionality (social, domestic and work related) within the participant group. The focus group participants described many ways in which they avoided potential painful situations or used aids/devices to help with their day to day living; similar to how people with other painful conditions have described strategies for dealing with their pain^(15, 28). To understand the lived experience of pain better, it is necessary to capture how people employ strategies to live with the pain; this knowledge may help in developing pain management strategies.

People with SSc-DUs talk about pain in many different ways. Single, unidimensional scales such as Visual Analogue Scales (VAS) that only assess the intensity of the pain experienced exclude the multi-faceted nature of DU pain. Multi-dimensional scales can potentially address this but are still limited in their ability to capture the context and lived experience of pain. People need to express their pain otherwise there is the risk that they become "*isolated in their pain*"⁽⁴⁸⁾, especially if scars have healed and there is the expectation that there should no longer be pain. This is especially pertinent for those with SSc-DUs as participants talked about pain from 'healed' ulcers as well as active ulcers⁽⁵³⁾.

Describing their extreme pain using graphic language seemed to come quite easily to the participants in the focus group setting. However, hearing these pain narratives in a clinical environment may be challenging for some healthcare professionals "*who need the understanding, the will and the humanity to hear beyond the words and see behind the picture.*"⁽⁴⁹⁾. As Pither⁽⁵⁴⁾ says "*Listening and acknowledgement are fundamental.*" The need to be believed by both friends and family and especially healthcare professionals came through strongly in studies of other conditions with the recommendation that pain is "*perhaps better captured as a dialogue between HCP and patient rather*

1 *than by the standardized rating scales alone.”⁽¹⁵⁾. Our study adds to the wealth of studies which*
2
3 *recommend listening to the patient narrative alongside capturing a mark on a pain scale^(15, 16, 55). The*
4
5 *extreme pain experienced and the specific effect of certain activities on those with DUs highlights the*
6
7 *potential value of a specific PROM for DUs that fully captures how patients ‘feel’ and ‘function’. Pain*
8
9 *would likely be a central feature. Such a PROM would need to capture the extreme nature of the pain,*
10
11 *its duration, when pain happens, and what activities are avoided due to the fear of pain.*
12

13 14 15 Conclusion

16
17 It is possible there are other descriptive tools which people with SSc-DUs use to describe their pain
18
19 experience that were not captured in this study. Data were collected from 29 participants across four
20
21 focus groups who were all able to converse in English so we have no information on how those who
22
23 express themselves in other languages may speak about pain. Participants in this study were
24
25 predominantly women. This is a weakness and further research could ensure that more men are
26
27 included, this is particularly advisable given that previous research has shown gender differences in
28
29 pain language^(24, 55).
30

31 This study was based on focus groups that were designed to understand the patient experience of SSc-
32
33 DUs including to inform the development of a new PROM for adults with DUs. Pain was not the only
34
35 element of their experience that was elicited; however, we found that it was a major one. Participants
36
37 were able to speak freely during the focus groups about their DU experiences and a novel, rich data
38
39 set has been collected and robustly analysed.
40

41 Patients’ experiences of SSc-DU pain are multi-dimensional; and understanding the complexity of SSc-
42
43 DU pain may support the clinical assessment and the development of novel ulcer outcome measures.
44
45 These could assess different elements of pain: nature and intensity, physical and psychological
46
47 reaction; comparing with other known pain events; how pain limits functionality, and how pain is
48
49 managed. Alongside this, it could be valuable for future PROMS to include items that elicit and record
50
51 ‘pain expression’ qualitatively⁽³⁶⁾, potentially including descriptions of ‘pain behaviours’, as well as the
52
53 impact of pain on mood.

54 Furthermore, in clinical practice decision-making might be enhanced when patients are enabled to
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56 speak about their pain in narrative form. Clinicians enabling and acknowledging patients’ lived
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58 experience of pain may help to legitimise the patient’s pain, thereby removing a number of potential
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60 barriers to shared decision-making about ulcer pain management. Furthermore, greater

1 understanding of how those with SSc experience DU pain could help to inform the development of
2 novel approaches to the treatment of SSc-DUs.
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7

8 **Author Statement:** MH, JP, RGH and AM were on the steering committee which designed the study
9 and developed the topic guide. MH undertook the focus groups. JP and AM observed one of the focus
10 groups. JJ and AM initially analysed the data, all authors discussed the analysis and contributed
11 towards the ongoing analysis of the dataset. JJ wrote the first draft of the paper, all authors
12 commented and edited drafts of the paper.
13
14
15
16
17

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22 and sharing their stories and experiences.
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31 **Data sharing statement**
32

33
34 The dataset used during this study is not available for further analysis as consent was only obtained
35 for the data to be used for this study.
36
37
38

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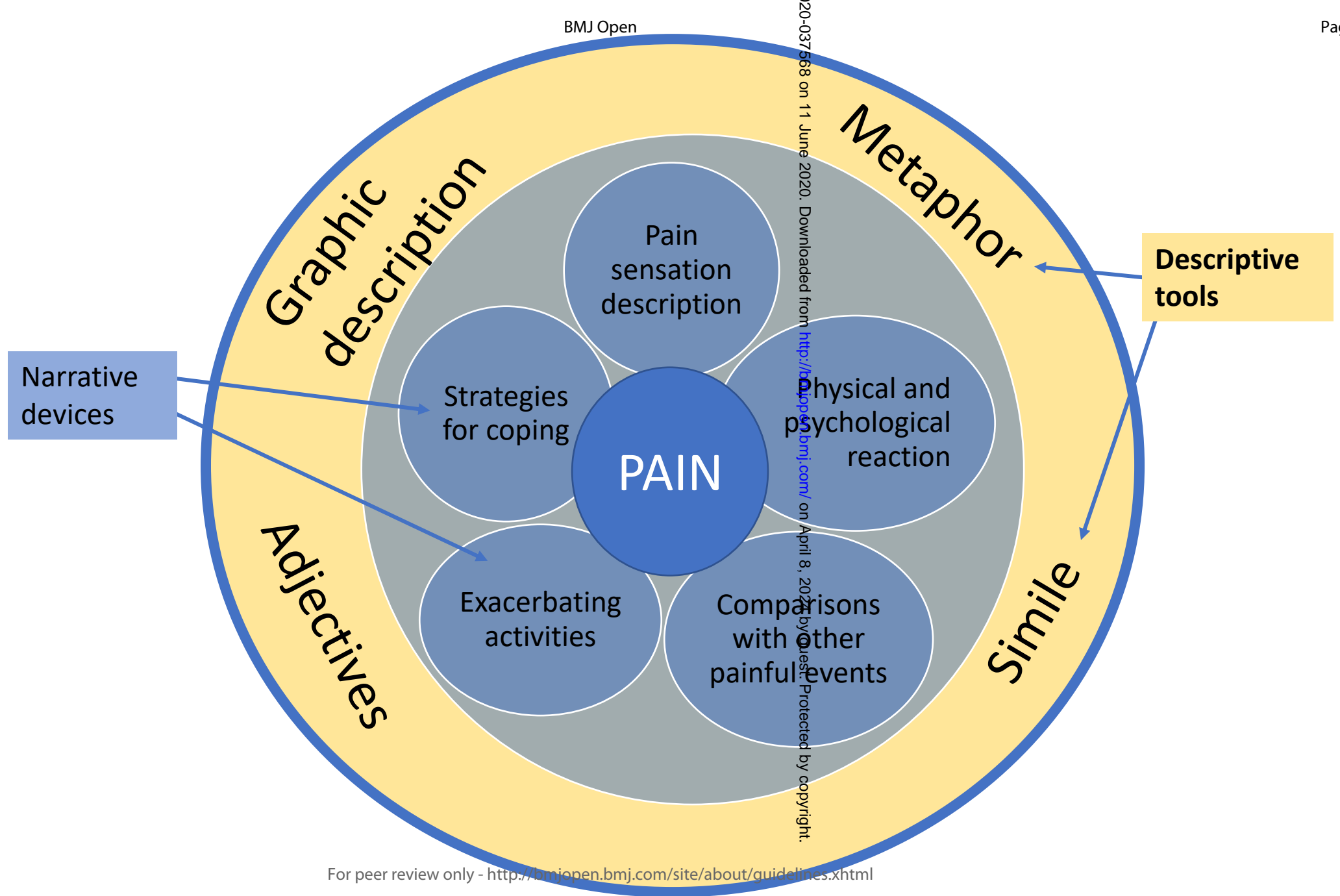
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1
2 Title for Figure 1:
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6 Figure 1: ***Pain experience: narrative devices and descriptive tools***
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Supplementary material. Appendix 1 The topic guide used for each focus group.

Development of a conceptual framework for a novel patient-reported outcome measure for the assessment of digital ulcers in systemic sclerosis

Focus Group Topic Guide

Preparation as patients arrive:

- Welcome patients, introduce ourselves, explain our role, offer refreshments
- Explain the reason for the focus group and the proposed duration (approximately 1.5 hrs)
- Explain that the focus group discussion will be audio-recorded, transcribed, and anonymised (by removing references to the names of people and places)
- Obtain written consent and offer travel expenses, give large name labels
- Ask participants to complete the Scleroderma Health Assessment Questionnaire (SHAQ)
- Remember to put audiorecorder on a cushioned surface to reduce background noise or on a separate table.

Introduction (5 minutes)

- Explain the ‘ground rules’ of respecting people’s views, allowing others to speak, and preserving confidentiality of what is said during the focus group discussion
- Explain that systemic sclerosis (SSc) and digital ulcers is highly variable and individuals can be affected in very different ways and treatments have evolved in recent years. Therefore not to be alarmed by anything unexpected they hear, but to speak to us afterwards should they have any concerns

Discussion (50 minutes) TURN ON THE RECORDER NOW

- 1) Ask everyone to introduce themselves and say how long they have had SSc and a history of digital ulcers
- 2) Ask participants to describe how they would define a digital ulcer?
 - What does that term mean to you personally?
- 3) What happens during as an ulcer is developing?
 - Prompts if needed:
Physical signs? Visual signs? Duration? What do they think are the triggers? Do they develop at certain times of the year (e.g. more in the winter?)
- 4) How do digital ulcers affect you?
 - Prompts if needed:
Pain? Quality of life? Psycho-social? Work? Leisure activities? Family roles? Body image dissatisfaction? Uncertainty?

5) How can you manage digital ulcers?

- Prompts if needed:
How do you manage them? Can you prevent them/what provokes them? Can you help to shorten them or reduce the severity (including drug treatments)?

6) How would you know a treatment for digital ulcers was working?

General prompts: Can you give me an example? What did that feel like? What did you do?

7) If you would want clinicians to focus on anything in particular about helping you to care for your ulcers what would it be?

Prompt: how could care be better for you? In an ideal world...

8) What are the most important research questions for you?

9) Are there any questions that we haven't asked or anything that you think is important that we haven't yet discussed in relation to caring for your ulcers?

Closing the group (5 minutes)

- Summarise the focus group discussion (based on notes) and ask participants to confirm whether it is an accurate reflection of the main points that emerged
- Thank participants for their time and input to the discussion

Reporting checklist for qualitative study.

Based on the SRQR guidelines.

Instructions to authors

Complete this checklist by entering the page numbers from your manuscript where readers will find each of the items listed below.

Your article may not currently address all the items on the checklist. Please modify your text to include the missing information. If you are certain that an item does not apply, please write "n/a" and provide a short explanation.

Upload your completed checklist as an extra file when you submit to a journal.

In your methods section, say that you used the SRQRreporting guidelines, and cite them as:

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	Reporting Item	Page Number
Title	#1 Concise description of the nature and topic of the study identifying the study as qualitative or indicating the approach (e.g. ethnography, grounded theory) or data collection methods (e.g. interview, focus group) is recommended	1
Abstract	#2 Summary of the key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results and conclusions	2
Introduction	#3 Description and significance of the problem / phenomenon studied: review of relevant theory and empirical work; problem statement	4

Purpose or research question	#4	Purpose of the study and specific objectives or questions	5
Methods			
Qualitative approach and research paradigm	#5	Qualitative approach (e.g. ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g. postpositivist, constructivist / interpretivist) is also recommended; rationale. The rationale should briefly discuss the justification for choosing that theory, approach, method or technique rather than other options available; the assumptions and limitations implicit in those choices and how those choices influence study conclusions and transferability. As appropriate the rationale for several items might be discussed together.	5-6
Researcher characteristics and reflexivity	#6	Researchers' characteristics that may influence the research, including personal attributes, qualifications / experience, relationship with participants, assumptions and / or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results and / or transferability	5-6
Context	#7	Setting / site and salient contextual factors; rationale	5-6
Sampling strategy	#8	How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g. sampling saturation); rationale	5
Ethical issues pertaining to human subjects	#9	Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	5
Data collection methods	#10	Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative	5

1			process, triangulation of sources / methods, and	
2			modification of procedures in response to evolving	
3			study findings; rationale	
4				
5	Data collection	#11	Description of instruments (e.g. interview guides,	5
6	instruments and		questionnaires) and devices (e.g. audio recorders)	
7	technologies		used for data collection; if / how the instruments(s)	
8			changed over the course of the study	
9				
10				
11				
12	Units of study	#12	Number and relevant characteristics of participants,	6-7
13			documents, or events included in the study; level of	
14			participation (could be reported in results)	
15				
16				
17	Data processing	#13	Methods for processing data prior to and during	5-6
18			analysis, including transcription, data entry, data	
19			management and security, verification of data	
20			integrity, data coding, and anonymisation /	
21			deidentification of excerpts	
22				
23				
24				
25				
26	Data analysis	#14	Process by which inferences, themes, etc. were	5-6
27			identified and developed, including the researchers	
28			involved in data analysis; usually references a specific	
29			paradigm or approach; rationale	
30				
31				
32				
33	Techniques to enhance	#15	Techniques to enhance trustworthiness and credibility	5-6
34	trustworthiness		of data analysis (e.g. member checking, audit trail,	
35			triangulation); rationale	
36				
37				
38	Results/findings			
39				
40	Syntheses and	#16	Main findings (e.g. interpretations, inferences, and	6-12
41	interpretation		themes); might include development of a theory or	
42			model, or integration with prior research or theory	
43				
44				
45				
46	Links to empirical data	#17	Evidence (e.g. quotes, field notes, text excerpts,	6-12
47			photographs) to substantiate analytic findings	
48				
49	Discussion			
50				
51				
52	Intergration with prior	#18	Short summary of main findings; explanation of how	13-16
53	work, implications,		findings and conclusions connect to, support,	
54	transferability and		elaborate on, or challenge conclusions of earlier	
55	contribution(s) to the field		scholarship; discussion of scope of application /	
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generalizability; identification of unique contributions(s) to scholarship in a discipline or field

Limitations	#19	Trustworthiness and limitations of findings	3,15
Other			
Conflicts of interest	#20	Potential sources of influence of perceived influence on study conduct and conclusions; how these were managed	3
Funding	#21	Sources of funding and other support; role of funders in data collection, interpretation and reporting	3

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