

1 Understanding the daily experiences of living with Sjögren’s
2 syndrome: insights for smartphone app co-design from qualitative co-
3 design workshops

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15

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1 **Abstract**

2 **Background:**

3 Sjögren’s syndrome (SS) is the second most common autoimmune rheumatic disease, and the range
4 of symptoms includes fatigue, dryness, sleep disturbances, and pain. Smartphone applications (apps)
5 may help deliver a variety of cognitive and behavioural techniques to support self-management in
6 SS. However, app-based interventions must be carefully designed to promote engagement and
7 motivate behaviour change.

8 **Objective:**

9 To explore self-management approaches and challenges experienced by people living with SS and
10 produce a corresponding set of design recommendations that inform the design of an engaging,
11 motivating, evidence-based self-management app for those living with SS.

12 **Methods:**

13 We conducted a series of eight co-design workshops and an additional three interviews with
14 participants who were unable to attend a workshop. These were audio recorded, transcribed, and
15 initially thematically analysed using an inductive approach. Themes were then mapped to the self-
16 determination theory (SDT) domains of competency, autonomy, and relatedness.

17 **Results:**

18 Participants experienced a considerable demand in the daily work required in self-managing their SS.
19 The mental, physical, and social work that the condition required was never stable, never
20 predictable, and never fully over. Participants employed a wide variety of techniques to self-manage
21 their symptoms, however their sense of competency was undermined by the complexity and
22 interconnected nature of their symptoms and affected by interactions with others. The daily
23 contexts in which this labour was taking place revealed ample opportunities to use digital health
24 aids. The lived experience of participants showed that the constructs of competency, autonomy and
25 relatedness existed in a complex equilibrium with each other. At times, they were disrupted by
26 tensions while on other occasions they worked together harmoniously.

27 **Conclusions:**

28 A SS self-management app needs to recognise the complexity and overlap of symptoms and the
29 complexities of managing the condition in daily life. Identifying techniques which target several
30 symptoms at once may reduce overwhelm for users. Including techniques which support

1 assertiveness and communication with others about the condition, its symptoms and users'
2 limitations, may support users in their interactions with others and support engagement in symptom
3 management strategies.

4 For digital health aids (such as self-management apps) to provide meaningful support, they should
5 be designed according to human needs of competence, autonomy, and relatedness. However, the
6 complexities between the three SDT constructs should be carefully considered, as they present both
7 design difficulties but also opportunities.

8

9 **Keywords:**

10 Self-Management; Mobile Health; eHealth; mHealth; Sjogrens Syndrome; Patient Participation;
11 Patient Involvement; Fatigue; Chronic Disease; Focus groups; complex intervention development

1 Introduction

2 Background

3 The need to improve the accessibility and quality of care for those with long-term conditions (LTCs)
4 is an international priority[1]. In England alone, LTCs affect 15 million people[2] and account for 70%
5 of healthcare spending[3]. Rheumatic diseases are LTCs with a particularly high UK and global
6 prevalence, having been estimated to affect up to one-quarter of Europeans[4, 5] and a similar
7 proportion of the population in the global south[6]. Sjögren’s syndrome (SS) is thought to be the
8 second most common autoimmune rheumatic disease[7] and is associated with poor quality of
9 life[8] and high disease burden[9].

10

11 SS is a heterogeneous LTC with a constellation of unpredictable and diverse symptoms[10, 11]. A key
12 characteristic of SS is mucosal dryness due to the destruction of exocrine (moisture-producing)
13 glands by the body’s immune system, which particularly affects the eyes, mouth and vagina[12]. In
14 addition to dryness, common extra glandular features include persistent fatigue[13], chronic
15 pain[14], sleep disturbances[15], and anxiety and depression[16]. People with SS report
16 experiencing these symptoms as being interconnected, with the exacerbation of one symptom
17 impacting others[17-19].

18

19 Like many other autoimmune diseases, SS does not have a cure[20]. Intervention efforts have
20 therefore focussed on reducing the severity of symptoms, for instance, topical treatments are used
21 for dryness management[21]. Drug treatments for the systemic management of SS, such as
22 hydroxychloroquine and rituximab, have had disappointing results in clinical trials[22, 23].
23 Behavioural interventions that aim to improve quality of life are a promising alternative; however,
24 few have been developed and evaluations of their impact have been of low quality[21, 24]. A recent
25 stakeholder engagement study found that support to self-manage symptoms was a key priority for
26 people with SS[25]. The term self-management has been defined as “the individual's ability to
27 manage the symptoms, treatment, physical and psychosocial consequences and lifestyle changes
28 inherent in living with a chronic condition” [26, 27]. To support the knowledge, behaviours, and
29 attitudes required, self-management interventions should deliver a range of educational,
30 behavioural, and cognitive techniques[28]. In SS, a targeted ‘complex’ intervention is required, which
31 delivers multiple techniques and targets multiple SS symptoms[29]. Our previous body of work with
32 SS patients found that people with SS require different levels of support. Some require more

1 complex individual support, but most people require lower levels with access to written information
2 and digital self-management tools[29] which could be provided in the form of a website or
3 smartphone application (app).

4

5 **Apps as a support for self-management**

6 SS shares multiple symptom and self-management similarities with other LTCs[30] including, but not
7 limited to neurological and autoimmune conditions such as rheumatoid arthritis(RA), myalgic
8 encephalomyelitis (ME), and multiple sclerosis (MS). Smartphone apps are a promising approach to
9 self-managing these LTCs [31, 32] [32] and others such as type 2 diabetes[33], asthma[34], and
10 hypertension[35]. Their increasing availability and functionalities enable complex intervention
11 techniques to be delivered in the context of users' daily lives when they are designed with
12 consideration for users' routines and choices[36]. User-centred design work on LTCs has produced
13 various app features and content[37] to support for example, user education and cognitive
14 strategies. However, app effectiveness can be limited through notoriously low levels of user
15 engagement[38, 39]. Intervention developers must therefore design apps that are more engaging,
16 and carefully consider how such engagement will ultimately lead to long-term behaviour change[40].
17 For example, beyond simply providing information on how to perform techniques, apps can be
18 designed to promote a sense of autonomy and motivation to engage in these self-management
19 behaviours over time[41, 42].

20

21 To increase user engagement, apps should be user- and person-centred[43], i.e. designed to fit
22 within individuals' current lives and daily activities[44]. People are more likely to use a new
23 intervention if it can be incorporated into their existing habits, routines and contexts[45, 46]. Self-
24 management interventions should therefore account for, and actively support, how people currently
25 manage their conditions[31, 47]. To facilitate this support and encourage user engagement, apps
26 should be user- and person-centred[43], i.e. designed to fit within individuals' current lives and daily
27 activities[44]. When exposed to any new intervention, people are more likely to adapt use a new
28 intervention if it can be incorporated into their existing habits, routines and contexts[45, 46]. Self-
29 management interventions should therefore account for, and actively support, how people currently
30 manage their conditions[31, 47]. Thus, to develop a useful, effective, and engaging app-based
31 intervention that supports those with SS, there is a need to first understand their current self-
32 management opportunities and challenges. To date, limited research has only gone as far as to

1 understand the lived experience of symptoms [17, 19, 48], and not explored self-management of
2 multiple diverse symptoms.

3

4 To gain an understanding of individuals' self-management contexts, co-design and user-centred
5 methods are useful[49]. These can involve practical design activities that elicit conversations around
6 a topic of interest (such as self-management) to inform the development of a design, product or
7 intervention, and have been used to develop digital health interventions[50, 51]. To then
8 understand how users in these contexts might best be supported in changing their self-management
9 behaviour, co-design findings can be interpreted using theories of motivation and behaviour change
10 [41].

11

12 Self-Determination Theory (SDT)[52] is one motivational theory widely used in interventions
13 promoting health behaviour change[53, 54], including those to self-manage chronic illnesses[55, 56].
14 SDT proposes that the constructs of competence, autonomy, and relatedness are required for
15 individuals to be internally motivated to perform behaviours and sustain these changes over time.
16 Situating qualitative findings within theoretical constructs facilitates the development of apps that
17 have a grounding in theory[42, 57]. While intervention developers use SDT to inform their
18 interventions, many do not explicitly link the theoretical constructs directly to their individual
19 components and we seek to bridge this gap. To our knowledge, there are no evidence-based, theory-
20 driven, self-management apps for SS.

21

22 **Study aims**

23 To employ an SDT framework to explore self-management challenges and approaches used by
24 people with SS and to produce a set of design and therapeutic recommendations for a supportive
25 and engaging app to aid self-management.

26

27 **Methods**

28 The methods and subsequent results have been reported in line with the COREQ guidelines for the
29 reporting of qualitative research[58] (Multimedia Appendix 1).

30 **Study design**

1 A consecutive series of eight workshops with people living with SS were conducted over seven
2 months, each involving design activities and focussed discussions (see Figure 1). The first two
3 workshops were open-ended to broadly understand participants' contexts (i.e., key self-
4 management challenges and overall self-management routines) and enable participants to get to
5 know each other and to feel comfortable while discussing potentially sensitive and personal topics.
6 We decided in advance to include a series of workshops, with each one dedicated to understanding
7 in depth the self-management activities, challenges and opportunities for each symptom. However,
8 the order of symptom workshops and their exact discussion topics and activities were not pre-
9 determined: their sequential nature enabled us to iteratively design topics based on findings from
10 the previous session. For example, a clear theme emerged around symptom interrelatedness, so
11 subsequent workshops included discussions on how participants managed interrelations between
12 their symptoms. Furthermore, fatigue was a priority for many participants and so two workshops
13 were dedicated to this symptom.

14 Participants were given the option to attend one or more workshops. Several were repeated to suit
15 participants availability. To enable those who could not physically attend any workshops to
16 participate, and to include the experiences of younger people living with SS, three further one-off
17 semi-structured interviews were conducted. These focussed on the key self-management practices
18 and challenges experienced by that individual. This qualitative study received ethical approval from
19 (anon.) University ethics committee (XXXX).

20

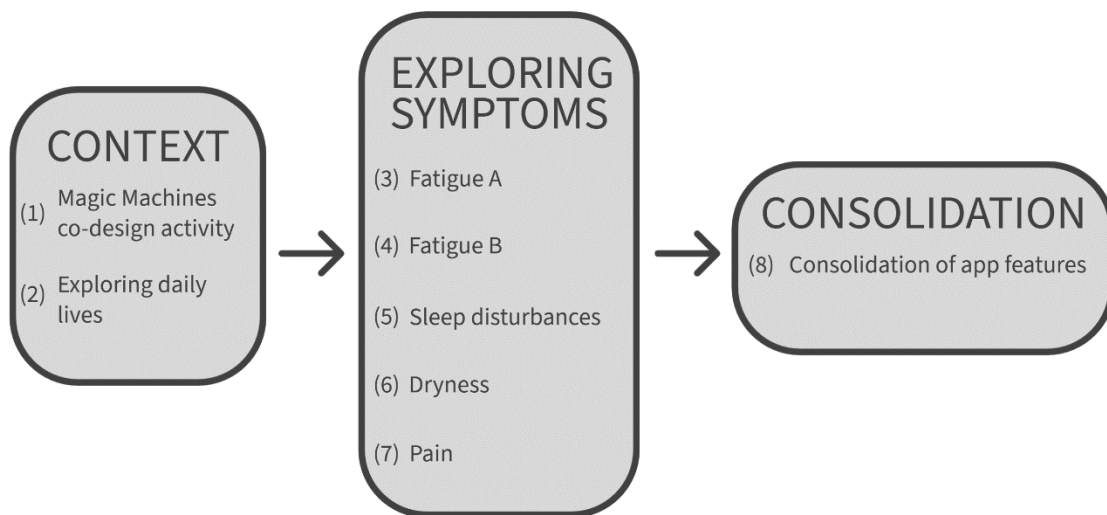
21 **Recruitment**

22 Workshop participants were purposively recruited from a regional UK SS support group (Northeast
23 Sjögren's Syndrome Association). Adverts were distributed via their member mailing list and
24 Facebook page, and the research team also presented the project at a support group meeting. The
25 call was open to those diagnosed with SS by a physician and potential participants were invited to
26 attend as many workshops as they liked. Interested participants unable to attend due to their
27 location or life commitments were invited to attend a video interview online. Additional participants
28 were recruited via social media (a single tweet on Twitter) and invited to participate in interviews
29 only.

30

31 **Data collection activities**

1 Informed consent was obtained before data collection and travel costs were reimbursed.
2 Workshops took place at (anon.) University, lasted approximately 90 minutes and included a 10-
3 minute comfort break. The interviews were 30-60 minutes long and conducted by telephone or
4 video conferencing software. Workshops were facilitated by three of the authors (xx, xx, xxx), all
5 female post-doctoral (PhD) researchers trained in qualitative research methods and experienced in
6 conducting qualitative research interviews and focus groups; one of whom was also an occupational
7 therapist (xx) with experience of SS symptom management. Several participants (n=4) had attended
8 clinics (conducted by XX) and two had taken part in previous research (conducted by xx and xxx). All
9 participants were briefed on the aims of the research. All workshops and interviews were audio-
10 recorded, and facilitators took field notes. Below we outline the focus of each workshop. The
11 individual workshop topic guides can be seen in Multimedia Appendix 2.



12
13

14 *Figure 1: The procedural flow and topics of the 8 design workshops*

15

16 **Workshop 1: Magic Machines[59] codesign activity**

17 This workshop introduced the series of workshops, and included discussion on some key self-
18 management issues participants experienced, and involved a Magic Machines[54] craft activity
19 where participants created some imaginative design solutions for another workshop participant. The
20 Magic Machines activity aimed to elicit a broad range of knowledge about participants' personal and
21 technological needs, through discussions about everyday problems relating to their condition and

1 potential solutions. Participants were asked to create an object, which addressed their partner's
2 daily challenge, out of household objects and craft items. Data capture was focused on the
3 conversations between participants about their "problem" while making their objects (a potential
4 "solution"), and when describing their object to the main group at the end of the session.

5 ***Workshop 2: Exploring daily lives***

6 The second workshop explored individuals' "daily lives" and the self-management of symptoms. The
7 daily lives discussion invited participants to discuss their 'typical day' in managing Sjögren's (i.e. their
8 habits and routines), how SS self-management was woven into these, and any related challenges
9 experienced.

10

11 ***Workshops 3, 4, 5, 6 and 7: Exploring Symptoms***

12 Workshops then explored self-management of specific symptoms and their interrelationships in
13 group discussions and invited participants to engage in basic sketching to articulate self-
14 management experiences and challenges. e We pre-selected the symptoms for discussion based on
15 our previous work where patients identified them as being important and impacting on their daily
16 activities[25].

17

18 ***Workshop 8: Consolidation***

19 Sketching was used to explore how an app might be structured to support symptom
20 interconnectedness and complexity. This design activity also elicited discussion around user
21 experience and usability issues. All participants engaged in sketching, but if short for time, they
22 were encouraged to further develop their ideas by articulating them verbally.

23 ***Interviews***

24 Three semi-structured online interviews were completed by xx following the workshops. The
25 interviews followed a schedule of open-ended questions to allow for flexibility (see Multimedia
26 Appendix 3).

27

1 **Data analysis**

2 Audio data was transcribed verbatim, pseudonymised, and combined into a corpus for analysis in N-
3 Vivo 12. Analysis was conducted in two phases using a hybrid approach, incorporating both
4 inductive and deductive methodologies, to harness the advantages of both[60]. First, an inductive
5 thematic analysis approach[61] was employed where two researchers (xx xxx) independently coded
6 the data generating an initial set of codes relating to participants' self-management perceptions and
7 experiences. These codes were then applied and refined through the arrival of each new transcript
8 and independent coding by (xx). Discussions at regular research team meetings relating to the codes,
9 their connections, importance, and relevance, were used to group codes into themes.

10 These inductive themes were then mapped to the three SDT[62] constructs of competency (the
11 sense of capability in performing activities/tasks), autonomy (experience of having control and
12 choice over one's actions/decisions), and relatedness (feeling of connection, belonging, and
13 meaningful interaction with others), by (xx). SDT was chosen over other motivational theories
14 because it emphasises social context as a key factor helping or hindering motivation, which matched
15 a prominent theme in our inductive thematic analysis on social relations, along with other major
16 themes we found relating to empowerment and autonomy, and capability (or "competency" in SDT).
17 The theory is also highly translational, enabling findings to inform intervention design[63]. Regular
18 research team meetings were used to review and reach consensus on the categorisations of themes
19 to the SDT constructs.. Opportunities to support participants' challenges associated with these
20 themes through an app were also arrived at through discussions. Methodological rigour and
21 credibility of findings were pursued through development of a codebook, maintenance of ongoing
22 reflexivity and peer debriefings, and data triangulation (from interviews, focus groups, and
23 observations during workshop activities).

24

25 **Results Participants**

26 In total, 17 people with SS took part in the workshops and interviews; 14 in the workshops (13
27 female, one male) and three undertook online interviews (all female). Participants ranged from 33-
28 76 years (mean 56.5 SD 13.95) and 14 had a diagnosis of primary SS. The remaining three
29 participants had a diagnosis of secondary SS. The mean number of years since diagnosis was 7.5 (SD
30 7.88). Regarding employment status, eight participants were retired, six were working full-time, one
31 was in part-time employment and two were not currently in work. All workshop participants had
32 links to a local SS support group in the north of England. One interviewee was part of the same

1 support group and the remaining two were from Spain and Canada (both <35 years). Workshop
2 group numbers ranged from 2-7 participants and 2-3 facilitators in each session. While some
3 participants came to some sessions and not others, a “core” group of five participants attended the
4 majority. One participant attended only the first workshop; others attended at least two.

5

6 **Overview**

7 Participants engaged in a wide range of self-management behaviours, including using prescribed and
8 over-the-counter medications and treatments (i.e., applying eye drops and gels; bathing and
9 massaging the eyes; use of humidifiers, skin creams, and vaginal lubricants; mouth care routines;
10 pain medication and hot and cold compresses). They also utilised cognitive and behavioural
11 techniques including activity pacing; goal setting; general exercise; relaxation; mindfulness;
12 distraction; napping; sleep management and wind-down routines; and social support. Participants
13 used various tools to support and facilitate the learning, use and practice of these techniques,
14 including books (e.g. on managing fatigue), diaries (paper and digital), websites and forums (e.g.
15 National Health Service or SS associations as both knowledge resources and social support), apps on
16 smartphones and tablets (such as for yoga, breathing exercises and mindfulness), wearables (to
17 track physical activity) and other devices (e.g. to provide relaxing music, “mindless” television, or
18 distracting podcasts and comedy). Not all participants owned or used smartphones. Tools were used
19 in addition to visiting friends and holistic wellness centres (e.g., spas, mindfulness classes) and
20 learning self-care techniques directly from health-care professionals (e.g., when to take eye drops,
21 and more complex techniques such as activity pacing and graded exercise).

22

23 In the following sections, we describe the challenges participants faced in managing their condition
24 and their psychosocial needs in terms of competency, autonomy, and relatedness.

25

26 **Competency**

27 Participants varied in the extent to which they felt competent and successful in self-managing their
28 SS, and this was related to how well they had established a self-care routine. One participant had a
29 very “*strict regime*”, which she felt was required to maintain her level of functioning. While hearing
30 about such self-management strategies from others, Jim reflected on his competencies:

31

1 *I've still got quite a lot to learn... although it has been a few years now, I think I still haven't*
2 *got a good routine... I listen to your explanation [of another participant's routine] and I think,*
3 *why can't I get myself like that? I'm supposed to be Mr Organised. I am known as that in my*
4 *life. My working life and my own home life. Yet with this, I have not gotten organised yet.*

5 [Jim]

6

7 Regardless of whether participants had routines or described habitual self-management behaviours,
8 their sense of competency in self-managing SS was still impeded by the complex nature of their
9 symptoms. Isolating and targeting individual symptoms was not only perceived to be difficult to do -
10 *"you can't separate the different symptoms"* [Jim] - but also sometimes unhelpful, as it did not
11 account for their accumulative negative impact:

12

13 *It is the overall effect to me. That three [symptoms] I can cope with and then the next day*
14 *one raises its head and floors me... That straw that broke the camel's back effect, you know.*

15 [Patricia]

16

17 Several participants believed they could better manage their symptoms through self-management
18 techniques capable of improving multiple symptoms simultaneously. Some had discovered these
19 types of techniques accidentally. For example, two recounted noticing, with surprise, that eye drops
20 had helped not only their dryness but also mental and physical fatigue. Other participants
21 purposefully sought out and regularly used techniques that targeted multiple symptoms at once.
22 Mindfulness and relaxation techniques provided a sense of control and the ability to *"keep a cap on"*
23 multiple symptoms before they became too severe. Others agreed that seeking these techniques
24 was worthwhile if it meant minor improvements across multiple symptoms. Despite valuing self-
25 management techniques that targeted multiple symptoms, most did not feel confident or
26 knowledgeable about which techniques were beneficial.

27

28 A further challenge to participants' sense of competency is how SS symptoms are not static but
29 change over time. Participants described instances where individual symptoms would rapidly
30 fluctuate in severity, for example: *"[They] come and go... one day you might have a headache, the*
31 *next day you don't"* [Jim]. Participants also explained experiencing longer periods where multiple
32 symptoms were severe (described using phrases such as a *"flare"*, *"phase"* or *"wave"*) or individual

1 symptoms persisted (such *“a dry patch”*). While sometimes the onset of symptoms appeared
2 *“gradually”*, at other times they changed rapidly, leaving participants feeling unprepared (*“a phase*
3 *hits you”*). Fatigue and pain were felt to be particularly volatile and could become severe with no
4 warning and *“like somebody just switched a switch”* [Penny].

5

6 Participants varied in how they managed such changing symptoms. Many attended to symptoms as
7 they arose or increased in severity on a moment-to-moment basis (i.e. an adaptive or reactive
8 approach). However, this often meant devising complex and intricate strategies and sequences to
9 manage the new combination of symptoms experienced in that moment. For example, sleep
10 disturbances which might be attributed to pain, dryness or anxiety required participants to vary their
11 approach to getting back to sleep, accordingly - *“depending on how I am”* [Penny] or *“what problem*
12 *I am having”* [Jim]. Other participants seemed to disregard the changing combination of symptoms
13 and addressed symptoms *“one at a time”* based on whether they felt successfully managed. For
14 example, Julie noted that *“I tend to find like I feel like my feet are sorted, so I am now sorting my*
15 *eyes, so I’m kind of going through this list”*. Addressing symptoms required constant adjustments for
16 participants. Their variable nature meant that just at the point that they started to feel in control of
17 one symptom, a flare of another may occur.

18

19 A final layer of complexity impacting participant competency was how symptoms often change due
20 to environmental factors. Dryness for example was exacerbated by air conditioning, bright lighting,
21 and other people’s aftershaves and perfumes, while navigating new and busy places could
22 exacerbate mental fatigue. The unpredictable nature of environments outside the home-made self-
23 managing symptoms more challenging. While home was characterised as *“familiar”* and
24 *“unchanging”*, participants felt they needed to continuously estimate the potential impact of
25 environments on their symptoms and plan accordingly:

26

27 *You have to be very wary of where you’re going... you’ve got to be careful. I will not walk*
28 *through [the shopping mall] in the perfumery because there is always somebody going to...*
29 *pick up a bottle of perfume and [spray]... I go, oh my eyes! [Geraldine]*

30

1 This planning itself was exhausting to several participants, and it also meant they lacked spontaneity
2 in their lives. Participants also felt symptoms were easier to manage at home because they could
3 easily perform physical relaxation and self-care techniques when required, particularly during a flare.
4 During such times Sarah remarked:

5

6 *I just don't want to leave the house, I don't want to do anything. I just want to go and have*
7 *like 2-3 baths per day. [Sarah]*

8

9 In contrast, when symptoms left them debilitated outside the house, participants had to adopt
10 different self-management techniques such as soothing self-talk, or organised being “rescued” by a
11 taxi or friend. Overall, being outside of the home meant that participants were less in control of
12 their environments, had to continuously plan and predict how these might impact their symptoms
13 which was mentally and physically tiring, and use different techniques to suit different
14 environments.

15

16 **Autonomy**

17 Our analysis identified many examples of participants feeling they had autonomy in the self-
18 management of their condition, although at times the same things that promoted autonomy also
19 reduced confidence and competence. Participants believed that the availability of various
20 techniques meant they had options in their self-management; there were multiple “different ways”
21 they could try to improve symptoms. The plurality of techniques appeared to provide reassurance
22 that at least one would be likely to be effective:

23

24 *I have six choices... I don't beat myself up when it doesn't work because I've already got*
25 *something else in mind. [Patricia]*

26

27 This plurality and optimism could provide a strong drive to continue in their self-management
28 activities.

29

1 Individuals varied in how they kept track of different available techniques. One had self-help books
2 in various locations in her home. Another explained that she had collated several techniques to
3 create her own book:

4

5 *I wrote myself a little book... [of] top tips... I just wrote maybe two dozen messages across*
6 *the book at random, things that might give me a clue. [Debra]*

7

8 Other participants employed an experimental approach – *“It’s about learning... through trial and*
9 *error... you’ll notice a pattern... you don’t know until you’ve done it for a few months”*
10 [Michelle]. These were similarly characterised by the desire to try different techniques, but also to
11 keep track of their effectiveness: *“With time and experience you begin to realise what works and*
12 *what doesn’t”* [Penny]. It was acknowledged that this required continuous effort and perseverance:

13

14 Having personal choice to decide which techniques to try, as opposed to being directed by a
15 healthcare professional, provided some participants with a sense of control. Debra likened creating
16 her book of techniques to developing a tailored smartphone app.

17

18 *“It is basically my own app that I’ve written for myself... I didn’t feel like being ordered*
19 *around by anybody else... I don’t necessarily follow it. If it’s inside my book, I think, well*
20 *alright, maybe I’ll try something else...I’ve still got some kind of control over things.” [Debra]*

21

22 Therefore, developing this herself meant she did not feel obligated to try any one technique.
23 Although participants appreciated having the autonomy to choose techniques in a personalised way,
24 the credibility of these techniques was also very important to them. Perceived credibility seemed to
25 give them confidence to go ahead and try them. Some participants indicated they understood the
26 distinction between evidence-based information and hearsay.

27

28 *I am pretty much someone who will try anything once if there’s some evidence to support its*
29 *effectiveness... Some people suggest real outlandish things, like you hear it and you’re like,*

1 *“okay!” I mean, I’m glad that it works for you, but I’m not really sold on trying that just yet.*

2 [Ellie]

3

4 Participants appreciated information they felt was credible. For example, Jim explained that simply
5 being presented with multiple self-management techniques and options, without a rationale for why
6 they might be helpful, would not sit well with him. Others stated that knowing information sources
7 was *“useful ... [for] controlling symptoms and trying to minimise [them]”* [Edith]. Information from
8 websites such as the UK National Health Service or regional and national SS organisations was
9 deemed trustworthy.

10

11 Although participants respected expert advice and implemented it in their self-management, expert
12 authority was often only one element in an autonomous process of symptom management decision-
13 making. For instance, when faced with a conflict between their preferred routines and expert advice
14 participants trusted their own expertise and experience. Jim outlined how he fell asleep with the
15 help of music or old comedy shows, and that he would simply *“ignore”* any potential prompts about
16 adjusting his bedtime routine if it meant removing his music from the bed (as may be advised as part
17 of a sleep intervention).

18

19 For some of the participants, smartphones and associated apps appeared to contribute to feelings of
20 autonomy around their self-management of SS. Those who used a smartphone reported using basic
21 note applications to track symptoms, or calendars built-in to the operating system to track feelings
22 of fatigue. Experienced smartphone users described how their ubiquitous nature enabled quick
23 access to information and could give access to techniques whenever and wherever needed
24 regardless of their location. In sessions where feedback was given on potential app designs,
25 participants expressed the value they would see in new apps that brought various techniques
26 together, provided reminders to take eye drops, and helped track symptoms more simply. For
27 instance, Julie suggested that *“a tracker or a journal ... or something like that on the app would be*
28 *helpful”* as this could help her manage her forgetfulness which she put down to *“brain fog”*.

29

30 However, while smartphones could enable autonomy, they also posed challenges that could impact
31 on their SS symptoms. Many noted that looking at the screen of a computer or phone for too long

1 exacerbated their eye dryness: *"It's okay [when it's] short, but you can't spend a long time looking at*
2 *the screen, because your eyes are just too sore"* [Mel]. To overcome this, participants used their
3 phones differently. Some described deliberately limiting the amount of time they used them in one
4 session and others described changing their device settings to increase the font size or darken
5 screens. As well as improving on-screen accessibility to reduce their eye strain, participants reported
6 listening to audio instead of reading text. One participant, Patricia, noted that when *"I am having my*
7 *brain fog"* the complexity of most apps *"would blow my mind"*. There was a sense amongst several
8 of the participants that smartphone use was closely related to experiences of mental fatigue from
9 their SS.

10

11 Overall, participants valued the diversity of SS self-management techniques that are available and
12 experienced this as enhancing autonomy. Smartphones and both generic and SS-specific apps were
13 viewed as an important part of this diversity and could provide in-situ tailored support. However, the
14 apparent abundance of techniques and availability of phones also posed a challenge to autonomy.
15 Patricia recounted that soon after being diagnosed with SS, she had felt overcome by the need to
16 learn about multiple symptoms and techniques from many sources. However, for her, this felt like
17 being *"shot at"* from multiple angles. What enables autonomy, can also, at times, constrain it.

18

19 **Relatedness**

20 Relatedness refers to the way participants operated in their social worlds and how their practices of
21 managing SS were enmeshed in this. Participants explained that SS profoundly impacted their
22 familial interactions, friendships, and other forms of social contact. Participants enjoyed managed
23 social activities and cherished positive relationships as a source of social support. Socialising and
24 taking part in activities with others provided a positive *"distraction"* from their symptoms. However,
25 self-management tasks could impact their ability to socialise and interrupt the flow of conversations:
26 *"When in company if you are out and about and talking to people... You have to keep popping off to*
27 *go and put eyedrops in, in the loo"* [Edith]. Furthermore, engaging in certain social activities such as
28 going to the cinema with friends required participants to perform additional self-care, for example
29 putting in eye drops more frequently, which could irritate the skin around the eyes. Geraldine
30 explained that although she enjoyed going to the theatre, she was now reluctant to go based on
31 previously being *"crucified"* by a smoke machine.

32

1 Pacing was a helpful technique when it came to staying on top of fatigue, but it was not always
2 received well by others in social situations and workplaces. Patricia recounted that she had been
3 regarded as “selfish” by family and friends for cancelling plans whilst trying to manage her energy
4 and fatigue levels. She also recognized that having to “book” people into her diary well in advance to
5 support her planning and pacing efforts “frightens some people off”. Edith recalled that the need to
6 take more breaks meant that she had to decide to leave her walking group as she was no longer able
7 to keep up with her friends. This in turn negatively impacted her feeling of belonging.

8

9 Communication was key when it came to managing illness demands and relationships. Some
10 participants created their own SS information sheets to hand to friends and healthcare
11 professionals. Creating opportunities to explain difficulties was conducive to receiving valuable social
12 support. Penny’s husband had delegated several household tasks to her, conflicting with her pacing
13 technique. Penny explained that after discussing the issue with him, he subsequently understood the
14 need to balance activities, and that they were able to do this together. Ellie noted:

15 *I do think that it is helpful to have people that you can talk to about Sjögren’s. I mean I have*
16 *a very close relationship with my family, and I have close friends who I do feel like I can*
17 *confide in, and that is really helpful for me. [Ellie]*

18

19 The freedom and ability to be open and honest about their SS symptoms with trustworthy family
20 and friends were central to well-being and helped symptom self-management. However, despite all
21 efforts to communicate effectively, many participants believed that often family and friends, and
22 even healthcare professionals, did not fully understand SS. They felt frustrated that symptoms were
23 dismissed, normalised, or wrongly attributed to other issues like “getting old” or menopause.
24 Dealing with invisible, ever-changing symptoms did not help. Multiple GP visits with complaints of
25 seemingly benign symptoms such as fatigue and thirst were sometimes received with perceived
26 scepticism, and the transient nature of these symptoms only made things worse: “Then you’re fine
27 and you think, ‘they’ll think I am putting it on’”. [Geraldine]. Any respite from symptoms had some
28 participants worried that those around them would not believe them the rest of the time. Carol
29 knew that relative to other conditions that may have one visible “major” symptom, her multiple
30 symptoms were unlikely to garner support and understanding, because of “Sjögren’s [and] all the
31 little things that it has” [Carol]. Some participants had stopped attempting to explain symptoms to
32 family and friends, saying that some symptoms were “very difficult for you to articulate... to

1 *somebody who doesn't feel it*" [Joan]. This was particularly detrimental to relationships with
2 healthcare professionals. When healthcare professionals seemed uncompassionate about their
3 symptoms, some participants talked about "*shutting down*" and making a choice to no longer discuss
4 their SS in consultations. This had negative consequences on participants who ended up feeling
5 rejected and disengaged, and there was a perception that at times healthcare practitioners were not
6 even aware of this relational and motivational shift.

7

8 When participants felt disbelieved, it led to experiences of self-doubt. Ellie said she was "*bounced*
9 *around like 4-5 practitioners*" to the point where she questioned her illness "*almost as if it is in your*
10 *head*". Carol resorted to keeping an activity diary, in part to monitor her fatigue, but also to preserve
11 her sanity. For her, the diary data provided a sense of external objectivity and an opportunity to feel
12 validated in the face of being questioned by other people:

13

14 *By doing the [diary] you think, yes... I've got a problem and that graph tells me... it is a*
15 *physical thing, it's not in my mind. [Carol].*

16

17 Being diagnosed with SS was a lonely experience for some participants due to the challenges of
18 family, friends, colleagues and health professionals not relating to participants symptoms or
19 condition. Social isolation was particularly pronounced for one younger person with SS:

20

21 *I don't know anyone else who has it. So, it is kind of isolating... I also had a hard time finding*
22 *people who are ... my age. So, I mean, I would definitely be interested in meeting younger*
23 *women who are working, who are finding strategies. [Ellie]*

24

25 Overall, connecting with others with SS was important and participants sought opportunities to
26 meet others with SS, to learn, and find the validation and understanding they did not necessarily
27 receive from others without SS. Some became support group members, and attended scientific
28 conferences to expand their social circle with other people with SS.

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30 However, not all social contact with others with SS was deemed to be helpful:

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Some of the interactions I had honestly more scared me than helped me because it was people who were really in the throes of severe illness and some who weren't coping well, and it was sort of anxiety-provoking. [Ellie]

Support from others with SS was therefore generally more welcome when it was helpful and positive as interactions with those who were struggling to cope could impact negatively on participants.

Within the construct of relatedness, even positive self-management was found to impact social activity but having highly supportive friends and family could mitigate this to some extent.

Describing and explaining the various, ever-changing symptoms to colleagues, friends and health professionals who did not fully appreciate the condition or symptoms, could be particularly challenging, but external resources such as using diary data could be a helpful tool to aid communication.

Discussion

We sought to understand current self-management approaches used by people with SS to inform the therapeutic ingredients and design recommendations of a self-management smartphone intervention. To date, most studies of lived experience in SS have focused on how specific symptoms are experienced[17, 18, 48]. To our knowledge, there are no studies exploring how people with SS do the day-to-day work of managing their condition and navigating challenges as they do so. This is an important consideration when designing interventions as those which draw upon users' expertise are more likely to be used[38]. Therefore, we analysed qualitative data collated through a series of workshops and interviews with people with SS inductively, before mapping the themes to the three constructs of SDT (competency, autonomy and relatedness)[52]. This theory was used as it can help identify psychosocial and practical needs required to support autonomous motivation to change and sustain healthy behaviours and to improve well-being in a population[52, 64]. Our work with the participants brought to light what Cartner first described in her qualitative research with SS participants: the *labour* of living with SS[65]. For her and our participants, competency was an ongoing effort, never a completed achievement. The complex, multi-symptomatic, volatile and unpredictable nature of the condition meant that their hard-won expertise was being constantly challenged. Having to adapt to an ever-changing and unpredictable challenge evokes the concept of stress, but more specifically it is captured by the notion of *allostasis*: the work that needs to be

1 done to find stability within a situation that is constantly changing. When allostasis is frequent or
2 continuous, more work needs to be done and our emotional, cognitive and biological resources can
3 become dysregulated. This is known as *allostatic load*, the psychophysiological wear and tear that
4 happens to a system that is constantly having to adapt and which has clear links to anxiety,
5 depression, morbidity and mortality[66].

6

7 The labour of the participants and its costs were also evident in the SDT domain of autonomy. There
8 was often a degree of forced autonomy, with participants having to do the epistemic labour of
9 figuring out how to manage their condition for themselves. This involved ongoing research and even
10 compiling their own resources. Discernment and discrimination were required to figure out what
11 advice to trust and follow, and how to balance that against their own experience. And although this
12 process was enabling, it was also potentially disabling as the very process of gathering and compiling
13 information worsened some SS symptoms.

14

15 Finally, in the realm of relationships, managing SS brings significant social labour. Participants were
16 often having to manage the expectations, lack of understanding, scepticism, and disbelief of others,
17 including health professionals, and these efforts were often only partially successful, if at all, leading
18 to self-doubt, isolation and lack of adequate care and support for their illness. This is not dissimilar
19 to the experiences others face with other fatiguing LTCs such as stroke, fibromyalgia, multiple
20 sclerosis and ankylosing spondylitis[67].

21

22 **Design recommendations**

23 Table 1 summarises our key findings, which have been mapped onto the three constructs of SDT,
24 with identified therapeutic approaches and design solutions for each. The findings within these SDT
25 domains were identified as targets for intervention by the participants. In the following, we will
26 review these domains and suggest what interventions might help, and how these interventions
27 could be adapted into an app to support self-management.

28

29 **Table 1: Key findings with potential therapeutic and design recommendations mapped onto Self-** 30 **Determination Theory domains**

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Self Determination Theory construct*	Current self-management practices and preferences	Self-management challenge	Therapeutic approaches and solutions	Potential design solutions
<p>Competency</p>	<p>To manage several connected symptoms, there is a preference for techniques that target more than one symptom at a time, where possible.</p> <p>Participants would like to know if they address a symptom individually, whether there is potential for a ‘knock-on’ positive impact on other symptoms.</p>	<p>Lack of knowledge of techniques that can target multiple symptoms.</p>	<p>Tips and tricks taken from multiple people with SS.</p> <p>Emphasise that no one size will fit all.</p> <p>Encourage users to experiment and trust their own experience.</p> <p>Prioritise the inclusion of evidence-based self-management techniques targeting more than one symptom, when possible.</p> <p>Provide a clear rationale for the inclusion of techniques and be explicit in how they aim to help with different symptoms or symptom combinations.</p> <p>Include self-help techniques that target individual symptoms, where there is no symptom cross-over.</p>	<p>Focus the intervention narrative/user flow on techniques that target more than one symptom simultaneously, where possible. Use these core techniques to organise the intervention (i.e. a ‘technique-led’ information architecture, where e.g. the main tabs in an app are techniques).</p> <p>Feature a rationale before each technique.</p>
	<p>To manage flares and rapidly changing symptoms, some adapted their strategies on a moment-to-moment basis and to prioritise the most severe symptom.</p> <p>Others reported disregarding their current state and persisting with self-management strategies for individual symptoms until they felt sufficiently managed in the long term.</p>	<p>Symptoms are volatile and difficult to control or influence.</p> <p>Participants rarely believed that they had sufficiently addressed or managed one symptom before ‘moving on’ to the next.</p>	<p>Acknowledge the volatility and difficulty of living with changing symptoms within text/content.</p> <p>Encourage self-compassion and acceptance of work and costs of dealing with the above (e.g. using Acceptance and Commitment Therapy[68] informed content).</p>	<p>Use design strategies to improve the sense of self-efficacy/sense of control/capability.</p> <p>Provide simple tools for the tracking of therapeutic techniques used, perceived self-efficacy in enacting them, and associated symptom effects.</p> <p>Design in community features that support the sharing of experiences</p>

			<p>Encourage users to identify valued activities that can be done with symptoms.</p> <p>Content to enhance self-efficacy and self-belief. Potential therapeutic techniques to support this may include the following: Graded activity, encouraging beliefs about capabilities, clear statement of “you are not alone”, problem-solving and goal setting.</p>	specifically associated with the management of multiple symptoms.
	<p>Participants reported needing to regularly assess the impact of the environment on their symptoms every time it changed (e.g. the air quality), particularly in new situations. They then needed a strategy to minimise the impact of these environmental fluctuations.</p>	<p>Planning how to respond to various possibilities can be exhausting and cognitively taxing.</p>	<p>Include an action planning tool that support users in breaking plans down into smaller tasks with flexibility for adjustments as required.</p>	<p>Design an activity planning tool with a simplified design to reduce overwhelm.</p> <p>Integrate features that use open-source air / environmental quality datasets to help users plan activities outside the home, and community tools to highlight potential “hotspots” for problematic spaces.</p>
Autonomy	<p>Having a multitude of self-management techniques to choose from provided a sense of optimism (there are others left to try).</p>	<p>Having numerous potential self-management techniques to choose from was felt to be overwhelming.</p>	<p>Provide users with a streamlined toolkit of relevant and appropriate therapeutic techniques. Provide a suggested order to try these techniques (if this is warranted therapeutically), while not restricting the user’s autonomy to choose or prioritise.</p> <p>Promote the use of experimentation to try one technique at a time.</p>	<p>Simplify the interface/reduce overwhelm through designing for simplicity and minimalism. One option is to present techniques one at a time, while still allowing users to override the order (avoid funnelling or a “tunnelled” information architecture).</p> <p>Help user to track what has worked for them and discard, mute, deprioritise those that have not or is not relevant to them.</p>
	<p>Some valued independence/control over choosing self-management techniques.</p>	<p>No challenge was identified.</p>	<p>Provide a rationale for techniques, ensure techniques are evidence-based, list credible sources,</p>	<p>Need for <i>optional</i> tailoring and guidance on techniques to try. Design these to be recommendations or options, which users can either ‘turn</p>

	<p>Others wanted guidance on which techniques to use. independent decision-making was supported by understanding the rationale for using a particular technique and the credibility of information.</p>		<p>and are kept up to date.</p> <p>Provide guidance on suggested techniques for those who require it.</p>	<p>off' or select.</p> <p>Use design strategies that enhance a sense of credibility, e.g. by providing links or references to credible sources that informed the content, using logos from sources (e.g., NHS) as appropriate.</p> <p>Provide community features that highlight the use of techniques by other users and perceived self-efficacy.</p>
	<p>Some used apps to self-manage and had created paper-based systems that they likened to an app.</p> <p>Participants reported several benefits of a proposed SS-focussed app (ubiquity/ability to learn about techniques anywhere), and the collated easy-to-find information rather than disparate sources, potential to remind them of new techniques to try and demonstrate how to do them, or time/context-specific reminders for when to use particular techniques (e.g. apply eye drops).</p> <p>Beyond apps, tactile features of wearables were appreciated, and audio features alongside text could potentially reduce the impact of eye symptoms.</p>	<p>Looking at the screen of a computer or phone for too long could exacerbate eye dryness and mental fatigue</p>	<p>Reminders to limit the length of sessions on the phone and take breaks from using technology.</p>	<p>Present information in a 'bite-sized' manner (not multiple menus/pages of text).</p> <p>Design to be compatible with accessibility options users already implement, including increased font size, ability to darken screens, the option to listen to an audio recording of the in-app text, and use tactile vibrations to convey information.</p> <p>Design apps that work across an ecosystem of connected devices – e.g., smart watches for reminders, and consider other forms of multi-modal interaction to reduce screen time (e.g., emphasise audio and vibration-based reminders).</p>
Relatedness	<p>Participants felt socialising with others provided a positive "distraction" from symptoms.</p>	<p>Symptoms and effective self-management (e.g. pacing and prioritising) limited participants from socialising and doing activities with others.</p>	<p>Plan, prioritise and pace social contact, as part of an activity management strategy. Treat social activities similarly to other activities and encourage users to have a balance of different types of activities within their week.</p>	<p>Include reminders for users to enter social events in a planning tool.</p>

			Get people on their side to act as “ambassadors” to decrease the communication burden.	
	<p>Explaining symptoms and self-management strategies to other people sometimes resulted in them being more understanding and accommodating.</p> <p>Some participants found ways to self-manage and structure their days that enabled them to continue their social life.</p> <p>Carrying a paper document around that explained the condition and symptoms was a useful way of communicating with others including healthcare professionals.</p> <p>Some people used completed activity diaries to demonstrate the impact of their SS on their daily lives to others.</p>	<p>Users may struggle to communicate their condition/symptoms/needs and therapeutic decisions to others, which impacts on their relationships and their ability to self-manage well.</p> <p>Explaining the condition and its symptoms to others takes effort. Other people sometimes seem to lack understanding of the invisible condition which can lead to feelings of frustration and self-doubt for the person with SS.</p>	<p>Provide assertiveness techniques to users, with which they can convey to others their needs without undue anxiety.</p> <p>Provide access to credible sources of information that can be used to evidence the credibility and validity of their symptoms when these are challenged.</p> <p>For those who struggle to communicate their needs - provide a written customisable template for communicating with others (particularly in new settings).</p>	<p>Include examples and exercises that train users in assertiveness and communication techniques.</p> <p>Include text such as pre-prepared communication cards for various audiences (employers, HCPs, work colleagues, etc.) to aid the story ability of the condition. This should speak on behalf of the user to others to succinctly explain the condition, symptoms and its impact.</p> <p>Include visual summaries of completed activity diaries to easily convey information to the user and others.</p> <p>Consider applications that are designed for spouses and family carers to support shared understandings of the conditions, and to distribute the social labour of advocating for oneself to friends and relations.</p>
	<p>Several described wanting to meet others with SS who understood what it was like to live with. Many accessed or wanted to access an SS forum.</p> <p>Speaking with role models could provide comfort.</p>	<p>Not knowing anyone like them with SS felt isolating and it did not feel easy to identify with celebrities who had SS.</p> <p>However, others who have SS could report negative experiences which may result in increased anxiety or frustration.</p> <p>Going outside the home required planning for any potential environmental impact on symptoms; getting out to meet others with SS could be difficult.</p>	<p>Encourage through the use of others’ stories.</p> <p>Make it clear that each has their path, and everyone struggles at times.</p>	<p>Include examples and quotes from real people with SS. Use validating language.</p> <p>Use persuasive language to facilitate self-efficacy.</p> <p>Include a feature that asks users to recall an incidence of overcoming a difficult issue.</p> <p>Provide community features that connect people with SS via simple asynchronous and synchronous (live, real-time) chat and audio conversations.</p> <p>Provide guidance on setting up virtual social visits for people with SS.</p>

1 *According to Self-Determination Theory (Deci & Ryan 2008)

1 A key finding within the competency domain was that SS was multi-symptomatic, volatile and
2 unpredictable. Participants were keen for interventions that would impact more than one symptom
3 at a time. A previous study investigating patient strategies in self-management of inflammatory
4 bowel disease (IBD) had similar findings[69]. Several treatment approaches and their components
5 discussed during the workshops could potentially address several symptoms together. For example,
6 activity and sleep management strategies such as pacing and reflective activity diaries have been
7 used to support self-management of pain, sleep disturbances and fatigue[70-74] and previous
8 studies evaluating interventions that target several symptoms have shown promising results. We,
9 therefore, suggest that when designing complex interventions for long-term conditions, intervention
10 developers map potential identified intervention content to behaviours and/or symptoms and select
11 techniques that target more than one where possible, thereby placing a smaller demand on the
12 user. Whilst this may not always be possible, streamlining intervention content where practicable, is
13 likely to decrease overwhelm and thereby support user competency.

14

15 The key challenge in the autonomy domain was the amount of work required by participants to
16 figure out how to manage their condition on their own. As with many other LTCs, a large part of the
17 “burden of treatment” is shouldered by the person with the condition[75]. Our findings broadly
18 indicate that technology-enabled symptom management could help with this work of illness
19 management. Participants liked the idea of a smartphone app to support self-management.
20 However, merely operationalising technology is not enough to promote and support self-
21 management. Güldenpfennig and colleagues found that poor design and well-meaning paternalism
22 for example through automated support which takes active choice away from the user, may
23 compromise autonomy and proactive self-management[76]. Furthermore, intervention designers
24 should aim to strike a more careful balance between the input of experts by experience and
25 professional experts[25, 76]. In our study, we found that people with SS managed their symptoms
26 using different approaches, but that all of them had arrived at their own set of strategies and
27 management regimes through experience, research, and trial and error. Acknowledging the
28 individuality of self-management, and the necessity to experiment with different approaches would
29 be a key part of any intervention. Having a repository of strategies in one centralised app, which
30 would also allow them to add their own, would seem like a potentially useful resource. This aligns
31 with previous research on apps that provide resources whilst allowing customisation and thus may
32 support a user’s sense of autonomy[77-80] and move away from a top-down
33 paternalistic/prescriptive approach to LTC management[81, 82]. An app for SS would need to
34 combine recognition of the labour of self-management whilst helping to support it in a way that

1 honours their autonomy and existing wisdom, providing ability to choose from a range of
2 therapeutic content and to determine the order in which they interact with it.

3

4 The most difficult and often fruitless area of labour fell within the relatedness domain. Participants
5 were having to manage others' expectations, lack of understanding, scepticism and disbelief often
6 leading to a smaller social world, isolation and difficulties accessing help from health professionals.
7 Again, any intervention needs to begin by acknowledging this labour and the emotional and social
8 costs of having a poorly understood and non-visible illness. Our findings also showed that there was
9 often a tension between illness management and maintaining relationships. For example, it could
10 become difficult to implement strategies such as pacing when others were involved, particularly
11 when the person with SS had not fully disclosed their symptoms and/or condition to the others
12 whom their self-management strategy may affect. Therefore, saying "no" could also be hard for
13 participants, particularly when it was perceived that others would not understand. Other
14 participants had found a solution, by working on their means of communicating their difficulties with
15 those around them. Winger and colleagues have found that greater practice of assertiveness and
16 communication skills was associated with reduced pain interference and psychological distress in
17 people with lung cancer[83] and assertiveness and communication is also a key component of an
18 effective fatigue management intervention for people with rheumatoid arthritis[84]. We, therefore
19 recommend including assertiveness and communication strategies within a therapeutic self-
20 management app for SS. When considering the design of the app, we recommend including some
21 text to help the user provide a brief explanation of their condition, its symptoms and their impact to
22 share with health professionals, colleagues, or people in social settings as needed. We also
23 recommend designing opportunities to practice assertiveness and communication skills within the
24 app for those who may find this helpful.

25

26 In summary, our findings would suggest that some of the key areas of concern for participants were
27 potentially addressable through an intervention. A common starting point for any approach should
28 be an acknowledgment of the real costs and the daily hard work of having an unpredictable, volatile
29 and multi-symptomatic LTC. Any therapeutic approach needs to be framed as helping with this
30 labour, acknowledging the social, emotional and physical costs of having and managing SS, and to
31 appreciate the wisdom that the "end-user" of the app or intervention will have already accumulated.
32 Strategies taken from Acceptance and Commitment Therapy[85] and Compassion Focused
33 Therapy[86] could be useful as these have been used to target the psychosocial impact of other

1 related health complaints such as chronic pain[68]. After that, specific strategies (e.g. pacing, sleep
2 management) that could help target multiple symptoms or single symptoms in sequence would be
3 useful. Finally, support to do some of the social labour of living with SS should be a key component.
4 In Table 1 we further specify the areas of intervention and suggest what broad therapeutic
5 approaches might be useful.

6 In terms of our use of the SDT framework, whilst useful to structure our thinking about intervention
7 development we also noted that constructs of SDT often existed in a state of tension with each
8 other, where fulfilling the requirements of one construct successfully meant diminishing functioning
9 in another. As noted above, this tension occurred between competence and relatedness where
10 symptom management conflicted with maintaining social bonds. Similar tension existed between
11 autonomy and competence: participants struggled to feel competent if presented with too many
12 self-management options. The SDT maintains that all three fundamental needs have to be met for
13 internally motivated, self-determined behaviour to occur[52], but we tentatively suggest that the
14 theory needs to consider moments when some needs stand in opposition to each other on
15 occasions. Making explicit the nature of these tensions to users of an intervention/app would be a
16 key part of its opening narrative.

17

18 **Limitations**

19 The extent of transferability of our findings to other LTCs, is not yet known. However, studies of
20 other autoimmune conditions have nevertheless evidenced the same need to self-manage
21 complexity: people with IBD reported that symptoms (pain, fatigue and diarrhoea) changed over
22 time, could be interconnected at different times, and required a highly individualized management
23 strategy to “balance” the illness and attend to dynamic fluctuations in symptoms[69]. Overall, our
24 findings may provide insight into how several other autoimmune conditions are self-managed or
25 could be self-managed with the use of an app. However, by the very nature of the complexity we
26 captured in the present study, transferability to other contexts may be limited. Out of the 17
27 participants in our study, there was only one male, which may mean that any unique difficulties
28 experienced by males with SS have been missed in our study. However, SS has a female-to-male
29 incidence rate of 16:1[87] and the gender makeup of our participants is representative of the wider
30 SS population. Another limitation was that we did not formally collate information on smartphone
31 ownership from participants. Such data should be collated in future similar studies. A final limitation
32 is that most of this work was done within the UK, with only two interviewees living outside of the

1 UK. We therefore cannot assume that similar findings would be replicated in other geographical
2 contexts.

3

4 **Future work**

5 Future research should operationalise the present findings to construct an intervention protocol that
6 could be implemented via a smartphone app for the management of SS and empirically optimise its
7 content through pilot and feasibility testing. Furthermore, future studies may wish to explore the
8 transferability of our findings to the self-management contexts of other autoimmune and fluctuating
9 conditions. Our target users are those with primary or secondary SS; future work should consider
10 how user age influences design requirements in this patient group.

11

12 **Conclusions**

13 In conclusion, therapeutic and design approaches for SS should be constructed both bottom-up (i.e.,
14 based on the self-management challenges prospective users already experience) and top-down
15 (according to the most effective treatments documented for SS). For people with SS, choosing to
16 involve an app in their self-management even has the possibility of being counter-productive - by
17 adding to their experience of fatigue and overwhelm. The design of a self-management app for SS
18 should therefore support the user in doing the physical, cognitive, emotional and social work of self-
19 management and should be careful not to add to their already high self-management costs.

20

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