



## LJMU Research Online

Lattimore, P and Harrison, F

**Pilot Study of an Online-delivered Mindfulness Meditation in Ehlers-Danlos Syndrome (hEDS): Effect on Quality-of-Life and Participant Lived Experience**

<https://researchonline.ljmu.ac.uk/id/eprint/17933/>

### Article

**Citation** (please note it is advisable to refer to the publisher's version if you intend to cite from this work)

**Lattimore, P ORCID logoORCID: <https://orcid.org/0000-0001-8714-7678> and Harrison, F (2022) Pilot Study of an Online-delivered Mindfulness Meditation in Ehlers-Danlos Syndrome (hEDS): Effect on Quality-of-Life and Participant Lived Experience. Disability and Rehabilitation. ISSN 0963-8288**

LJMU has developed [LJMU Research Online](#) for users to access the research output of the University more effectively. Copyright © and Moral Rights for the papers on this site are retained by the individual authors and/or other copyright owners. Users may download and/or print one copy of any article(s) in LJMU Research Online to facilitate their private study or for non-commercial research. You may not engage in further distribution of the material or use it for any profit-making activities or any commercial gain.

The version presented here may differ from the published version or from the version of the record. Please see the repository URL above for details on accessing the published version and note that access may require a subscription.

For more information please contact [researchonline@ljmu.ac.uk](mailto:researchonline@ljmu.ac.uk)

<http://researchonline.ljmu.ac.uk/>



## Pilot study of an online-delivered mindfulness meditation in Ehlers-Danlos syndrome (hEDS): effect on quality-of-life and participant lived experience

Paul Lattimore & Fay Harrison

To cite this article: Paul Lattimore & Fay Harrison (2022): Pilot study of an online-delivered mindfulness meditation in Ehlers-Danlos syndrome (hEDS): effect on quality-of-life and participant lived experience, *Disability and Rehabilitation*, DOI: [10.1080/09638288.2022.2140843](https://doi.org/10.1080/09638288.2022.2140843)

To link to this article: <https://doi.org/10.1080/09638288.2022.2140843>



© 2022 The Author(s). Published by Informa UK Limited, trading as Taylor & Francis Group.



Published online: 03 Nov 2022.



Submit your article to this journal [↗](#)



Article views: 72



View related articles [↗](#)



View Crossmark data [↗](#)

ORIGINAL ARTICLE



# Pilot study of an online-delivered mindfulness meditation in Ehlers-Danlos syndrome (hEDS): effect on quality-of-life and participant lived experience

Paul Lattimore<sup>a</sup>  and Fay Harrison<sup>a,b</sup>

<sup>a</sup>School of Psychology, Faculty of Health, Liverpool John Moores University, Liverpool, UK; <sup>b</sup>The Ehlers-Danlos Support, Devonshire House, Borehamwood, UK

## ABSTRACT

**Purpose:** Ehlers-Danlos syndromes (EDS) are connective tissue disorders with multi-systemic symptoms. Management of chronic pain and other symptoms of EDS is a challenge for patients and clinicians. Mindfulness-based approaches for chronic pain produce improvement in pain symptoms. Mindfulness meditation could be an acceptable and readily accessible therapy for pain in EDS. This study evaluated the effect of daily practice of mindfulness meditation on pain experience and quality-of-life in EDS.

**Materials and methods:** A pre-post design enabled assessment of change in quality-of-life (SF-36) after practicing internet-delivered mindful meditation for two weeks. Thematic analysis of interviews ( $n = 10$ ) documented lived experience of meditation practice for pain symptoms. One hundred fifty-seven were recruited from UK EDS charities mailing lists. Seventy six completed the two-week intervention.

**Results:** There was a statistically significant improvement in the SF-36 mental component summary score of medium effect size; the change was clinically important. Thematic analysis revealed three key themes of lived experience relating to increased body awareness, reduction in pain intensity, and barriers to practicing meditation.

**Conclusions:** This study provides novel evidence about the effect and experience of meditation for symptoms in people with EDS. The outcomes warrant further research with appropriate control groups to determine efficacy.

## ARTICLE HISTORY

Received 10 February 2022

Revised 18 October 2022

Accepted 23 October 2022

## KEYWORDS

Ehlers-Danlos; pain; mindfulness; meditation; quality-of-life

## ► IMPLICATIONS FOR REHABILITATION



- Mindfulness-based interventions are effective for chronic pain relief.
- Hypermobile Ehlers-Danlos syndrome (hEDS) patients experience chronic pain that affects quality-of-life.
- Online-delivered mindfulness meditation was clinically significant in reducing pain intensity and improving quality-of-life outcomes.
- hEDS patients found the online-delivered intervention acceptable and recommended development to make it flexible to meet their specific needs by varying types and duration of meditations offered.

## Introduction

Ehlers-Danlos syndromes (EDS) are heterogeneous heritable connective tissue disorders characterized primarily by generalized joint hypermobility, skin hyperextensibility, tissue fragility, and chronic pain [1]. Classification includes Hypermobile Ehlers-Danlos syndrome (hEDS) and Hypermobility Spectrum Disorder (HSD; formerly joint hypermobile syndrome or JHS) [2]. The higher prevalence (3.4%) of association between JHS and widespread pain, a potential proxy for hEDS, suggests that hEDS is the most common systemic connective tissue disorder. Epidemiological studies estimate 2 million (UK), 10 million (USA), 17 million (EU), and 255 million worldwide affected by hEDS/JHS [3]. Long-term management of chronic pain in hEDS/JHS is difficult for both patients and clinicians [4]. Clinical trials have not determined efficacious therapeutic management of pain in hEDS/JHS [4–6]. Pain is not merely

a physical experience, it manifests in cognitive, behavioral, and emotional dimensions [7] that are interdependent and contingent on the physical pain. However, many patients never receive psychosocial treatment for chronic pain [8], which is concerning given the limited efficacy of medical/physical approaches in hEDS/JHS [9].

Cognitive-behavioral therapy (CBT) is a gold standard psychological intervention for management of chronic pain [8,9]. Systematic reviews and meta-analyses have established its efficacy for reducing pain, distress, and pain interference with activities. Integration of CBT into collaborative chronic pain care models is lacking [8,10]. Randomized controlled trials of CBT for hEDS/JHS do not exist. Mindfulness techniques have been incorporated in psychological therapy for chronic pain [11] and systematic reviews and meta-analyses indicate small effects for relief of pain symptoms [12]. Evidence for the differential efficacy of mindfulness and

CONTACT Paul Lattimore  [p.j.lattimore@ljmu.ac.uk](mailto:p.j.lattimore@ljmu.ac.uk)  School of Psychology, Faculty of Health, Tom Reilly Building, Liverpool John Moores University, Byrom Street, Liverpool L3 3AF, UK

© 2022 The Author(s). Published by Informa UK Limited, trading as Taylor & Francis Group.

This is an Open Access article distributed under the terms of the Creative Commons Attribution-NonCommercial-NoDerivatives License (<http://creativecommons.org/licenses/by-nc-nd/4.0/>), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited, and is not altered, transformed, or built upon in any way.

CBT interventions for chronic pain is inconclusive [10]. Evaluation of mindfulness interventions for pain or quality-of-life in hEDS/JHS have not occurred. Mindfulness is defined as “the awareness that emerges through paying attention on purpose, in the present moment, and non-judgmentally to the unfolding of experience moment by moment” [13]. Techniques include focused attention on the sensations of the breath. When attention wanders, the practitioner acknowledges the distracting event without engaging and gently re-directs attention to the sensations of the breath. The aim is to gain mental control and stability of attention which naturally leads to open monitoring of thoughts, sensations, and emotions so that they are experienced without evaluation [14]. Regarding pain experience, patients would learn to sustain attention on the breath and suspend judgment of thoughts and emotions that accompany pain experience. Even after one week of repeated practice, meditation produces significant changes in pain intensity and subjective ratings of unpleasantness. Longer term practice does not further reduce physical pain intensity but does influence the subjective dimension of unpleasantness of pain [15]. It appears that adept meditators can engage a difficult pain sensation without appraising or evaluating it, whereas novice meditators reduce pain by effortful reappraisal of the pain sensation [15]. This explanation implies that although meditation may not directly change physical pain, it can reduce its adverse impact on cognitive and emotional experience, and quality-of-life indicators. In this respect, it can facilitate a state of mind where it is possible to acknowledge pain without overidentifying with its sensory qualities or engaging in catastrophic thinking and furthermore reappraise the painful experience. The reappraisal process facilitates alleviation of secondary psychological “suffering” arising from the primary pain sensation [16].

Mindfulness meditation as an adjunctive therapy for hEDS/JHS could be acceptable to patients because it alters pain experience through multiple psychological and neural processes [15]. The current study evaluated the effect of mindfulness meditation on pain experience and quality-of-life in hEDS/JHS patients and documented their lived experience of using meditation. We predicted that practicing meditation daily would lead to improved quality-of-life and reduced physical pain.

## Materials and methods

### Study design

Accessibility to therapeutic interventions can be facilitated by remote access *via* the internet designed as alternative or adjunct to face-to-face care [17]. The current study used the internet to deliver instructions for hEDS/JHS patients to practice mindfulness meditation daily at home for two weeks. Interviews were conducted with a sample of participants to discover their experience of using the meditation. A single-group pre/post-test intervention assessed change in pain and quality-of-life over a two-week period.

### Recruitment of participants and sampling

Opportunity sampling recruited participants by email newsletters and social media channels of the UK Ehlers Danlos Support charity (EDS UK) and the UK Hypermobility Syndromes Association (HMSA). Inclusion criteria were diagnosis of EDS, HSD, or JHS; over 16 years of age, access to internet, fluent English speaker, and willingness to practice mindful meditation daily for two weeks. One hundred fifty-seven met the inclusion criteria, 81 dropped out after completing baseline assessments, and 76 completed the

intervention and all assessments (see Table 1). Participants who completed the intervention and opted for interviews ( $n=34$ ) were randomly selected to achieve a sample of 10 interviewees. The University Research Ethics Committee granted ethical approval and the authors adhered to the British Psychology Society Ethical Principles and Code of Conduct for Psychologists.

### Intervention

The mindfulness meditation exercise required participants to view a pre-recorded video explanation of mindfulness, its potential benefits, a description of the guided meditation, and directions about how to practice it daily. The guided meditation was adapted from the widely used mindful breathing space meditation [18] so that it lasted 6 min. The first author (PL accredited mindfulness teacher) produced pre-recorded video instructions and guided meditation MP3. The guided meditation comprised three elements followed in sequence: (1) *noticing*—acknowledging current experience, becoming aware of thoughts, feelings, and sensations. Bringing non-judgmental recognition and awareness to inner experience; (2) *gathering attention*—re-directing and narrowing attention to the ebb and flow of breath without changing anything. Anchoring self in the present moment. Re-attending to breath when attention wanders; and (3) *expanding*—Broadening attention to include the whole body. Notice sensation within the body without reacting or attempting to change. Expanding attention to external surroundings. Making intention to carry the experience of present moment awareness into daily activities.

### Data collection

#### Quality-of-life

The SF-36 healthy survey was used to assess physical and mental components of quality-of-life [19] that include: physical functioning, bodily pain, role functioning due to physical problems, role functioning due to emotional problems, emotional wellbeing, social functioning, energy/fatigue, and general health. Scale score computation used the t-score normalization method [20]. The reliability and validity of the SF-36 is acceptable [21]. Participants responded at pre-test considering the past four weeks, and at post-test considering the past two weeks.

#### Demographics and eligibility assessments

Multiple option and free-text questions assessed age, gender, ethnicity, diagnosis, source of diagnosis, date of diagnosis, duration of condition prior to, and since diagnosis, and treatment and support currently received from medical professionals and family (see Table 1). One question assessed willingness to meditate: “On a scale from 0 to 10, how willing are you to practice a guided meditation daily for two weeks using an MP3 download?”. Participants scoring below six were not eligible and excluded. Participants reported days per week and times per day spent meditating.

### Semi-structured interview

Interviews questions were developed to gain an understanding of satisfaction with, and feasibility of the mindfulness intervention. The interviewer (second author) volunteers part-time as a support worker by the EDS UK charity, has direct experience of hEDS, and at the time of data collection registered as an MSc candidate at first author’s institution. The interview comprised five main questions supplemented by probing questions where appropriate to gain richer context and clarify: (1) How easy was it to do

Table 1. Descriptive statistics of participants pre-intervention.

	Not completed <sup>a</sup> (N = 81)	Completed <sup>b</sup> (N = 76)	p
Age in years <i>M (SD)</i>	32.9 (10.8)	38.6 (12.5)	0.003
Willing to meditate <i>M (SD)</i>	9.6 (0.7)	9.6 (0.8)	0.915
Gender			0.347
Female	93%	98%	
Male	5%	1%	
Prefer not to say	2%	1%	
Ethnicity			0.042
Caucasian	83%	75%	
Mixed ethnic	6%	1%	
Black <sup>c</sup>	11%	24%	
Diagnosis			0.332
hEDS	62%	66%	
JHS	25%	16%	
HSD	14%	18%	
Source of diagnosis			0.615
GP	4%	5%	
NHS consultant	48%	55%	
Private consultant	25%	18%	
Physiotherapist	6%	3%	
Other <sup>d</sup>	17%	18%	
Time (years) since diagnosis <i>M/SD/range</i>	5.8/7.4/0.5–44	6.4/7.1/0.5–50	0.115
Duration of illness (years) <sup>e</sup> <i>M/SD/range</i>	21.7/13.1/1–60	23.7/15.4/3–65	0.115
Treatment			
Medication	75%	71%	
Surgery	12%	7%	
Genetic counselling	5%	5%	
Occupational therapy	24%	12%	
Physiotherapy	52%	53%	
Psychological	17%	16%	
Alternative treatments			
Yoga	24%	24%	
Massage	43%	36%	
Reflexology	4%	3%	
Acupuncture	20%	20%	
Reiki	1%	9%	
Meditation	26%	37%	
Herbal	20%	29%	
Quality-of-life/SF-36 <i>M (SD)</i>			
Physical functioning	38.9 (23.9)	31.3 (20.3)	0.34
Role limitations due to physical health	5.3 (12.9)	9.7 (19.7)	0.20
Role limitation due to emotional problems	27.2 (35.8)	30.0 (39.2)	0.89
Energy/fatigue	19.8 (11.1)	18.0 (15.5)	0.16
Emotional wellbeing	48.6 (19.2)	51.5 (18.7)	0.31
Social functioning	42.4 (21.3)	38.8 (24.7)	0.31
Pain	39.4 (15.3)	38.4 (16.5)	0.76
General health	22.3 (16.9)	22.9 (16.4)	0.76
Physical component summary	27.2 (7.2)	26.8 (7.7)	0.49
Mental component summary	36.3 (10.2)	35.8 (10.1)	0.58

<sup>a</sup>Not completed = participants who dropped out after completing pre-test measures.

<sup>b</sup>Completed = participants who completed pre-and post-test measures and meditation practice.

<sup>c</sup>Reported by rheumatologist, geneticist, cardiologist.

<sup>d</sup>Black/African/Caribbean/Black British.

<sup>e</sup>Duration of illness = participants were asked "How long have you been suffering with this condition (to the nearest year)?."

meditation (any obstacles you found)? (2) Did you notice any benefits during or after meditation? (3) What could be done to improve or strengthen the intervention? (4) How satisfied were you with the intervention?, and (5) Do you feel mindfulness may be useful for managing the symptoms of EDS?

### Procedure

The intervention and assessments were delivered online using the Qualtrics survey platform (<https://www.qualtrics.com/uk/>), thereby enabling automated delivery of assessments and instructions by email address collected at registration. Participants were invited

to register to check if they met inclusion criteria, and thereafter read a participant information sheet prior to consenting to take part. Eligible participants completed demographic questions and the SF-36 followed immediately by viewing a video of intervention instructions which triggered an email containing a link to the video and the MP3 guided meditation. Meditation practice frequency was assessed after 7 and 14 days. Fourteen days after starting, participants completed the SF-36 again followed by a debrief and an invitation to take part in an interview about their experiences of the intervention. Scheduled interviews were conducted by Zoom by the second author between May and June 2021. Recording of the Zoom meeting commenced after

participants made at ease, interview format explained, and consent obtained for recording and transcribing of interviews. Participants had opportunity to ask questions before recording commenced.

### Statistical analysis

The Shapiro-Wilk and z-test for skew and kurtosis ( $z \pm 2.58$ ,  $p < 0.01$ ) checked normality of data. Non-normality of all SF-36 scales at pre- and post-test was evident in terms of significant skew and kurtosis. Wilcoxon ranked sum distribution free test analyzed change in SF-36 scales. Effect sizes were calculated as  $r = z/\sqrt{N}$ . Effect sizes were assessed according to Cohen's classification of effect sizes: 0.1 = small effect, 0.3 = medium effect, and 0.5 and above = large effect [22]. Minimal clinically important difference (MCID) was calculated as  $0.5 \times SD$  of the difference between pre- and post-test SF-36 values [23]. Mean and standard deviation normalized t-scores are presented.

### Qualitative analysis

Interview transcripts were analyzed using inductive thematic analysis which allows identification of key themes from discourse [24,25]. Thematic analysis permits flexibility and opportunity to gain a broad overview of meaning from discourse. Transcripts were checked for accuracy against the audio recording and then uploaded to NVivo software. Coding of transcripts followed a recursive process of revisiting and revising the codes to identify and organize a set of themes and subthemes. The inductive approach to coding allowed for theme identification based on participant responses rather than predetermined interview questions. Short phrases that reflected common experience across participants summarized the themes that emerged from coding. Quotes from transcripts are reported to illustrate themes. Coding and thematic analysis was only completed by the second author.

## Results

### Descriptive statistics

Table 1 displays information about participants according to completion status. Eighty-one completed pre-test questions only and then dropped out. Seventy-six completed pre- and post-test assessments and engaged in meditation practice. Most participants were Caucasian females, though completers comprised

significantly more Black participants. The age of completers was significantly older than non-completers. Participants self-reported their diagnosis as predominantly hEDS and most received this diagnosis from an NHS consultant. Completers and non-completers did not differ in time since diagnosis or duration of illness. Approximately three quarters of all participants were using medication and half receiving physiotherapy. Participants used a range of alternative treatments, the most common being massage, yoga, acupuncture, and meditation. Differences between completers and non-completers on pre-test SF-36 dimensions were non-significant (all  $p > 0.16$ ; Mann-Whitney test).

### Adherence

To assess adherence to meditation practice, participants reported number of days they had successfully managed to practice meditation over 14 days. Sixty-five (85%) reported meditating between 9 and 14 days compared to 11 (15%) who reported meditating between 5 and 8 days. Sixty-two (82%) participants reported meditating two to three times per day, and 14 (18%) meditated five or more times per day. Meditation was practiced on average 11 days out of 14 ( $M = 11.2$ ,  $SD = 2.5$ ,  $MED = 12$ , range: 5–14 days) and 21 times over the course of 14 days ( $M = 20.7$ ,  $SD = 10.1$ ,  $MED = 18$ , range: 12–60 times). Using the duration of the meditation task (6 min) as a multiplier, the estimated accumulated average time participants spent meditating over 14 days was 4 h ( $M = 232$  min;  $SD = 123$  min;  $MED = 192$  min, range 60–600 min; computed as days\*times per day\*6 min).

### Pain and quality-of-life

Descriptive and inferential statistics for pre-post comparisons of completers quality-of-life scores are displayed in Table 2. Prior to practicing meditation, hEDS/JHS participants scored below the 50-point level on all SF-36 dimensions, except emotional wellbeing. There were significant increases in quality-of-life dimensions except for physical functioning. There was a significant increase in the mental component summary score but not the physical component summary score. Effect sizes were in the medium range. MCIDs range from 2 to 18. Participants remained below the 50-point level on all dimensions except for emotional wellbeing and pain.

Table 2. Descriptive statistics<sup>a</sup> of change in quality-of-life (SF-36) domains for participants who completed intervention ( $N = 76$ ).

SF-36 health survey <sup>b</sup>	UK norms <sup>c</sup>	Pre-intervention		Post-intervention		Wilcoxon test			MCID <sup>d</sup>
	<i>M</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>Z</i>	<i>p</i>	<i>r</i> <sup>e</sup>	
Physical functioning	89.8	31.3	20.3	31.5	21.2	−0.365	0.7149	−0.041	2
Role limitations due to physical health	85.3	9.7	19.7	17.0	27.6	−2.666	0.0077	−0.305	13
Role limitation due to emotional problems	83.8	30.0	39.2	45.4	42.7	−3.127	0.0018	−0.358	18
Energy/fatigue	60.5	18.0	15.5	24.8	19.0	−3.928	0.0001	−0.450	8
Emotional wellbeing	73.1	51.5	18.7	58.1	19.6	−3.746	0.0002	−0.429	7
Social functioning	88.6	38.8	24.7	44.7	25.4	−2.729	0.0063	−0.313	9
Pain	80.5	38.4	16.5	47.5	18.1	−3.965	0.0001	−0.454	9
General health	74.1	22.9	16.4	25.3	16.5	−3.010	0.0026	−0.345	4
Physical component summary	...	26.9	7.7	27.4	8.2	−1.116	0.2641	−0.128	3
Mental component summary	...	36.9	10.2	41.3	11.7	−3.878	0.0001	−0.444	4

<sup>a</sup>Descriptive statistics are computed using normalized t-score method [19].

<sup>b</sup>Pre-intervention, participants were asked to respond to questions considering past four weeks; post-intervention, they were asked to respond to questions considering the past two weeks.

<sup>c</sup>UK norms are average of Health Survey for England, Oxford Health Life Survey and British ONS [28]. Averages were computed for females 35–44 years old.

<sup>d</sup>MCID = minimal clinically important difference calculated as  $0.5 \times SD$  of difference between pre- and post-test values.

<sup>e</sup>Effect size calculated as  $r = z/\sqrt{N}$ .



**Table 3.** Themes uncovered using inductive thematic analysis.

Key themes	Subthemes
Increased body awareness through mindfulness	Enhanced attention
Reduction in pain intensity	Relaxation and sleep
Challenges and barriers to completing daily mindfulness meditation	Suggested improvements

### Feasibility and satisfaction

Ten participants completed interviews to explore their experience of the mindfulness intervention. Nine participants were diagnosed as hEDS and one as HSD. Participant age (38.9 years) and duration of illness (23.4 years) were equivalent to the full cohort of completers (38.6 years and 23.7 years, respectively). Time since diagnosis (9.7 years) was longer than the full cohort of completers (6.4 years). In terms of those participants who are quoted, all were diagnosed with hEDS, were of similar age to the full cohort of completers (36.9 years). Time since diagnosis (11.6 years) and duration of illness (16.7 years) were longer than the full cohort of completers. Thematic analysis of 10 interview transcripts identified 131 codes which translated to three key themes summarized in Table 3 along with their subthemes.

### Theme 1: increased body awareness

Participants reported that the mindfulness intervention enhanced awareness of their body and pain. Eight participants described how practicing meditation influenced recognition and response to pain, and how this was helpful:

it's a benefit for me because it just focuses your mind on what and where your pain is, so I didn't give it much thought what my pain is I just knew I had pain, so I think that's what I have got out of it; focus more on what my pain is and deal with things better that way. (Participant 2)

Participants reported that opportunity to recognize the location of their pain and type of pain was helpful, for example, it was a chance to acknowledge tensing: "you discover where you are guarding" (Participant 10). This heightened awareness can enable individuals to take action that may help improve their pain: "you then know maybe where you need to take it from there like your physio or Pilates or whatever you use to strengthen" (Participant 10). However, some participants they found this increased awareness led to a greater focus on their pain which was not helpful:

yeh, I think it made me more hyper aware. So, for example my toe was hurting, and I hadn't really noticed that before. So, I think like I say it can be a pro or a con. (Participant 3)

Participants agreed that mindfulness had fostered greater awareness of the body and their current pain sensations, viewing this as a benefit which could influence positive behavior changes. Others perceived it as a disadvantage as they focused more on their pain. Additionally, participants reported that mindfulness could help with attention and feeling a greater sense of mental clarity: "my head just feels clearer I haven't got that whirling going around in my head since I have started" (Participant 2) and "feeling a bit more focused that was a really good benefit" (Participant 6). Enhanced attention is associated with effective self-regulation and maintaining focus on goals. In this respect, meditation practice may allow participants to feel they have better control over the impact of their pain on other domains such as work and social life.

### Theme 2: reduction in pain intensity

All participants reported reduction in pain intensity associated with mindfulness meditation practice, an effect illustrated clearly by a participant who woke up during the night due to pain and by practicing the brief meditation achieved relief from the pain:

That I would be in that much pain and be able to manage myself out of that and back to sleep because as you probably know if you are in that much pain that you don't sleep then the not sleeping it makes the pain worse and it is like a circle from hell. But that I was able to break through that ... that was pretty amazing to me. (Participant 1)

Other participants recognized that during meditation attention shifted away from the intensity of pain experience thereby providing relief from the pain sensations:

The middle bit the breathing bit I think that's the bit I found most beneficial as I was able then to just focus on the breath and that's it and at that moment, I sort of not forgot but didn't pay so much attention to the pain or being uncomfortable. (Participant 8)

Participant 5 reported a similar experience that altered perception of pain intensity: "this is not a magical cure but it's just that focus on something else focused on breathing just takes that pain down the level." Meditation enables acknowledgement of a difficulty, in this case pain, without overidentifying with associated sensations, achieved by maintaining attention on sensations of the breath and letting go of other sensations, thoughts, emotions in peripheral awareness. The participant quotes illustrate how they "discovered" this and appear to reveal a reappraisal process in terms of coping with pain.

A subtheme of relaxation and improved sleep related to altered pain perception. Participants described their difficulties with insomnia due to pain or depression and others outline their battle with chronic fatigue. Participants reported that practicing meditation allowed them to relax and unwind. The majority used references to be feeling "relaxed" and "calm." Participants identified that this helped promote sleep and alluded to better sleep quality helping with pain and fatigue:

if I was getting a good quality of sleep, like it's all theoretical but if I was getting more sleep, I would be able to handle my pain better and get a bit more energy. (Participant 3)

Most participants stated they would be continuing with the practice following the intervention to help support their pain because they found the mindfulness intervention valuable. Participants acknowledged that meditation had benefits, helping with relaxation, sleep, and mental health, which provided motivation to continue the practice of meditation.

### Theme 3: challenges and barriers to completing daily mindfulness meditation

The third theme illustrates the barriers experienced by participants completing daily mindfulness meditation and ways the practice was adapted to make it more accessible and relevant. It also uncovers recommendations for future use. The main barriers identified were "forgetting" and "distractions." Some found it helpful to set reminders to prompt them or to practice at a certain time each day. Several participants described how they were put off meditating because they wanted to avoid the thoughts and sensations that might arise when meditating: "struggle to sit with my pain and that's what I have always found that mindfulness does for me" (Participant 6). There was a worry that mindfulness could make pain or anxiety worse.

It emerged that a barrier to continued practice was boredom because only one format of guided meditation was available.

Many participants suggested that a variety of mindfulness exercises, variation of narrator (male and female), and varied duration of guided meditation would be useful:

I tend to sort of mix and match like what I listen to just for a bit of a variety and I found having like the same the same thing each time I just kind of stop listening to what was being said like I just kind of zoned out. (Participant 4)

Several participants suggested use of a variety of meditation, such as body scans and guided visualizations. For example, some may be suitable for pain and others for fatigue and anxiety. Some participants felt the length of practice was short enough “the information you thought about there was enough there to keep your concentration, so I thought it was ok for me” (Participant 2) and was helpful for those new to meditation. Those with previous experience of meditation felt that a longer practice would be beneficial. Therefore, providing more choice would enable individuals to tailor mindfulness to their own needs and facilitate regular engagement with meditation practice.

## Discussion

The aim of the current study was to evaluate the effect of practicing mindful meditation on pain experience and quality-of-life in patients with hEDS. The numerical data support our prediction that practicing meditation daily for a brief time leads to improvements in quality-of-life and pain experience. Participants provided valuable insight of their lived experience of meditation practice related to pain suggesting that meditation practice was acceptable and enabled participants to reappraise their pain experience in a beneficial way. This study provides novel information regarding the effect and feasibility of internet-delivered mindfulness meditation for pain and quality-of-life in people with hEDS/JHS although limited in scope due to lack of an appropriate control group.

Without an appropriate inactive or active control group it is not possible to draw firm conclusions about the effects of meditation practice on quality-of-life. Nonetheless, there was a significant improvement in all quality-of-life indicators on the SF-36, except for physical functioning and the physical component summary score, with medium effect sizes for role limitations due to physical health and emotional problems, energy/fatigue, emotional well-being, social functioning, pain, general health, and the mental component summary score. Minimally clinically significant improvements (MCID) on the SF-36 ranged from 2 to 13 for physical domains and 4 to 18 for mental domains. Previous studies of MCIDs in patients suffering pain conclude that interventions improving MCIDs by less than five on the Physical and Mental Health components of the SF-36 are unlikely to be perceived as beneficial by patients [26]. In the current study MCIDs were above five on role limitations due to physical health and emotional problems, energy/fatigue, emotional wellbeing, social functioning, and pain. At face value such improvement is notable; however, the method used to calculate MCID is sample size and distribution dependent. Distributions of SF-36 dimensions were non-normal which can bias MCIDs [27]. Notwithstanding the lack of a control group, interpretation of the effect sizes and MCIDs requires caution as hEDS/JHS participants did not move above the 50-point level SF-36 domains except for emotional wellbeing and pain. The mean values on all SF-36 dimensions for hEDS/JHS participants were lower than available normative data for the UK population [28]. The low values on SF-36 dimensions and physical/mental summary scores are comparable with recent evidence documenting the adverse effect of the hEDS/JHS condition on quality-of-life

[29]. Therefore, although significant improvements in quality-of-life domains are observed in the current sample, whether such improvements translate into clinically important changes compared to active control therapies requires further investigation. Systematic reviews and meta-analyses of mindfulness-based approaches for chronic pain indicate mindfulness meditation is associated with small effects in terms of improved pain symptoms and improvements in quality-of-life, albeit from evidence that does not meet high standards of quality [12]. The evidence from the current study is in accord with those conclusions and does not add to quality in terms of methodological rigor because it lacks a control group. However, mindfulness-based approaches have not been evaluated in hEDS/JHS patients and thus our novel evidence points to a need for rigorously controlled studies to establish the potential benefit of meditation for this group. In terms of clinical practice, our evidence suggests that for those who engage with mindfulness meditation they experience quantifiable and subjective improvements in pain experience and related quality-of-life. For participants motivated to try meditation, it is reasonable to recommend this approach if contextualized and delivered alongside standard care.

Thematic analysis of interview transcripts generated three key themes that identified the nature of experience of the meditation practice. Firstly, participants described a heightened awareness of their body, location of pain, type of pain, and physical tensing. Several participants referred to this as a difficulty, but many reported this heightened awareness enabled choice of how to respond to the pain. Practicing mindful meditation will lead to heightened awareness of sensations [15] and the transcripts suggested that some form of reappraisal is enacted by participants, which accords with the mechanistic explanation of meditation effects in novice meditators [15]. Greater awareness of the nature of pain coupled with balanced reappraisal may empower patients to communicate more effectively with health practitioners and make more skillful choices on how best to manage pain. Secondly, participants expressed reductions in pain intensity suggesting this was because of focused attention on the breath. This experience accords with the explanation of how meditation works. By training attention to focus on an object, for example the breath, a state of non-elaborative, non-judgmental awareness of the present moment experience emerges [14]. In the case of chronic pain this enables the person to notice the pain sensation without overidentifying with it, which in turn enables adaptive responding rather than habitual reacting. By consequence, the subjective unpleasantness of pain may attenuate thereby enabling clear focus on daily living and life goals such as work and socializing. Thirdly, participants spoke of barriers to practice and suggestions for improvement of the intervention to make it more attractive to use. In relation to barriers participants suggested improvements in delivery and content of the intervention with an emphasis on offering choice. Participants referred to the importance of opportunities to select different mindfulness sessions that are easy to engage and not repetitive. Providing different durations of practice was also noted as being beneficial and having formal and informal mindful practice is often recommended [18]. Expanding the breadth of meditation practice to include variety could reduce barriers identified, such as boredom and distraction.

The amount of meditation practice reported by completing participants varied. The majority reported meditating between 9 and 14 days and 82% reported practicing two to three times per day. This study was not statistically powered to assess the contribution of meditation time to changes in outcome measures. Evidence that even one week of meditation practice can lead to



changes in pain intensity and subjective ratings of unpleasantness [15] suggests that SF-36 outcome measures could have been affected by practice time. Assuming that completers were novice meditators, reduction in pain intensity and unpleasantness may occur from meditation by learning to effortfully reappraise pain sensation [15]. Additionally, the reappraisal process may enable change in fatigue and emotional wellbeing. Further research can establish evidence for this possibility. Given the pilot nature of the study and its mode of delivery, future work should address factors that impact a high drop-out rate. Despite meeting the criterion of willingness to practice meditation daily for two weeks, over half of those recruited did not complete post-intervention assessment. Inspection of demographic and related data does not explain this drop-out rate. Completers were significantly older than non-completers but did not differ on other measures. The high percentage of females with a diagnosis of hEDS accords with epidemiological evidence [1]. Women may have been more inclined to register for the study because they more readily access alternative therapy in addition to conventional treatments than men. However, willingness to use alternative therapies did not differentiate completers and non-completers. It is reasonable to speculate that those who dropped out may have disliked the meditation after watching the video instructions, were put off by the prospect of practicing daily for two weeks or that internet errors may have occurred that prevented completion of instructions at pre-test stage. Without interviewing non-completers, it is difficult to offer a viable explanation. Based on the feedback from interviewed participants, future studies of mindfulness for hEDS patients should consider creating variety in content of meditation offered, variable duration and gender narration of guided meditation, flexibility to adapt meditation for differing needs, and instruction about how best to deal with forgetfulness and distractions.

### Limitations

It is unclear why so many participants dropped out after completing pre-intervention assessment. Although there were slight differences in age and ethnicity between non-completers and completers, there was no difference in terms of quality-of-life domain scores. A high drop-out rate is a challenge to the internal validity of the study design. The sample was mostly Caucasian and thus the outcomes do not generalize to other ethnic groups. The study did not assess or control for meditation experience. Thus, it is possible that motivation to take part may have arisen from previous experience of meditation. Sampling of participants for thematic analysis was based on interest to engage with the researcher. This sampling could be biased by overrepresentation of those who had a positive experience using the meditation. Additionally, it was not possible to employ a second coder to validate coding and themes derived from analysis of transcripts

### Conclusion

This mixed-methods study of the effects of mindfulness on pain and quality-of-life in patients with hEDS reveals that meditation practiced daily over two weeks has a beneficial impact on patient experience. The prediction that significant improvement in quality-of-life scores on the SF-36 was supported and is reflected in patient reports of their lived experience of practicing the meditation. People who suffer from hEDS experience a wide range of social, emotional, and physical symptoms. The study outcomes echo recent recommendations that clinical management of hEDS

should address psychological aspects of patient experience that affects perception and adaptation to pain [9]. Improvements in SF-36 dimensions seen in the current study could be explored further. Most interviews referred to coping better with pain, yet improvements were seen across a range of quality-of-life dimensions. Future studies could structure interviewing more precisely by focusing on the nature of different SF-36 dimensions. The novel aspect of our study involved brief intervention *via* the internet, thereby reaching more people than in-person contact. The interview data indicates that this approach is acceptable and feasible. However, patients emphasized the need to improve the intervention by offering more choice and variety in duration of meditation exercises. Integration of internet-delivered mindfulness meditation to the standard care model would provide opportunity to explore more cost-effective management of the symptoms of hEDS patients, for example, combining physiotherapy with mindfulness meditation for pain and mobility care. Indeed, mind-body therapeutic approaches that include mindfulness meditation have a proven record of benefit for chronic pain patients [12,15]. Ultimately, the current or revised intervention needs to be assessed in a randomized controlled study with an appropriate control group. Further multidisciplinary research is warranted to establish the efficacy of self-guided mindfulness initiatives because they are low-cost interventions that may complement standard care approaches.

### Acknowledgments

The authors express gratitude to the UK Ehlers Danlos Support charity (EDS UK) and the UK Hypermobility Syndromes Association (HMSA) for assistance with recruitment of participants, and all participants for their cooperation.

### Disclosure statement

No potential conflict of interest was reported by the author(s).

### Funding

The author(s) reported there is no funding associated with the work featured in this article.

### ORCID

Paul Lattimore  <http://orcid.org/0000-0001-8714-7678>

### References

- [1] Tinkle B, Castori M, Berglund B, et al. Hypermobile Ehlers-Danlos syndrome (a.k.a. Ehlers-Danlos syndrome type III and Ehlers-Danlos syndrome hypermobility type): clinical description and natural history. *Am J Med Genet C Semin Med Genet.* 2017;175(1):48–69.
- [2] Malfait F, Francomano C, Byers P, et al. The 2017 international classification of the Ehlers-Danlos syndromes. *Am J Med Genet C Semin Med Genet.* 2017;175(1):8–26.
- [3] Voermans NC, Knoop H, van de Kamp N, et al. Fatigue is a frequent and clinically relevant problem in Ehlers-Danlos syndrome. *Semin Arthritis Rheum.* 2010;40(3):267–274.
- [4] Castori M. Pain in Ehlers-Danlos syndromes: manifestations, therapeutic strategies and future perspectives. *Expert Opin Orphan D.* 2016;4(11):1145–1158.

- [5] Chopra P, Tinkle B, Hamonet C, et al. Pain management in the Ehlers-Danlos syndromes. *Am J Med Genet C Semin Med Genet.* 2017;175(1):212–219.
- [6] Reyckler G, De Backer MM, Piroux E, et al. Physical therapy treatment of hypermobile Ehlers-Danlos syndrome: a systematic review. *Am J Med Genet.* 2021;185(10):2986–2994.
- [7] Linton SJ, Shaw WS. Impact of psychological factors in the experience of pain. *Phys Ther.* 2011;91(5):700–711.
- [8] Ehde DM, Dillworth TM, Turner JA. Cognitive-behavioral therapy for individuals with chronic pain efficacy, innovations, and directions for research. *Am Psychol.* 2014;69(2):153–166.
- [9] Baeza-Velasco C, Bulbena A, Polanco-Carrasco R, et al. Cognitive, emotional, and behavioral considerations for chronic pain management in the Ehlers-Danlos syndrome hypermobility-type: a narrative review. *Disabil Rehabil.* 2019;41(9):1110–1118.
- [10] Pardos-Gascon EM, Narambuena L, Leal-Costa C, et al. Differential efficacy between cognitive-behavioral therapy and mindfulness-based therapies for chronic pain: Systematic review. *Int J Clin Hlth Psych.* 2021;21(1):1–17.
- [11] Day MA. Mindfulness-based cognitive therapy for chronic pain: a clinical manual and guide. Chichester: John Wiley & Sons Inc.; 2017.
- [12] Hilton L, Hempel S, Ewing BA, et al. Mindfulness meditation for chronic pain: systematic review and meta-analysis. *Ann Behav Med.* 2017;51(2):199–213.
- [13] Kabat-Zinn J. Mindfulness-based interventions in context: past, present, and future. *Clin Psychol-Sci Pr.* 2003;10(2):144–156.
- [14] Malinowski P. Neural mechanisms of attentional control in mindfulness meditation. *Front Neurosci-Switz.* 2013;7:1–11.
- [15] Zeidan F, Vago DR. Mindfulness meditation-based pain relief: a mechanistic account. *Ann N Y Acad Sci.* 2016;1373(1):114–127.
- [16] Kabat-Zinn J. Full catastrophe living: using the wisdom of your body and mind to face stress, pain, and illness. Rev. ed. New York (NY): Bantam Books Trade Paperback; 2013.
- [17] Parikh SV, Huniewicz P. E-health: an overview of the uses of the internet, social media, apps, and websites for mood disorders. *Curr Opin Psychiatry.* 2015;28(1):13–17.
- [18] Williams M, Penman D. Mindfulness: a practical guide to finding peace in a frantic world. St Ives: Piatkus; 2011.
- [19] Ware JE. SF-36 health survey update. *Spine.* 2000;25(24):3130–3139.
- [20] Ware JE, Kosinski M, Dewey JE. How to score version two of the SF-36 health survey. Lincoln (RI): Quality Metric Incorporated; 2000.
- [21] Chan EKH, Zumbo BD, Zhang W, et al. Medical outcomes study short form-36 (SF-36) and the World Health Organization quality of life (WHOQoL) assessment: reporting of psychometric validity evidence. *Soc Indic Res Ser.* 2014;54:243–255.
- [22] Cohen J. Statistical power analysis for the behavioral sciences. 2nd ed. Hillsdale (NJ): Lawrence Erlbaum Associates; 1988.
- [23] Norman GR, Sloan JA, Wyrwich KW. Interpretation of changes in health-related quality of life - the remarkable universality of half a standard deviation. *Med Care.* 2003;41(5):582–592.
- [24] Braun V, Clarke V. Successful qualitative research: a practical guide for beginners. Los Angeles (CA): SAGE; 2013.
- [25] Braun V, Clarke V. What can "thematic analysis" offer health and wellbeing researchers? *Int J Qual Stud Heal.* 2014;9:1–2.
- [26] Ogura K, Yakoub MA, Christ AB, et al. What are the minimum clinically important differences in SF-36 scores in patients with orthopaedic oncologic conditions? *Clin Orthop Relat Res.* 2020;478(9):2148–2158.
- [27] Brigden A, Parslow RM, Gaunt D, et al. Defining the minimally clinically important difference of the SF-36 physical function subscale for paediatric CFS/ME: triangulation using three different methods. *Health Qual Life Out.* 2018;16:1–7.
- [28] Bowling A, Bond M, Jenkinson C, et al. Short form 36 (SF-36) health survey questionnaire: which normative data should be used? Comparisons between the norms provided by the omnibus survey in Britain, the Health Survey for England and the Oxford Healthy Life Survey. *J Public Health Med.* 1999;21(3):255–270.
- [29] Molander P, Novo M, Hallstam A, et al. Ehlers-Danlos syndrome and hypermobility syndrome compared with other common chronic pain diagnoses-a study from the Swedish quality registry for pain rehabilitation. *J Clin Med.* 2020;9(7):1–16.