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Developing a self-management intervention to manage hypermobility spectrum disorders (HSD) and hypermobile Ehlers-Danlos syndrome (hEDS): an analysis informed by behaviour change theory

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ABSTRACT

Purpose: Hypermobility Spectrum Disorders (HSD) and Hypermobile Ehlers-Danlos Syndrome (hEDS) are heritable connective tissue disorders associated with joint instability and pain, but with scant guidance for supporting patients. The aim was to determine recommendations for an HSD/hEDS self-management intervention.

Materials and methods: Barriers to self-management were mapped onto the Theoretical Domains Framework (TDF) and Capability, Opportunity, Motivation-Behaviour (COM-B) model in a behavioural analysis. A modified Nominal Group Technique was used to prioritise behaviour change technique (BCT) interventions ($n = 9$ women).

Results: Possible BCTs incorporated:

Education: Incorporating self-help strategies, education to improve their knowledge of HSD/hEDS, and how to judge information about HSD/hEDS.

Training: In activity pacing, assertiveness and communication skills, plus what to expect during pregnancy, when symptoms can worsen.

Environmental restructuring and enablement: Support from occupational therapists to maintain independence at work and home.

Modelled behaviour: That illustrates how other people with HSD/hEDS have coped with the psychosocial impact.

Conclusions: This study is the first to apply theoretically-informed approaches to the management of HSD/hEDS. Participants indicated poor access to psychological support, occupational therapy and a lack of knowledge about HSD/hEDS. Future research should evaluate which intervention options would be most acceptable and feasible.

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► IMPLICATIONS FOR REHABILITATION

Patients with Hypermobility Spectrum Disorders or Hypermobile Ehlers-Danlos Syndrome can be active partners in the co-design of behaviour change interventions.

Behaviour change interventions should target psychological support and patient education, particularly patient information.

Additional behaviour change interventions included environmental restructuring and enablement; adaptations to participants' environment with input from occupational therapy.

Participants were keen to suggest opportunities for behavioural modelling; positive first-person modelling narratives, written by those with HSD/hEDS, which addressed how they coped with the psychosocial impact of their condition.


Background

Hypermobility spectrum disorders (HSD) and hypermobile Ehlers-Danlos syndrome (hEDS) are heritable disorders of connective tissue characterised by chronic joint pain, muscle pain, and multisystemic symptoms [1]. Patients can experience restricted physical and psychological functioning, and impaired quality of life [2,3]. A lack of awareness of the syndromes can cause considerable delay

in diagnosis, and patients have indicated that healthcare professionals may struggle to understand or manage their condition [4,5].

Historically, generalised joint hypermobility with additional symptoms of joint pain and instability was referred to as Joint Hypermobility Syndrome (JHS), or Ehlers-Danlos Syndrome Hypermobility Type (EDS-HT). These were thought to be the same

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 Supplemental data for this article can be accessed [here](#).

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condition, due to overlaps in presenting symptoms [6]. In 2017, the diagnostic criteria were updated to make clear the distinction between Hypermobility Spectrum Disorders (HSD) and Hypermobile Ehlers-Danlos Syndrome (hEDS), which has more selective diagnostic criteria [7]. For clarity and relevance, the newer terms will be used throughout this paper. The prevalence of the condition is unknown, however 30% of those screened at a musculoskeletal triage clinic in London were found to meet the former diagnostic criteria [8].

There has yet to be any high-quality research with this population exploring how to overcome the challenges identified by patients in daily life. This is important, as poorly managed chronic pain and recurrent injury in hEDS and can be significantly disabling [9,10]. This may lead to fear of movement and catastrophising responses to symptoms, leading to muscle deconditioning, and fear of injury and pain, potentially resulting in an over-reliance on emergency care [11,12]. Described as a physical and psychological decline in the HSD/hEDS literature, poorly managed symptoms can lead to substantial emotional costs such as low confidence, anxiety, depression and social isolation [10].

Patients who have HSD and hEDS have expressed that they would like to receive greater support and guidance in managing their condition, but that provision for this was not available [13]. As HSD and hEDS are lifelong chronic conditions, self-management is considered a mainstay of treatment [14]. However, compared to the substantial recommendations for patient education and self-management for other musculoskeletal conditions such as inflammatory or degenerative arthritis [15], there is very little recognition or patient support for patients with HSD/hEDS.

Self-management interventions have been shown to be very beneficial for patients with chronic pain conditions such as osteoarthritis, with improvements in mood, self-efficacy, anxiety, depression, and associated healthcare costs [16,17]. Participants with HSD/hEDS have the potential to benefit from self-management interventions to better control their symptoms [18–20]. Supported self-management is also a feature of the NHS Long Term Plan, which aims to support individuals to develop the knowledge, skills and confidence to live well with their long-term conditions [21]. With shifts in care towards supported self-management, rather than being seen as a patient with symptoms that require treatment, patients will instead be recognised as active partners in the management of their health and wellbeing [21].

When designing self-management interventions, recent research has highlighted that self-management interventions underpinned by psychological theories, are more effective compared to interventions that are not [22]. The Theoretical Domains Framework (TDF) was developed by a consensus group of behavioural scientists and stakeholders [23] to make the assortment of behaviour change theories more accessible to other disciplines, such as public health [23,24]. The TDF is based on a synthesis of thirty-three theories of behaviour and behaviour change, clustered into firstly twelve [24], and later fourteen domains to create one combined theory of behaviour change [23]. The TDF includes a wide range of factors that are likely to influence participant behaviour change. Additionally, the TDF domains can be mapped to the 'COM-B model' in what is termed a behavioural analysis. The COM-B model indicates that for a desired behaviour (B) to happen, each person must have the three factors. 'Capability' (C) can be defined as the physical or psychological capacity of a person to engage in the activity, including the required knowledge and skills; 'Opportunity' (O) refers to the factors external to the individual that can enable or prompt the behaviour; and 'Motivation' (M) signifies the processes that encourage and direct

behaviour, including habits, emotional responses and decision-making [25]. Mapping to the TDF and COM-B allows the identification of possible behaviour change techniques (BCTs, Table 1).

The mapping process involves the matching of barriers to self-management in HSD/hEDS, as identified by past research, to subsections of the TDF, and then to the corresponding COM-B domain. For example, prior research had indicated that participants with HSD/hEDS experienced fear and catastrophising relating to their condition, with fears of sudden declines in mobility [5,26,43]. These mapped to the TDF domains 'Knowledge' (as patients view their illness representation as degenerative) and 'Emotion' (fear). The corresponding COM-B domains were psychological capability (participants knowledge about their condition), and automatic motivations (representing the automatic 'fear' response). The intervention functions, or what the intervention would do, were 'education', 'persuasion', 'incentivisation', 'coercion', 'modelling and 'enablement'. The possible intervention for fear and catastrophising associated with HSD/hEDS was 'education for patients addressing knowledge and management of HSD/hEDS, pain control, self-help measures and fears about decline.'

In order to fully understand the impact of HSD/hEDS on participants, and their preferences for intervention content, the views of patients with the conditions were vital to inform these findings. This has the advantage of providing feedback from key patient stakeholders. In addition, these findings can be used to inform the content and delivery of any proposed intervention, which would be grounded in patients' own views and experiences.

The aim of the present study was to determine recommendations for the components of a self-management intervention for people with HSD/hEDS. The TDF and COM-B were used to conduct a behavioural analysis and identify potential intervention options (see Table 1), based on prior published research with patients who had HSD/hEDS. This is the first study to use behaviour change theory to identify intervention options with a HSD/hEDS population. Prior results from research with patients who had HSD/hEDS was used to prioritise elements of a self-management intervention informed and driven by patients' own experiences and preferences [13,26].

Methods

Study design

This study comprised three stages. Firstly, the TDF and COM-B were used to conduct a behavioural analysis. Secondly, the outcomes of Stage 1 were presented to a focus group of key patient

Table 1. Definitions of BCT interventions adapted from Michie et al. [25].

Intervention	Definition
Education	Increasing knowledge or understanding
Persuasion	Using communication to induce positive or negative feelings or stimulate action
Incentivisation	Creating expectation of reward
Coercion	Creating expectation of punishment or cost
Training	Imparting skills
Restriction	Using rules to reduce the opportunity to engage in the target behaviour (or reduce the opportunity to engage in competing behaviours)
Environmental restructuring	Changing the physical or social context
Modelling	Providing an example for people to aspire to or imitate
Enablement	Increasing means or reducing barriers to increased capability or opportunity

Table 2. The 20 potential BCT interventions agreed after discussion during the Stage 2 focus group, and reviewed in the Stage 3 NGT groups.

Psychological capability	Social opportunity	Physical opportunity	Automatic motivation	Reflective motivation
1. Skills development training for patients focusing on advocacy, assertiveness and communication, to improve interpersonal communication of their needs.	7. Behavioural modelling examples to show how to communicate your needs to others in social situations.	10. To safeguard from negative or unreliable information: a) Identify and restrict access to unreliable information sources. b) Enable ease of access to reliable information within an easily-located webpage or source.	11. Educational examples of behaviours, including self help strategies for coping with injury and pain.	16. Educational programmes with a focus on self-help and coping strategies for injury or pain.
2. Education for patients addressing knowledge and management of HSD/HEDS, pain control, self-help measures, fears about decline.	8. Modelling narratives that emphasise independence from family members in completing daily tasks.		12. Positive first-person modelling narratives that address some of the negative aspects of HSD/HEDS (depression, distress, frustration, sexual dysfunction feelings of loss) and how they coped.	17. Education to manage beliefs and perceptions about body image.
3. Pregnancy with HSD/HEDS - Improved education, training and information for participants regarding what to expect.	9. Environmental restructuring and enablement: altering the physical environment, with occupational therapy input, in order to achieve tasks independently.		13. Modelling of coping strategies from mothers with HSD/HEDS who have had children.	18. Persuasion- Emphasising patient's capability in social situations to reduce fear.
4. Establish guidance regarding trusted, accurate sources of information for HSD/HEDS.			14. Education regarding the likelihood that their child will inherit HSD/HEDS and signposting for support.	19. Persuasion of capability regarding physical ability, to reduce fear of injury.
5. Training in pacing skills where individuals can learn to actively manage cycles of activity and rest to achieve increased participation in daily activities.			15. Develop templates outlining examples of increased HSD/HEDS symptoms during pregnancy and what to do, to act as a support tool.	20. Education regarding consequences of overexertion and exacerbations of pain/fatigue.
6. Promote information to improve knowledge of accessible seating or parking.				

partner and researcher stakeholders who selected potential behaviour change interventions. Thirdly, two modified Nominal Group Technique (NGT) groups with patient stakeholders were conducted to rank intervention options, and to discuss these in terms of appropriateness, acceptability, feasibility, perceived barriers and potential solutions.

Full ethical approval was granted by the University of the West of England Faculty Research Ethics Sub-committee (HAS.18.03.128).

Stage 1: behavioural analysis

Procedure

When collecting information to identify what needs to change, Michie et al. [27] recommended that data is collected from as many relevant sources as possible, as the most accurate representation will be gained by using input from multiple perspectives [27]. By triangulating data from multiple published sources, all using first-hand qualitative interactions with participants who have HSD/hEDS, we can gain a greater insight into the potential barriers to self-management. Therefore, in order to gather as broad a range of data as possible, a systematic review and thematic synthesis of patients' lived experiences of JHS and EDS (prior terms for HSD/hEDS) was conducted [26]. Secondly, semi-structured telephone interviews with 17 purposively selected participants (14 women, 3 men) asked about the psychosocial impact of the syndromes on their lives [13]. The current paper will focus on the mapping of this published information against the TDF and COM-B.

Analysis

All barriers to successful self-management of HSD/hEDS were transferred from NVivo 12 qualitative analysis software into Microsoft Word. A coding framework was generated, in which each barrier to self-management of HSD/hEDS was inductively coded against the TDF and COM-B by the first author (SB), who had completed Behaviour Change Technique (BCT) taxonomy training. [Supplemental File 1](#) shows the results of the mapping process. Great care was taken during the mapping process to ensure checking and accuracy of the mapping results. Final coding for each element of the framework was discussed with co-authors (NW, TM, SP) to reach consensus. The results were reviewed by a patient research partner who had personal experience of JHS, to ensure that the materials were clear, comprehensive and relevant to patients with HSD/hEDS. Results were also discussed with an external researcher with experience of mapping behaviour change techniques to the TDF [28].

Stage 2: focus group with key stakeholders

Sample

To identify which BCTs should be included, the previously agreed mapping results were presented to a research team and external members in a focus group. Participants in the Stage 2 focus group comprised researchers with experience of qualitative research, the COM-B behavioural change framework [29], knowledge mobilisation and implementation in osteoarthritis and a patient research partner with JHS.

Procedure

Potential intervention options were presented in a table developed by the first author (SB, see [Supplemental File 2](#)). The table

Table 3. Participant demographics for attendees of both focus groups ($n = 9$).

ID	Pseudonym	Age	Gender	Ethnicity	Diagnosis	Year diagnosed	Five-item score ^a
001	Helen	43	Female	White	JHS & HSD	2015	3
002	Alex	41	Female	White	JHS & EDS-HT	2018	3
003	Kelly	57	Female	White	EDS-III & EDS-HT	1998	5
004	Kris	25	Female	White	JHS	2002	5
005	Jody	51	Female	White	JHS & HSD	2012	3
006	Heather	39	Female	White	JHS	2018	4
007	Elsa	34	Female	White	JHS & HSD	2016	5
008	May	57	Female	White	JHS	2013	2
009	Julie	28	Female	White	JHS & HSD	2015	4

^aFive Item Hypermobility Score, where scores ≥ 2 indicate hypermobility, Hakim and Grahame [33].

EDS-HT: Ehlers-Danlos Syndrome Hypermobility Type; EDS-III: Ehlers Danlos Type III; an earlier diagnostic term for EDS-HT, HSD: Hypermobility Spectrum Disorder; JHS: Joint Hypermobility Syndrome.

listed each potential BCT under the relevant section of the COM-B, along with definitions of key terms used in the document, an overview of the TDF and COM-B, and definitions of intervention functions, or what each intervention would do. For example, for the barrier “participant fear of decline or catastrophizing” the intervention function ‘education’ was defined as ‘increasing knowledge or understanding’, with the possible BCT outcome as ‘education for patients addressing knowledge and management of HSD/hEDS, pain control and self-help measures, fears about decline.’ These were emailed to focus group participants prior to the meeting, in order for participant to have time to review definitions and intervention options prior to discussion. The focus group lasted for 90 minutes.

Analysis

Supplemental File 3, Table S2, shows the changes made to the potential BCT options including reasoning and supporting quotes, as a result of focus group discussion at Stage 2. Each BCT intervention option was discussed with the team to reach consensus. Following the suggestions of group members, some BCTs were removed, and others altered to further clarify the options available for NGT participants to vote on in Stage 3. BCTs were judged based on how well they represented the self-management of HSD/hEDS at the individual patient level. For example, BCTs that referenced broader aims, such as the training or information needs of healthcare professionals (“*informative HSD/hEDS leaflets and guidance to increase healthcare professional knowledge and understanding.*”) were excluded at this stage (see Supplemental File 3). From this focus group discussion, the BCTs were refined and reduced from 41 to 20 potential intervention options to change the behaviour of patients with HSD/hEDS. The focus group lasted for 90 min. A list of the 20 refined BCTs is available in Table 2.

Stage 3: nominal group technique

Sample

A modified NGT method was chosen due to its benefit in gaining reliable qualitative information from expert participants within a face-to-face focus group [30]. The NGT is a highly structured group process that involves equal participation and input from all participants [31], which ensures that, unlike traditional focus groups, there is no risk of one or more participants dominating discussions [32].

For the modified NGT groups, a convenience sample of participants was recruited through online social media advertisements posted by two patient charities; The Hypermobility Syndromes Association (HMSA) and Ehlers-Danlos Support UK (EDS-UK) inviting eligible participants to email the principal investigator if they

wished to take part. Convenience sampling was chosen due to the face-to-face nature of the tasks, as participants had to be able to travel to sites in London and Bristol for focus groups. Perhaps as a result of this method, fewer participants expressed an interest in participating in the study, and for this reason purposive sampling was not possible. This has been further discussed within the study limitations.

Participants were all women of white ethnicity aged between 25 and 57 (mean age = 42 years). Seven participants were from the South-West of England, one from the Midlands and one from the North-East. Although the 2017 diagnostic criteria for the Ehlers-Danlos Syndromes had been published [1], participants had yet to be re-assessed for an updated diagnosis. However, all had a self-declared diagnosis of JHS, HSD, EDS-HT or EDS-III, diagnosed between 1998 and 2018, and had been assessed using the Five Part Questionnaire (5PQ), a self-reported test for identifying generalized joint hypermobility, a potential indicator of HSD/hEDS [1,33]. Participant demographics are described in Table 3.

Procedure

The meeting was facilitated by SB, supported by SP. Participants were first invited to consider a paper copy of the 20 behaviour change intervention options and to privately rank these by importance. If participants had additional ideas or contributions, they were invited to take note of these to share at the discussion stage of the process. Next, participants were presented with each of the potential BCTs electronically projected on a large screen (available in Supplemental File 4). Participants were given a TurningPoint[®] ResponseCard[®] keypad (an electronic device with numbered keys) enabling them to individually and anonymously vote for each potential BCT. Participants were asked to respond to each of the 20 BCTs while considering the proposed question:

“Which of these factors would you consider important in a HSD/hEDS-related intervention?”

For each of the BCTs, clarifying information (such as the underlying themes and features of the qualitative data informing the creation of the BCT), and definitions of each term used were made clear, and participants given the option to ask any clarifying questions before voting. Participants could respond using a four-item Likert scale; from 1 = Not important/not applicable to 4 = Very important.

The next step in the NGT process involved participants verbally proposing any additional ideas in turn, around the group, without debate [34]. Participants’ ideas were edited and refined by the NGT group facilitators (SB & SP), with input on wording and general suggestions from group participants, to ensure a good fit with the COM-B and TDF definitions. Care was taken to match the new suggestions with agreed definitions and methods of behaviour change, as set out by Michie et al. [27] to ensure as close a

fit with the previous 20 identified options as possible. When the refinement of new BCTs was complete, participants were asked to vote on each new idea, using their TurningPoint® ResponseCards® keypad (an electronic device with numbered keys) and the same 4-item likert scale.

Next involved a group discussion. Each participant was invited to revisit their initial ranking of the original 20 BCTs (step one) along with the additional BCTs identified by participants. Participants were then asked to verbally identify their top two preferred intervention options and to discuss these with the rest of the group. The question posed was;

“From the items discussed, which 2 items would you prioritise as most important in a HSD/hEDS-related intervention?”

Participants were asked to discuss the appropriateness, acceptability, feasibility, perceived limitations and proposed solutions relating to a HSD/hEDS intervention. The NGT groups lasted an average of 3-4 h.

Analysis

The NGT process was recorded using a digital recorder and transcribed verbatim (identifying information removed to ensure participant confidentiality). Participants' contributions using TurningPoint® keypads were saved within the software before being imported for summation using Microsoft Excel.

Results

Results from stage 1: behavioural analysis

Mapping in this case refers to the matching process between qualitative barriers identified by participants against the relevant subsections of the TDF and COM-B and then to intervention functions. The potential intervention functions, with their associated mapping categories, were presented in a table with themes represented under each of the six COM-B subdivisions. All TDF domains were coded as part of the analysis. The most frequently coded TDF domains were Knowledge; Emotions; Social influences; Social/professional role and identity; Skills; Beliefs about capabilities; and Beliefs about consequences. While not coded as frequently, codes were also identified for: Environmental context and resources; Pessimism; Person and environmental interaction; and Behavioural regulation. An overview of the results is provided in [Supplemental File 1 Table 1](#).

Results from stage 2: focus group to choose potential BCTs to include in a HSD/hEDS intervention

During the process of discussion, the focus group actively discussed and deliberated over the correct wording and definitions for each potential intervention function, whilst being mindful to keep the intended focus on individual self-management of HSD/hEDS. Some BCTs were removed following discussion and 6 amended to improve clarity. [Supplemental File 3, Supplemental Table 2](#) shows the changes and decisions made in the discussion and refinement from 41 to 20 potential BCT options, including reasons for exclusion. By excluding a selection of potential behaviour change interventions, this enabled the process to continue to focus on self-management of HSD/hEDS at the individual, patient level. The potential BCTs for patient self-management were rewritten based on group feedback in preparation for Stage 3 ([Supplemental File 4](#)).

Results from stage 3: potential interventions in the self-management of HSD/hEDS

Both Group 1 and 2 ranked each of the 20 BCT interventions twice; before and after a group discussion. Participants in the Bristol and London NGTs proposed a total of 4 (Bristol) and 3 (London) additional self-management ideas (see [Table 4](#)). Ethical approval was granted to allow for participants to vote on all additional ideas on completion of both NGT groups. However, few participants responded to follow-up. For this reason, the additional ideas from the Bristol NGT were considered for final inclusion, as these had the highest number of final votes.

Before data collection began, it was agreed that participants' consensus score would be calculated by combining likert scale scores into categories of important (3 = Somewhat important; and 4 = Very important), and unimportant total scores (1 = Not important/not applicable; 2 = Somewhat unimportant). A tally was then made of the number of participants scoring each option as 'important' and this was expressed as a percentage of all participants. For example, if all 9 participants scored 3 = Somewhat important; or 4 = Very important, then the consensus score would be $9/9 = 100\%$. Consensus was reached when agreement was

89%, or scores of 8/9 or higher. Items with the highest total score (combined scores of 9 or 8) in the second round of NGT ranking were prioritised for intervention content. Although other examples of NGT prioritisation have used cutoffs of 80% [35], a cutoff of 7/9 (78%) gave a participant intervention prioritisation list of 20 items. After discussion, a pragmatic decision was made by the study co-authors to raise the consensus Level to 8/9 (89%) in order to focus on the top 14 items. The preferred interventions are no longer classified according to the COM-B model as consensus was drawn from the voting process in the NGT. The final 14 preferred intervention options, consensus scores, and supporting quotes from participants' discussions are presented in [Table 5](#).

Discussion

This is the first study of its kind to prioritise interventions for participants to self-manage their HSD/hEDS using the TDF and COM-B. The overall aim of this research was to determine the components of a self-management intervention for people with HSD/hEDS. In order to achieve this we identified what would be required to change for patients to better manage HSD/hEDS, using the COM-B model and TDF to conduct a behavioural analysis. Next, potential intervention components were decided through extensive discussion and input by an intervention development focus group (including expert researcher and patient research partner input), in order to decide which behaviours identified by the COM-B mapping exercise were viable for inclusion in a self-management intervention, as recommended by Michie et al. [27]. Finally, with input from key stakeholders with HSD/hEDS, a participant-centred modified NGT method was used to identify which BCTs should be prioritised and participant preferences for intervention content.

Participants prioritised 14 of the 27 potential BCTs (20 refined, 7 newly identified by NGT group members) in a systematic modified NGT for potential implementation. The remaining interventions, while not reaching a high enough threshold for inclusion in the final prioritised list, received a wide range of scores from participants, indicating that a holistic and multi-faceted intervention is required.

A number of options for patient education were identified for self-management of HSD/hEDS. The first of these was for patients to receive education interventions, including those that address

Table 4. Results from NGT groups ($n = 9$) participants' additional ideas for self-management interventions.

Additional Ideas: Bristol NGT	NGT final consensus score	Supporting quotes from NGT group discussions
1. Tailored information only when appropriate.	77% 7/9	"I think that sometimes [patient education and information] has the opposite effect ... I think there can be a lot of fear-mongering as well ... If I hadn't been pregnant, or had a child ... if I was being fed loads of information about things that might go wrong ... I think that might kick in the anxiety, and that anxiety and pain, for me ... So, I would rather have less information ... " [Alex] "Sex with arthritis,' ... Seventy-year-olds in the front? Smiling? (group laughter) and they're just giving you all these books, and there is not anyone under the age of seventy, in any of them ... reading it you just feel like, this doesn't really apply ... to you." [Julie]
2. Education: How to evaluate information.	100% 9/9	"There needs to be like, education, on like, how to identify what is negative or unreliable information? Because sometimes they're like, 'Oh, have a look on the Internet,' You know? To get some advice, but also, you need to be careful ... Anxious people can stir up a storm online, can't they?" [Julie] "We are going to have to face some stuff that we wouldn't like to hear, at times. And just because it's negative, doesn't mean it's not important information to have? ... I've got pelvic floor problems ... Again, it would have been nice to know about the risks of pelvic floor issues [with HSD/hEDS]" [Jody] "When I [was diagnosed] it was in the Nineties ... it was just ... 'here's a book' ... So it is obviously all been vetted, they put it together ... It's not some bloke on Facebook making up a load of rubbish" [Kelly]
3. Education: How to navigate social support	89% 8/9	"I've not been entitled to any benefits ... but I learnt the other week that I could be entitled to Access to Work funding? [.] I'm still ... Not optimistic about what I'm going to get! (Laughs) ... but, it's something I didn't know about until three weeks ago." [Elsa] "I've got a friend ... she was going through the [Personal Independence Payments] benefits thing ... And she had some help ... how do you even navigate that? Where do you start? Where else can I get support? [Heather]
4. Enablement: Access to emotional support e.g., mindfulness, counseling, CBT or books.	89% 8/9	"Try this strategy, and try this strategy, has been more helpful for me to ... To talk through kind of, the issues, and the acceptance, and stuff, rather than actually having ... A list of things to do, you normally come away with a list of physio exercises ... You come away with a list of medications, so I don't want to (laughs) come away with a list of like ... mindfulness exercises as well! It's hard work." [Elsa]
Additional Ideas: London NGT	NGT final consensus score	Supporting quotes from NGT group discussions
1. Education for others regarding HSD/hEDS- what it is and how it affects people.	22% 2/9	"I think [Additional Idea 1] is about expectation, so for me that's all part of the 'it's invisible, you can't see it, but it's real' thing." [May] "People will be able to lead with what they can do- their capability and their functionality ... not predetermining what that person's abilities would be". [May]
2. Education regarding common behaviours and lived experiences of HSD/hEDS (compared to other people who don't have the conditions).	22% 2/9	"It's a different kind of education, I think, it's not necessarily a medical intervention, it's a, 'This is the skeleton you were born with, this is the impact it might have, here's some really good ideas about how to sort some stuff out.'" [May] "So it's like identifying the discrepancies between the [HSD/hEDS] world, and the ... normal connective tissue world." [May]
3. Modelling examples from a mentor with HSD/hEDS.	33% 3/9	"A real person, because I think that creates two things, a touchpoint, which is a viewpoint where other people understand where you're coming from ... So if you have a mentoring structure, that allows the mentors to share knowledge." [May]

The aim of the NGT was to determine recommendations for the components of a self-management intervention for people with HSD/hEDS. The question posed to the group was 'Which of these factors would you consider important in a HSD/hEDS-related intervention?'

their knowledge and management of HSD/hEDS, pain control, self-help measures and fears about decline. In order to actively manage and take responsibility for their condition, participants need to take on knowledge and skills about how to manage their own health, especially as professional knowledge about their specific condition may be poor [36,37]. Improving patients' knowledge of their condition has been used to great effect in other chronic pain self-management interventions. However, participants in a diabetes self-management education programme were keen for knowledge about their condition that they could tailor to their own circumstances, and their own personal situation [38]. The need for tailored information links with participants' newly suggested education intervention for 'tailored information when appropriate', rather than generic information about their syndromes [36]. Delivering tailored information is considered an important factor in person-centred care, particularly for those with long-term conditions [39]. Recommendations for patient

education in rheumatoid arthritis (based on a systematic review of the literature) also indicated the need for patient education information to be tailored to each individual patient's needs, including educational needs such as knowledge and management of their disease, knowledge of risk factors and side effects [40].

The recommended BCTs also aimed to reduce participants' fears and catastrophising regarding their HSD/hEDS. Distress experienced with chronic pain, including anxiety, depression and fear of pain or injury has been shown to negatively affect participants' ability to self-manage their condition [41]. Fears identified by participants with HSD/hEDS have been related to injuries, pain and medical treatment [4,13,42,43], fears of suddenly declining [13] and fears relating to heritability, pregnancy and childbirth [4,13]. These beliefs were typically grounded in patients' lived experiences, or in information found on the Internet [13,26]. Interventions to manage fear have been very successful for chronic pain, such as self-management interventions to manage fear-avoidance

Table 5. Results from the Nominal Group Technique (NGT) focus group voting.

TDF domain	Possible BCT interventions for patient self-management in HSD/hEDS	NGT final consensus score	Supporting quotes from NGT group discussions
Education	Education for patients addressing knowledge and management of HSD/hEDS, pain control, self-help measures, fears about decline.	100% (9/9)	<i>"I haven't necessarily found a huge amount of help from medical professionals, I've certainly found that most of the knowledge I've gained has been from fellow hypermobile sufferers, or EDS, people who had those diagnosis ... I tend to look more towards those sorts of people as the people who are knowledgeable"</i> [Heather,]
	Educational examples of behaviours, including self help strategies for coping with injury and pain.	100% (9/9)	<i>"Figuring out stuff- ... self- help ... just ... chatting with other people today has been brilliant! (Laughs) ... You don't often meet people, you know, who've got similar experiences? I mean, you see them all on Facebook but ... It can be a bit negative"</i> [Kelly]
	Educational programmes with a focus on self-help and coping strategies for injury or pain.	100% (9/9)	<i>"The education never ends, you have just constantly keep going, learn how to ... sort yourself out, so now I like, Google the anatomy, find out which muscle is hurting, and then look up different physio stretches ... So you kind of just end up treating yourself, after a while!"</i> [Julie]
	Education regarding consequences of overexertion and exacerbations of pain/fatigue.	100% (9/9)	<i>"I'm the first one in my family with [EDS-HT], ... I feel if I'd known stuff before, I probably would have done [things] differently, or helped differently. I feel like I'm gathering information for trying to prepare my children for the issues that they may possibly have in the future"</i> [Alex]
	How to evaluate information. ^a	100% (9/9)	
	Templates outlining examples of increased HSD/hEDS symptoms during pregnancy and what to do, to act as a support tool.	89% (8/9)	
	Establish guidance regarding trusted, accurate sources of information for HSD/hEDS.	89% (8/9)	
Training	Education regarding how to navigate social support e.g., Blue Badge, charity support, benefits and ATW or DSA funding. ^a	89% (8/9)	
	Skills development training for patients focusing on advocacy, assertiveness and communication, to improve interpersonal communication of their needs.	100% (9/9)	<i>"Yeah, about being able to advocate for yourself, communicate, find the right people that can help- ... communicate [to] them in a way that they actually ... Understand what you're talking about ... I feel like I'm, you know, Talking different languages? On a different planet"</i> [Jody]
	Training in pacing skills where individuals can learn to actively manage cycles of activity and rest to achieve increased participation in daily activities.	100% (9/9)	<i>"The pregnancy, for me, like really snowballed everything, it really did."</i> [N/A] [Helen]
Environmental restructuring and enablement	Improved education, training and information for participants regarding what to expect during pregnancy.	89% (8/9)	<i>"I made the assumption, that maybe my hypermobility symptoms would go away [while pregnant] when in fact, it's the opposite ... And I only learned that today "I made the assumption, that maybe my hypermobility symptoms would go away [while pregnant] when in fact, it's the opposite ... And I only learned that today? So I would rather a doctor had been able to give me that information, than me assume?"</i> [Jody]
	Altering the physical environment, with occupational therapy input, in order to achieve tasks independently.	89% (8/9)	<i>"[Occupational Therapy is] just one of those small things that make such a big difference ... small adjustments you can make to your life that just has such a positive [impact]."</i> [Alex]
Modelling	Access to emotional support e.g., mindfulness, counselling, CBT or books. ^a	89% (8/9)	<i>"The amount that us, as individuals ... cost the NHS ... because were not having the right support, so we go to A&E ... I haven't had to go to A&E in the last year that I've been at [National HSD/hEDS treatment centre] ... And I've had the things I needed at home, to be able to deal with it."</i> [Elsa]
	Positive first-person modelling narratives that address some of the negative aspects of HSD/hEDS (depression, distress, frustration, sexual dysfunction feelings of loss) and how they coped.	89% (8/9)	<i>"When you're talking to somebody who's been through it ... Their knowledge and experience sometimes seems a bit more valid than someone who's read a lot of stuff in textbooks."</i> [Heather]

ATW: Access to Work; CBT: Cognitive Behavioural Therapy; DSA: Disabled Students' Allowance; a: Additional participant-proposed idea from NGT focus group 1.

beliefs involving catastrophising and fear of movement [44]. Fears relating to decline in HSD/hEDS can be associated with catastrophising; the belief that new or unusual symptoms are a signal of inevitable or impending physical decline [13]. Therefore, tailored educational information emphasising a general lack of evidence for physical decline in the majority of patients with HSD/hEDS would be beneficial.

In a recent systematic review of self-management interventions for people with chronic pain, practicing core self-management skills was found to improve participants' acceptance of their condition [41]. Acceptance of HSD/hEDS has been identified as an important factor in successful self-management [13,14] and can be a predictor of successful adjustment in chronic pain [45]. Modelled behaviours, performed by similar peers or family members, may help participants to have greater acceptance of their

condition and improved self-efficacy and confidence in their ability to master a skill [46].

Participants also indicated preferred options for training, including skills development focusing on self-advocacy, assertiveness and communication.

An exploration of a humanisation approach to managing HSD/hEDS recommended giving patients the agency to manage their own condition, such as through self-referral to physiotherapy [47]. A recent qualitative systematic review and meta-synthesis of self-management interventions identified a need for better communication across all stakeholders (patients, healthcare professionals, family and friends) in order to positively support patient self-efficacy and self-management of chronic pain [41].

Participants identified two interventions that related to environmental restructuring and enablement, and these related to the

physical and psychosocial impact of HSD/hEDS. Participants were keen for self-management, supported by occupational therapy input, to adjust their environment to complete tasks independently from their family members. The key barrier in this case was participants' lacking the physical capability to manage their own activities, and instead relying upon family members or partners for assistance. However, depending on others for support with daily activities can cause feelings of guilt and shame [26,43]. Occupational therapy input has successfully been used to influence the self-management of other chronic pain conditions, including Fibromyalgia [48]. Due to joint instability, participants with EDS-HT have also been reported as likely to fall [49]. As many as 96% of those with EDS-HT surveyed had experienced a fall within the previous 12 months, with 68% reporting balance problems such as unsteadiness and stumbling when walking [49]. Learning how to make adaptations to one's own environment can enhance feelings of independence and provide a sense of successful adjustment [50]. Therefore, the inclusion of occupational therapy in a self-management strategy for managing HSD/hEDS could be beneficial, in terms of improving participants' confidence; with the potential to reduce incidence of accidental injuries and participants' associated fear of movement.

Lastly, many were keen for an intervention to feature enablement, in order to access emotional support such as mindfulness, counselling or CBT. Enablement can be defined as increasing means or reducing barriers to increased capability or opportunity [51]. CBT skills, in addition to exercise and relaxation techniques have successfully been used with patients who experienced Fibromyalgia-related chronic pain [52]. In recent European recommendations for managing rheumatoid arthritis, the need for patient education to include discussion of emotional issues, psychological support, and support from healthcare professionals in managing emotional distress has been highlighted using methods such as mindfulness, breathing exercises, and stress-management skills to promote acceptance, enhance wellbeing and alleviate emotional distress [40].

Although this study focused on the barriers to self-management in HSD/hEDS, in any formal self-management intervention, facilitators to the self-management of HSD/hEDS, such as aerobic exercise or strength training would also be included. For example, since this study was conducted, novel research examining an inpatient self-management intervention for patients with HSD and hEDS was conducted in France [53]. The inpatient intervention programme for patients with hEDS included occupational therapy input, physical activities to improve muscular endurance and coordination, proprioception exercises to improve balance, relaxation exercises, and self-management workshops to improve patients' knowledge of hEDS [53]. However, although kinesiophobia and quality of life were significantly improved by the end of the programme, this was not maintained at 6 weeks post-intervention, which raises questions for patient management and support following the study [53].

The current study has a number of strengths. Firstly, the COM-B and TDF models have a strong theoretical underpinning and have facilitated the development of recommendations for a self-management intervention for patients with HSD/hEDS, through targeting a number of behavioural barriers to self-management. Using this method, it was possible to identify a number of influences on participants' behaviour that would have been difficult to identify using quantitative methods. For example, prior research had identified participants with HSD/hEDS as being significantly more fearful than the general population but with this qualitative approach a number of specific fears in relation to automatic

motivation and knowledge about HSD/hEDS have been identified; from fears about potential injury [5,14,43] to fears about future deterioration of their condition leading to catastrophising about symptoms [13].

A second strength was the involvement of participants with HSD/hEDS at both the identification stages (our patient research partner), and at the NGT stage (the nine participants), which also encouraged discussion and debate of each proposed BCT. Consultation with patients is likely to result in material that is more relevant, understandable and readable to patients [54] and there was evidence during the NGT discussions that participants found the BCTs proposed to be very relevant to their lived experiences, and were freely able to alter the wording of the additional interventions that they proposed to improve understanding and coherence. The use of a modified NGT and focus group methodology enabled group consensus to be established regarding preferred items for a HSD/hEDS self-management intervention. The methodology generated useful quantitative and qualitative data regarding participants' use, experiences, and preferences. However, it is acknowledged that by utilising NGT group discussions, thorough exploration of more sensitive HSD/hEDS-related issues (such as sexual dysfunction or prolapse) were less likely to be voiced by participants than in an interview setting.

One limitation is the convenience sample, with a small number of participants in the NGT study who were not particularly diverse. One reason for this may be the face-to-face nature of this methodology. Feedback from participants who were unable to attend the focus groups indicated that the significant effort required to travel and attend the group in person may have been a barrier to those who wanted to take part, particularly if they had more severe chronic pain or disability. Although the modified NGT process enabled priorities to be identified, this was based on the opinion of 9 women, and therefore may not be as representative of the priorities that others may choose, such as men or women from more diverse communities and ethnic backgrounds [55].

Participants' additional ideas for intervention options had to be matched to the COM-B and TDF definitions by the group facilitators (SB & SP) at the time of the NGT meetings. While every effort was made to ensure that these definitions both reflected what participants wished to see in the proposed intervention, and that these were true to other interventions proposed by the method, the fact that these additional ideas could not be given the same time and consideration as the other twenty proposed interventions is a potential limitation. However, in consultation, refinement and discussion of key concepts with participants at each of the modified NGT meetings, participants were content that the additional ideas proposed reflected what they would like from a self-management intervention. A further important limitation to note is that not all participants from Focus Group 1 responded to requests to rate the additional interventions proposed in Focus Group 2. Those interventions were therefore not voted on by all participants and could not meet the consensus threshold. A further limitation is the lack of consideration of aerobic exercises and strength work as part of this study. As this study focused on barriers to self-management, and participants considered exercise to actually be a positive and very beneficial facilitator to self-management of HSD/hEDS, this was therefore not a consideration of the mapping process. The barriers to completing exercises as part of HSD/hEDS self-management focused instead on participants' poor proprioception (a poor awareness of where their joints are). This poor awareness made it more difficult for some participants to complete their physiotherapist-prescribed

exercises at home [26,56]. In contrast, many participants spoke positively of regular aerobic exercise and support from physiotherapists; modifying their activities to better achieve their goals [13].

In summary, this study has provided a valuable insight into what patients with HSD/hEDS would like to see in a self-management intervention. Future research with professionals from a variety of disciplines would be valuable, in order to gain feedback regarding the acceptability and feasibility of such an intervention in the context of healthcare resources. Further work could then develop supporting materials to allow for the effective delivery of the identified BCTs. Information generated by the current study is thus vital in informing future research and development of a holistic, patient-centred intervention for the management of HSD/hEDS.

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