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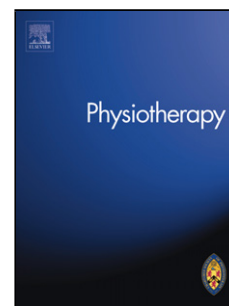
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## TITLE PAGE

**Title:** Physiotherapy management of joint hypermobility syndrome – a focus group study of patient and health professional perspectives.

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Accepted Manuscript

24 **TITLE**

25 Physiotherapy management of joint hypermobility syndrome – a focus group study of  
26 patient and health professional perspectives.

27

28

29 **ABSTRACT**

30 **Objective:** To develop an understanding of patient and health professional views  
31 and experiences of physiotherapy to manage joint hypermobility syndrome (JHS).

32 **Design:** An explorative qualitative design. Seven focus groups were convened,  
33 audio recorded, fully transcribed and analysed using a constant comparative method  
34 to inductively derive a thematic account of the data.

35 **Setting:** Four geographical areas of the UK.

36 **Participants:** 25 people with JHS and 16 health professionals (14 physiotherapists  
37 and 2 podiatrists).

38 **Results:** Both patients and health professionals recognised the chronic  
39 heterogeneous nature of JHS and reported a lack of awareness of the condition  
40 amongst health professionals, patients and wider society. Diagnosis and subsequent  
41 referral to physiotherapy services for JHS was often difficult and convoluted. Referral  
42 was often for acute single joint injury, failing to recognise the long-term multi-joint  
43 nature of the condition. Health professionals and patients felt that if left undiagnosed,  
44 JHS was more difficult to treat because of its chronic nature. When JHS was treated  
45 by health professionals with knowledge of the condition patients reported satisfactory  
46 outcomes. There was considerable agreement between health professionals and  
47 patients regarding an 'ideal' physiotherapy service. Education was reported as an  
48 overarching requirement for patients and health care professionals.

49 **Conclusions:** Physiotherapy should be applied holistically to manage JHS as a  
50 long-term condition and should address injury prevention and symptom amelioration  
51 rather than cure. Education for health professionals and patients is needed to  
52 optimise physiotherapy provision. Further research is required to explore the specific  
53 therapeutic actions of physiotherapy for managing JHS.

54 **Key Words:** Benign hypermobility syndrome, Ehlers-Danlos Syndrome,  
55 Hypermobility Type, Physiotherapy, focus groups, life experiences

56

57

## 58 **INTRODUCTION**

59 Musculoskeletal problems represent some of the most common reasons for seeking  
60 primary health care [1]. Joint hypermobility syndrome (JHS) is a heritable connective  
61 tissue disorder, characterised by excessive joint range of motion and symptoms of  
62 pain, fatigue, proprioception difficulties, soft tissue injury and joint instability [2].

63 Many experts now consider JHS to be indistinguishable from Ehlers Danlos  
64 Syndrome - Hypermobility Type (EDS-HT) [3]. This paper uses the term JHS.

65 Physiotherapy is generally the preferred management option, however, if patients  
66 are referred for an acute injury rather than for JHS, it is possible that physiotherapy  
67 could exacerbate symptoms [4].

68

69 Generalised joint laxity (often described as being 'double jointed') is very common  
70 and generally asymptomatic, occurring in 10-20% of Western populations, with  
71 higher prevalence in Indian, Chinese, Middle Eastern and African populations [5, 6,  
72 7]. JHS is thought to be under-recognised [8], although there is a lack of high quality  
73 epidemiological data on its true prevalence, complicated by the historical use of

74 different diagnostic criteria. The revised Brighton 1998 criteria are now  
75 recommended for diagnosis [9]. A key component of the Brighton criteria is the  
76 Brighton score, a nine-point score of joint mobility in clinical usage for many years  
77 [6]. One point is awarded for being able to place the hands flat on the floor whilst  
78 keeping the knees straight. One point is also awarded for left and right joints as  
79 follows: 10° knee hyperextension; 10° elbow hyperextension; 90° extension of the 5<sup>th</sup>  
80 finger metacarpophalangeal joint; and opposition of the thumb to touch the forearm.  
81 The Brighton criteria incorporate other clinical features to exclude other differential  
82 diagnoses. However, diagnosing JHS is often challenging, as symptoms may easily  
83 be attributed to other causes. Patients report a wide range of fluctuating symptoms in  
84 addition to pain, and it has been suggested that many patients presenting in primary  
85 care with everyday musculoskeletal conditions may have unrecognised JHS [10].  
86 Indeed use of the Brighton criteria has revealed that a very high prevalence of JHS  
87 in musculoskeletal clinics, with rates of 46% of women and 31% of men referred to  
88 one rheumatology service [11]; 30% of those referred to a Musculoskeletal Triage  
89 Clinic in the UK [12]; and 55% of women referred to physiotherapy services in Oman  
90 [13].

91  
92 Physiotherapy, particularly exercise, is the mainstay of treatment for JHS [13].  
93 However, there is little empirical evidence supporting its efficacy. Two recent  
94 systematic reviews included only a handful of eligible trials of physiotherapy and  
95 occupational therapy interventions for JHS and found limited evidence for their  
96 clinical and cost-effectiveness [14, 15]. The current lack of evidence on the most  
97 effective management options for JHS may contribute to anecdotally reported  
98 negative experiences of management [16, 17]. Higher quality multi-centre trials are

99 clearly required to investigate the clinical and cost effectiveness of physiotherapy for  
100 JHS. However, before such trials take place, there is a need to develop a clearer  
101 understanding of patients' and health professionals' attitudes towards, and  
102 experiences of, physiotherapy to manage JHS. Such information could help to inform  
103 the development of effective intervention packages. The study reported here  
104 therefore aimed to qualitatively explore patients' and health professionals' views on  
105 physiotherapy management of JHS.

106

107

## 108 **METHOD**

### 109 **Participants**

110 Seven focus groups were conducted between January and February 2013 in four UK  
111 locations. The purposive sampling strategy aimed for diversity with regard to  
112 professional discipline (for health professionals); socio-economic situation (for  
113 patients); and age, gender, and geographical location (for both groups). All  
114 participants were recruited via mailed invitations. Potential patient participants were  
115 identified as follows: 1) from clinical records at two NHS Trusts; 2) people with JHS  
116 who previously expressed interest in assisting with research at two Universities; 3)  
117 members of the Hypermobility Syndromes Association (HMSA) who lived locally to  
118 the same two Universities (identified by the HMSA). Eligible patients were aged 18  
119 or over, had previously received a diagnosis of JHS, had attended physiotherapy  
120 within the preceding 12 months and were able to speak English. Other known  
121 musculoskeletal pathology causing pain was an exclusion criterion. Potential health  
122 professional participants were identified by lead physiotherapists within the two NHS  
123 Trusts and by lead academic researchers from two Universities (including previous

124 attendees on courses relevant to JHS management). Eligible health professionals  
125 were post-qualification health professionals who had some interest or involvement in  
126 treating people with JHS. There were no specific exclusion criteria. Ethical approval  
127 was obtained from the North East NHS Research Ethics Committee (12/NE/0307)  
128 and all participants gave written consent.

129

### 130 **Procedure**

131 Focus groups were conducted in meeting rooms distant from clinical physiotherapy  
132 departments (to preserve confidentiality and facilitate open and honest discussion).  
133 The focus groups were facilitated by two researchers. One researcher (SP) led the  
134 discussion using open-ended questioning techniques to elicit participants' own  
135 experiences and views and to ensure all participants had an opportunity to take part.  
136 Another researcher (JH) summarised the discussion, audio-recorded the session  
137 and noted down who was speaking to aid transcription. Each focus group lasted  
138 between 71 and 100 minutes. Topic guides, developed and refined by the research  
139 team (including patient research partners), were used to facilitate discussions and, in  
140 line with an inductive approach, were revised in light of emerging findings. A further  
141 researcher (KR) attended the first patient focus group as an observer and  
142 contributed to subsequent refinement of the topic guides. Topic guides explored  
143 experiences of physiotherapy and views regarding education, advice, exercises and  
144 support. Separate focus groups were conducted with patients and health  
145 professionals.

146

### 147 **Data Analysis**



148 All focus groups were audio-recorded, transcribed, anonymized, checked for  
149 accuracy and then imported into a qualitative software package (NVivo 10) to aid  
150 data analysis. Thematic analysis [18], using the constant comparison technique [19]  
151 was used to identify and analyse patterns across the dataset. Transcripts were  
152 examined on a line-by-line basis with codes being assigned to segments of the data  
153 and an initial coding frame developed. An inductive approach was used to identify  
154 participants' perceptions of their experiences. To enhance analysis and enable team  
155 discussion and interpretation, team members (RT and JH) independently coded  
156 transcripts; any discrepancies were discussed to achieve a coding consensus and  
157 maximise rigour. Scrutiny of the data showed that data saturation had been reached  
158 at the end of analysis, such that no new themes were arising from the data [20]. All  
159 participants were assigned a letter as a pseudonym.

160

161

## 162 **RESULTS**

163 In total 4 focus groups were conducted with 25 patients (3 men and 22 women; aged  
164 19-60 years) and 3 focus groups with 16 health professionals (3 men and 13 women;  
165 0-30 years post qualification; 14 physiotherapists and 2 podiatrists) (Table 1). Three  
166 themes, developed from the analysis, related to: 'JHS as a difficult to diagnose,  
167 chronic condition' 'Physiotherapy to treat JHS' and 'Optimising physiotherapy as an  
168 intervention for JHS'.

169

### 170 **JHS as a difficult to diagnose, chronic condition**

#### 171 **The chronic, heterogeneous nature of JHS**

172 Both patients and health professionals described the chronicity of JHS and its  
173 symptoms. Patients recognised that they were *“going to have it forever”* [Female  
174 *patient E, age 19, FG6*] and that *“you won’t be fine, not completely”* [Female patient  
175 *C, age 40, FG1*]. Similarly, one health professional described having JHS as *“almost*  
176 *like a recovering alcoholic, you are always a recovering hypermobility person”*  
177 [Female health professional B, 28 years post-qualification, FG4]. The diverse nature  
178 of the symptoms was also noted by patients, that *“everyone with hypermobility has*  
179 *different symptoms”* [Female patient F, age 44, FG1] and by health professionals,  
180 who explained *“it’s the heterogeneous group that makes it very interesting”* [Female  
181 *health professional D, 22 years post-qualification, FG4*].

182

### 183 **Scepticism and lack of understanding amongst health professionals**

184 As joint laxity is sometimes perceived as an asset, and JHS symptoms fluctuate and  
185 vary, patients’ reports of problematic symptoms to health professionals were often  
186 met with scepticism.

187

188 *“... there’s still quite a prevalent view that it’s all in the mind, that [...] “I don’t*  
189 *believe in hypermobility” [...] it’s a kind of ... there are people who don’t feel it’s a*  
190 *genuine diagnosis, that it’s something psychological and you, you know, just need to*  
191 *be a bit braver.”* [Female patient A, age 60, FG2].

192

193 Both patients and health professionals therefore felt that JHS is not a widely  
194 understood or recognised condition amongst health professionals.

195

196           *“when I went back to physio for strengthening exercises to help my joints*  
197 *after the hypermobility diagnosis, there was ... I got that a little bit, I’m not sure*  
198 *about this hypermobility ...” [Female patient B, age 34, FG2].*

199

200           *“I work in a rheumatology department who don’t recognise joint hypermobility*  
201 *as an entity and in fact, probably a lot of people tend to get diagnosed with things*  
202 *like fibromyalgia more than normal” [Female health professional E, 30 years post*  
203 *qualification, FG3].*

204

205           Consequentially, health professionals perceived *“a lot of mismanagement”* of JHS by  
206 health professionals [Female health professional E, >20 years post qualification,  
207 FG4] and that patients may be given erroneous information by some health  
208 professionals. One patient described a rheumatologist who said, *“in his opinion, his*  
209 *professional opinion, that hypermobility doesn’t cause pain” [Female patient C, age*  
210 *53, FG2].* JHS trained health professionals felt that they were required to *“undo*  
211 *misconceptions, other health professionals’ understanding and what they have*  
212 *taught or implied to the patient about their condition. So for us we sort of have to*  
213 *unravel an onion so to speak, and it’s quite hard, yeah challenging I think” [Female*  
214 *health professional E, >20 years post qualification, FG4].*

215

216           Patients felt that JHS does not generally fit with health professionals’ models of  
217 acute injury and recovery and that this may be a source of frustration for health  
218 professionals.

219

220            “[physiotherapists] get frustrated because their model of physiotherapy and  
221            what they’re taught and how joints move and how they get better, hypermobility is  
222            totally the opposite of what they’re expecting and they can’t understand that. I’ve had  
223            physios before say ‘well stop the shoulder dislocating” [Female patient B, age 32,  
224            FG1].

225

## 226            **Diagnosis of JHS and subsequent referral**

227            The heterogeneous nature of JHS symptoms, lack of recognition of the syndrome  
228            and subjective diagnostic criteria were seen to contribute to often slow and  
229            convoluted diagnostic trajectories. Patients commonly remarked that “*it takes so*  
230            *many years to get diagnosed*” [Male patient E, age 36, FG5]. Health professionals  
231            highlighted the difficulties in diagnosing JHS using the criteria available.

232

233            “*I think it’s the diagnostic criteria for hypermobility syndrome that’s actually*  
234            *part of the problem [...] So it’s almost going right back to the start, finding a slightly*  
235            *more sensitive diagnostic criteria that can help us to then manage it*” [Female health  
236            professional, 11 years post-qualification, FG7].

237

238            For patients, receiving a diagnosis was considered essential in order to access  
239            appropriate treatment: “*the sooner you get the treatment the less likely it is that it is*  
240            *going to have such a great impact on your life*” [Male patient E, age 36, FG5].

241            However, health professionals felt that care pathways for JHS were not well defined  
242            and, as a result, patients may develop more complex problems or chronic pain  
243            issues.

244

245           *"I see the other end. I think we don't have a structured pathway of care for*  
246 *hypermobiles, which is what I'm interested in developing, but we don't have it. So*  
247 *there's no rheumatologist in the trust that has a special interest in hypermobility, and*  
248 *my God I've tried to find one [...] So there isn't a defined pathway of care for*  
249 *someone with generalised - with hypermobility syndrome, so"* [Female health  
250 *professional C, 25 years post qualification, FG4] .*

251

252           *"So for me I feel that's a key problem because I think we end up getting them*  
253 *too late, and if ((name)) had the support I feel to get these pathways better earlier"*  
254 *[Female health professional E, >20 years post qualification, FG4].*

255

## 256 **Physiotherapy to manage JHS**

### 257 **Physiotherapy for acute individual joint problems is unhelpful**

258 Physiotherapy is the mainstay treatment for JHS symptoms. However, both patients  
259 and health professionals emphasised that physiotherapy would not be effective if  
260 individual joints were treated in isolation and described difficulties in treating JHS  
261 within some National Health Service (NHS) constraints:

262

263           *"Because of, I think, the way – at least in my experience – that the NHS*  
264 *seems to approach things, they have a sort of, 'you're here for one joint' approach,*  
265 *which is quite difficult, because you go: 'Well, I'm floppy all over,.'. And then you*  
266 *have to have the conversation about 'Well, which is the most difficult?' You're like*  
267 *'Well, it's kind of all related', so if, like, if my knee is stronger and I'm doing less weird*  
268 *things with my knee, then my hip will feel better because - and I can say that, and to*  
269 *me it's obvious, that if you fix - just because it's your hip that hurts it doesn't mean*

270 *that it is actually the problem. It could well be that your knee is the issue, making you*  
271 *do weird things with your hip, but there's this, 'This is the joint, and we will deal with*  
272 *this joint,' when that isn't really ..."* [Female patient C, age 53, FG2].

273

274 Patients and health professionals reported that in the NHS, 'usual care' was normally  
275 up to six physiotherapy sessions to treat a specific joint. However, it was felt that this  
276 was not necessarily appropriate for JHS.

277

278 *"They've got us as their clinical leads telling them to look at people globally,*  
279 *pick up this diagnosis, but then they've got their managers telling them you have to*  
280 *do six sessions [...] I should really be saying "I know you've got hypermobility, I*  
281 *know it's all related, but actually I need six sessions with your back, I need six*  
282 *sessions with your shoulder and I need six sessions with your knee, and we need to*  
283 *negotiate that with your PCT because otherwise ((place name)) is not going to get*  
284 *paid"* [Female health professional E, 30 years post qualification, FG4].

285

286 In all focus groups, the need for continuous, ongoing access to physiotherapy was  
287 highlighted. One patient felt: *"the difficulty is, it's a chronic condition and the only*  
288 *time you are actually able to access any care in the NHS is when you have an acute*  
289 *incident from it"* [Female patient G, age 48, FG2]. Health professionals, unless  
290 practicing privately, were equally frustrated by the lack of flexibility in the number of  
291 treatment sessions that could be offered.

292

293           *“And I think the limitations of, like, if you were receiving NHS treatment, then*  
294 *you’re only going to get so many sessions” [Female health professional D, newly*  
295 *qualified, FG3].*

296

297 In addition to the perceived limited number of sessions, physiotherapy may also be  
298 unsuitable and exacerbate symptoms if it ignores the complexity of JHS symptoms:

299

300           *“Then, as you say, being given some more exercises that weren’t helpful*  
301 *because they did seem to cause more pain which then sets you back even more and*  
302 *then you seem to get into the cycle of never sort of making any progress and then*  
303 *the treatment’s over because you only get a few sessions” [Female patient G, age*  
304 *48, FG2].*

305

306 Although most patients described negative experiences of physiotherapy, for  
307 example when being referred without a diagnosis of JHS, once patients had been  
308 diagnosed and referred to JHS trained physiotherapists, many participants reported  
309 beneficial treatment.

310

311           *“I was originally seen by a physio who hadn’t diagnosed with the*  
312 *hypermobility and then went back to a musculoskeletal specialist who then put me*  
313 *forward to specialist hypermobility physiotherapist and since then it’s been amazing I*  
314 *feel like it’s been worthwhile and it felt like the right thing to do and I’ve been really*  
315 *enjoying it” [Female patient B, age 27, FG 5].*

316

317 **Physiotherapy is less effective if diagnosis is delayed**

318 Both physiotherapists and patients recognised that if JHS remained undiagnosed,  
319 chronic pain may develop which may be less responsive to physiotherapy. The  
320 biopsychosocial impact of living with untreated or inappropriately treated  
321 symptomatic hypermobility may lead to a more multidisciplinary approach being  
322 required.

323

324 *“And you see by the time - for me they come with quite a lot of psychological*  
325 *baggage, and you know, they are difficult patients. And then you’re trying to unravel*  
326 *what’s the primary and secondary issue here, is it that your mental health is actually*  
327 *what’s driving your hypermobility, or is it the fact you have such debilitating joints is*  
328 *making you mentally unwell. But by the time they get to us that’s so hard to deal*  
329 *with, [...] and they almost then, it’s a cry for help. So they’re desperate to get help*  
330 *so the psychological side comes out because the physical manifestation of what*  
331 *they’re suffering with is just so severe” [Female health professional E, >20 years post*  
332 *qualification, FG4].*

333

334 *“actually, there’s some that do quite well [with physiotherapy] as well in terms*  
335 *of .... especially I think if you catch them early, really the key is, before they develop*  
336 *a lot of the chronic pain” [Male health professional B, 8 years post qualification, FG*  
337 *7].*

338

339 Patients also recognised that delays in diagnosis may result in maladaptive  
340 responses to JHS, for example, compensatory postures, which are then difficult to  
341 rectify.

342



343           *"I was 15 when I was diagnosed and that was even too late really for me*  
344 *because the way I stand, the way I move, everything, my Pilates teacher - her*  
345 *grandson was 3 when he was diagnosed and he has Pilates, and physiotherapy now*  
346 *so he will get into habits of a life time"* [Female patient G, age 30, FG 1].

347

#### 348 **Optimising physiotherapy to manage JHS**

349 All focus groups were able to provide descriptions of an 'ideal' physiotherapy  
350 intervention or suggested improvements based upon their own experiences of giving  
351 or receiving treatment. Health professionals' and patients' descriptions of ideal  
352 physiotherapy were notably similar (Table 2). Both felt it was important to have  
353 continuity of therapist, who was trained in JHS and who provided reassurance. Both  
354 patients and health professionals described the importance of flexible treatment  
355 delivery; patient led treatment that meets and manages goals and expectations;  
356 taking a holistic, long term approach; and treating JHS rather than its acute  
357 manifestations. The importance of ongoing, 'maintenance' physiotherapy was also  
358 highlighted.

359

#### 360 **Central role of education in managing JHS**

361 Both patients and health professionals considered education to be key to optimising  
362 physiotherapy for JHS.

363

364 ***Education for health professionals:*** many patients felt that education was  
365 required for health professionals.

366

367           *“I think actually it’s the health professionals that need education [...] I mean*  
368 *there’s lots of things I still need to know about hypermobility but on the flip side I do*  
369 *think it’s the health professionals that need to know more” [Female patient G, age*  
370 *42, FG 5].*

371

372 Because of the lack of understanding that patients perceived to be common amongst  
373 health professionals, some felt that they were providing education for the health  
374 professionals, and that this was not necessarily beneficial for them.

375

376           *“So there’s this odd situation where I’m explaining how it works to them and I*  
377 *think that it isn’t ideal and I think there does need to be better education for the*  
378 *physios because I think that is quite important that they tell you how and why things*  
379 *are happening to you, rather than vice versa because that’s unhelpful” [Female*  
380 *patient E, age 21, FG2].*

381

382 Health professionals also highlighted the need for better education and suggested a  
383 variety of educational sources, including websites, special interest and support  
384 groups and further professional training. One health professional highlighted the  
385 value of evidence based guidelines: *“because if you get a patient in front of you, you*  
386 *need to be able to think, okay, what can I look at? What is the most effective? So*  
387 *guidelines that you were talking about, or maybe you can do, would be very helpful”*  
388 *[Female health professional E, 30 years post qualification, FG3].*

389

390 **Education for patients:** Health professionals felt that patient education was  
391 necessary to facilitate a greater understanding of the condition.

392

393 *“I think a large part of it, as well, is to the education. To think that the patients*  
394 *don’t necessarily understand the condition. [...] Sometimes they don’t actually,*  
395 *nobody has never actually sat down and explained to them what that is and the*  
396 *implications. And what can actually be done to help them. So I think that’s a large*  
397 *part of it” [Female health professional D, newly qualified, FG3].*

398

399 Health professionals felt that education is necessary for patients to develop realistic  
400 expectations of treatment and a better understanding of the rationale for particular  
401 treatment plans.

402

403 *“A lot of ... I think what is ... is education, “this is why I’m doing it”, and*  
404 *making sure they understand why I’m getting them to do these exercises [...] ...*  
405 *even if it doesn’t work and goes horrendously wrong, that’s fine, we can change that,*  
406 *but they’ve got to have an understanding of what we’re asking them to do and why*  
407 *we’re asking them to do it” [Male health professional B, 8 years post qualification,*  
408 *FG7].*

409

410 Patients similarly recognised that education helped them to fully engage with  
411 treatment.

412

413 *“because I kind of understand and have an interest in it, I think it makes it*  
414 *really easy and go really quick so I suppose it’s where someone who doesn’t really*  
415 *know about it, they’ve got to learn about it first because you can’t tell someone to do*  
416 *it if they don’t understand it” [Female patient D, age 21, FG1].*

417

418 **Measuring success, and managing expectations, of physiotherapy**

419 All participants recognised the aim of physiotherapy was to manage, rather than  
420 cure, the symptoms of JHS; that ‘successful’ therapy did not mean being pain free;  
421 rather, the aim was for the patient to be able to manage their pain.

422

423 *“I think measuring success should be more about reaching a point of*  
424 *continuity where you know you might not be great all the time or you might not be*  
425 *really bad all the time but you’re manageable” [Female patient G, age 30, FG1].*

426

427 *“... you may not be expecting to get them pain free, but if they’re happy and if*  
428 *they’re managing the problem better, you know what to do to manage it, then you’re*  
429 *there” [Female health professional C, 19 years post qualification, FG3].*

430

431 However, some health professionals raised concerns about patient expectations;  
432 that patients were expecting to gain more than they could realistically offer. For  
433 example, one health professional felt that patients often wanted, and expected, a  
434 ‘cure’.

435

436 *“I don’t want them to go away and think, well, she’s done nothing, when they*  
437 *expected me to fix it. So I have to say from the beginning, well, I can’t fix it, but this*  
438 *is what I can do. And to a point, that’s all you can do, isn’t it, really?” [Female health*  
439 *professional E, 19 years post qualification, FG7].*

440

441 Some patients considered that physiotherapy would be successful if it resulted in  
442 some reduction in pain intensity, in some parts of their body. But contrary to some  
443 health professionals' perceptions, patients did not appear to hold unrealistic  
444 expectations about treatment:

445

446 *“You can measure it [i.e. the success of physiotherapy] by parts of body I*  
447 *guess because I, although I don't feel remotely better in many parts I still say that my*  
448 *last physiotherapy was a success because it significantly helped me with my*  
449 *shoulders so that I, I like suffer a lot less pain in that area of the body now, so I call it*  
450 *a success but when you get to my knees and ankles and neck and back it did do that*  
451 *much, the neck surgery was a success because that significantly reduced the neck*  
452 *pain although I still get probably more muscular now than any joints but that's still*  
453 *again one part of it, so there's lots of other areas that are still very bad, so erm I*  
454 *guess that in order to say that I'm better every bit would have to have improved*  
455 *significantly to say that they didn't affect my day to day life, but to have individual*  
456 *parts improve is still a success” [Female patient F, age 19, FG5].*

457

458

## 459 **DISCUSSION**

460 This is the first in-depth qualitative exploration of patients' and health professionals'  
461 perspectives on physiotherapy for JHS. As such, it provides invaluable information to  
462 help reflect upon and enhance management of this complex long term  
463 musculoskeletal condition.

464

465 Both patients and health professionals described JHS as a painful, chronic condition  
466 with heterogeneous and evolving symptoms, in line with other empirical research  
467 [21]. Patients and health professionals reported a lack of recognition and  
468 understanding about JHS and even some scepticism. Patients reported difficulties in  
469 being diagnosed and how they had encountered health professionals who they felt  
470 didn't believe or understand their descriptions or experiences of JHS [22]. Both  
471 patients and health professionals recognised that a diagnosis was essential in order  
472 to facilitate effective treatment. Previous research has similarly referred to the  
473 significance and the sense of relief for patients when a diagnosis is received  
474 following many years of frustration and searching for a reason for their symptoms  
475 [23]. Recent surveys have also highlighted the need for further education to improve  
476 recognition, diagnosis and management [24, 25].

477

478 Although physiotherapy is the mainstay treatment for JHS, there is a lack of  
479 empirical evidence to indicate the optimum type, frequency or means of delivering  
480 physiotherapy interventions [26]. In the current study, participants indicated that the  
481 success of physiotherapy appears to be dependent upon having a prior diagnosis of  
482 JHS and receiving physiotherapy from a therapist trained in JHS. Recent surveys of  
483 physiotherapists treating adults with JHS identified that between 68% and 51%  
484 reported receiving no training in JHS [24, 25]. Only 9.8% received undergraduate  
485 training in hypermobility [24]. Development of appropriate learning opportunities and  
486 resources for health professionals would seem warranted. Health professionals and  
487 patients also highlighted the importance of early diagnosis and intervention to  
488 prevent the establishment of maladaptive postural habits or movements. It is  
489 possible that many symptoms of JHS could be prevented or ameliorated by

490 addressing issues such as joint control (posture, motor control, muscle  
491 strengthening, and proprioception), education, physical activity and physical fitness  
492 [27]. On the other hand, our focus group data suggest that once chronic pain had  
493 developed, JHS management may become much more complex due to its  
494 substantial psychological impact [15]. Moreover, as previous research has implicated  
495 acute pain episodes in the subsequent development of chronic pain [10], further  
496 research is required to investigate the extent to which repeated acute pain episodes  
497 influence chronic pain development. The extent to which other variables influence  
498 the efficacy of physiotherapy also requires further exploration (for example age;  
499 severity and duration of pain symptoms; the degree of joint hypermobility and  
500 instability; psychological dysfunction [15]; and concurrent conditions such as postural  
501 tachycardia syndrome [28], dysautonomia [29] and gastro-intestinal dysfunction  
502 [30]). Further research is required to assess the value of physiotherapy and a  
503 feasibility trial is underway to investigate the acceptability of a tailored physiotherapy  
504 programme for JHS (ISRCTN29874209).

505

506 There were many similarities between patients' and physiotherapists' descriptions of  
507 an 'ideal' physiotherapy service (Table 2). Their descriptions also reflected 'best  
508 practice' in some physiotherapy services specialising in JHS. Whilst some health  
509 professionals felt that patients may hold unrealistic expectations of the extent to  
510 which physiotherapy could help, in fact, patients in our focus groups recognised that  
511 their condition would never be cured, and that amelioration of their symptoms was  
512 the most that they could hope for.

513

514 There was a consensus from participants that patients would benefit from health  
515 professionals who understood JHS and its complexities. A central aim of  
516 physiotherapy should be to equip the patient to manage JHS over the life course and  
517 education was seen as the most salient factor to facilitate a correct and timely  
518 diagnosis, to raise awareness within society and to enable those with the condition to  
519 maximise their function.

520

### 521 **Limitations and strengths**

522 A particular strength of the research is the fact that data were gathered from both  
523 patients and health professionals, allowing a clearer understanding of views  
524 regarding physiotherapy for JHS. Employing focus group methodology allowed  
525 consensus to be gained regarding physiotherapy treatment, although it is recognised  
526 that focus groups do not permit as much in-depth exploration of issues as other  
527 forms of data collection such as interviews. Greater diversity in health professional  
528 perspectives would have been welcome. Unfortunately an occupational therapist,  
529 osteopath and rheumatologist who expressed an interest in taking part were  
530 subsequently unable to attend the focus groups.

531

532 The congruence between patients' and health professionals' descriptions and  
533 perceptions of JHS was notable. Whilst this is encouraging, it should be noted that  
534 the health professionals in these focus groups were experts in the field, providing  
535 specialist care for JHS. Further research is required to understand the perceptions  
536 and experiences of other health professionals and to develop an understanding of  
537 any potential barriers to providing appropriate care.

538



539 **Conclusion and future directions**

540 Physiotherapy is likely to be helpful for JHS, but may be more beneficial if used to  
541 manage JHS holistically rather than to treat acute injuries in isolation. Physiotherapy  
542 services need to recognise the chronic nature of JHS, and the aim of physiotherapy  
543 should be long term injury prevention and symptom amelioration. It appears that  
544 physiotherapy may be particularly beneficial for JHS patients who have not  
545 developed chronic pain syndromes. For JHS patients with chronic pain,  
546 physiotherapy may also be valuable, but treatment is more complex and may require  
547 input from a multidisciplinary pain service. Education for health professionals and  
548 patients and raising awareness of the condition is essential to optimise  
549 physiotherapy provision for JHS. Research is required to explore the specific  
550 therapeutic action of physiotherapy and its role within the wider multidisciplinary  
551 team.

552  
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564

565

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646

647 **Table 1. Participants' demographic characteristics.**

<b>Patients (total n=25)</b>		<b>N (%)</b>
Age	18-29	8 (32)
	30-39	7 (28)
	40-49	6 (24)
	50-59	2 (8)
	>60	3 (12)
	mean, (median)	33 years, (36)
Gender	Female	22 (88)
	Male	3 (12)
Ethnicity	'White'	23 (92)
	'Other'	2 (8) (both self-reported as 'British White and Chinese')
Socio-Economic Status (SES)*	1 (affluent)	8 (32)
	2	8 (32)
	3	4 (16)
	4	3 (12)
	5 (most deprived)	1 (4)
Education	Schooling to 16 yrs	3 (12)
	College diploma/equivalent	6 (24)
	University degree/equivalent	10 (40)
	Post graduate degree	6 (24)
Employment	Employed full time	7 (28)
	Employed part time	8 (32)
	Student full time	4 (16)
	No paid job	5 (20)
	Retired	1 (4)
<b>Staff (total n=16)</b>		<b>N (%)</b>
Gender	Female	13 (81)
	Male	3 (19)
Role	Physiotherapists	14 (88)
	Podiatrists	2 (13)
Years since qualifying	Newly qualified	1 (6)
	≤5 years	1 (6)
	6-20 years	7 (44)
	>20 years	7 (44)

648 \* Measured as Index of Multiple Deprivation (IMD) quintile from home post code  
649 (Source: Office for National Statistics)

650

651

**Table 2. Suggestions for an 'ideal' physiotherapy service.**

<b>Suggested improvements</b>	<b>Illustrative excerpt from patient</b>	<b>Illustrative excerpt from health professional</b>
<b>Regarding therapist</b>		
Continuity of therapist to improve patient-therapist interaction/relationship	<i>"They get to know you as well, don't they, and they know your lifestyle and they know what you do day in day out and therefore they can start to understand any triggers, ... they get to know you as a person" [Female patient G, age 30, FG1].</i>	<i>"For everybody, all patients, is continuity. But it's especially difficult [for JHS patients] because they have so many different problems" [Male health professional A, 6 years post qualification, FG3].</i>
Therapist should be JHS expert	<i>"... the two physiotherapists I've had who've known about [erm] hypermobility have been a lot better than ones I've had in the past where they obviously haven't had a clue" [Female patient C, age 60, FG 6].</i>	<i>"... if they see somebody who hasn't had an interest in that then they're learning along with the patient at the same time. ... So that's quite difficult. It's much better, isn't it, to be seen by a specialist straight away who has got a broader knowledge base to be able to tap into their tools and skills" [Female health professional E, 30 years post qualification, FG 3].</i>
Therapists should provide reassurance and encouragement	<i>"quite often I'll come out of the next physio feeling much happier because they've reassured me that it's not the end of the world and you know sometimes you have a bad week but it doesn't mean that you won't then have a good week" [Female patient F, age 44, FG1].</i>	<i>"I think you've got to set achievable goals, then you've got to give a lot of reassurance and positive feedback" [Female health professional B, 28 years post qualification, FG 4].</i>
<b>REGARDING Physiotherapy</b>		
Flexibility in treatment, (e.g. number of sessions, content, specific techniques, mode of delivery, structure and focus)	<i>"... Or consider the person's life style, ... and that sort of flexibility, not just on what they're asking the patient to do, even being flexible on the times of day or you know when these things can happen, you know make it</i>	<i>"Ideally, you'd want to have a service offer where they could tap into the service where they wanted to. If they suddenly got a flare up of something, say their hands started to give way or become more of a</i>

	<i>interesting, you know we can't all get in at 11 o'clock in the morning or 2 o'clock in the afternoon, we do need the half past 7's the 8 o'clock in the morning, and the evening appointments" [Female patient C, age 40, FG1].</i>	<i>problem, then they could come back to you" [Female health professional E, 30 years post qualification, FG3].</i>
Patient led treatment, whilst managing and understanding patient expectations.	<i>"I think being patient led, ... what it is that they want to achieve out of it and how the best way they can do that, and you know with a bit of guidance, like..." [Female patient B, age 32, FG 1].</i>	<i>"You try and tease out, you know, what are your expectations? No idea. So your hopes? No idea. I don't know what I'm supposed to be doing ... Forget that, what would you like to be doing? .... Then you start to offer things and start to treat or start to address ..." [Male health professional D, 5 years post qualification, FG 7].</i>
Meeting individual goals, to manage rather than cure	<i>"Or consider the person's life style, you know consider what is going to be feasible, what they need to be able to get to in terms of achievement and you know and that sort of flexibility not just on what they're asking the patient to do ..." [Female patient C, age 40, FG1].</i>	<i>"Because we're very good at having goals, but you know, it's making sure that the patients, they are the patients' as well" [Female health professional G, 23 years post qualification, FG4].</i>
Holistic, long term approach	<i>"It's not just your joints, it is all the other bits around it and that sort of slightly bigger picture, you're probably going to be like this always, you need to think of different ways to manage different things" [Female patient E, age 34, FG2].</i>	<i>"... obviously if there's a mechanical element to it we'd have to go into that, but as I say, the hypermobility is something that needs to be addressed more holistically" [Female health professional E, 19 years post-qualification, FG7].</i>
Recognition of the need to treat multiple joints for JHS rather than individual problematic joints	<i>"I think they need to take notice that it is a full body condition rather than just individual, rather than just like one area, it is individual parts but they often concentrate on one area and then forget that the rest of the body hurts as well and that the pain can be</i>	<i>"If it was classified as a condition, [unclear 31:00] spondylitis or all those other rheumatological conditions which are, extend beyond one section, it's treated differently isn't it, so it's got to do with its recognition presumably. It's</i>



	<i>interlinked” [Male patient E, age 36, FG5].</i>	<i>multi systemic, therefore you can treat multiple sites and therefore it may take longer in the end” [Female health professional D, 22 years post qualification, FG 4].</i>
Focus on core strengthening and ‘correct’ movement	<i>“basically you’ve really got to give them a comprehensive set of useful exercises that will cover a whole range of joints, you know because most of our joints are affected, but particular core stability” [Female patient E, age 44, FG1].</i>	<i>“but really just concentrating on ... on kind of core, and ... good posture .. concentrate on how they’re exercising, what they’re doing, technique rather than just exercising. Because a lot of them just ... they find the most bizarre ways of doing things that I could never do in a million years” [Male health professional B, 8 years post qualification, FG7].</i>
Maintenance physiotherapy for a chronic condition rather than acute problems arising from JHS	<i>“If it’s like say the diabetic clinic, where you get called every year to see them. ... So could they not do a package where you actually went back every six months to see somebody regardless of how you were feeling” [Female patient A, age 60, FG2].</i>	<i>“So what we’ve tried to do is ...a sort of self-referral back into the service, so they’re not having to go round the houses, and we pick them up quickly when they’re starting to get a flare up or a deterioration” [Female health professional E, &gt;20 years post-qualification, FG4].</i>

652