

**Full title:** Diagnosis, management and assessment of adults with joint hypermobility syndrome:  
a UK-wide survey of physiotherapy practice

**Short title:** Survey of physiotherapy for joint hypermobility

**Word count:** 3,167

**Names and affiliations of all authors:**

Shea Palmer<sup>1</sup>, Fiona Cramp<sup>1</sup>, Rachel Lewis<sup>2</sup>, Shahid Muhammad<sup>1</sup>, Emma Clark<sup>3</sup>

<sup>1</sup>Department of Allied Health Professions, Faculty of Health & Applied Sciences, Glenside Campus, Blackberry Hill, Bristol, UK, BS16 1DD

<sup>2</sup>Department of Physiotherapy, North Bristol NHS Trust, Southmead Hospital, Westbury-on-Trym, Bristol, UK, BS10 5NB

<sup>3</sup>Musculoskeletal Research Unit, University of Bristol, Southmead Hospital, Westbury-on-Trym, Bristol, UK, BS10 5NB

**Corresponding author:**

*Address:* Professor Shea Palmer, Professor of Musculoskeletal Rehabilitation, Department of Allied Health Professions, Faculty of Health & Applied Sciences, Glenside Campus, Blackberry Hill, Bristol, UK, BS16 1DD.

*E-mail:* [Shea.Palmer@we.ac.uk](mailto:Shea.Palmer@we.ac.uk)

*Tel:* +44 (0)117 3288919

*Fax:* +44 (0)117 3288437

**Sponsor of the research:** This research was supported by a grant from the Faculty of Health & Applied Sciences, University of the West of England, Bristol.

**ABSTRACT (248 words)**

Joint Hypermobility Syndrome (JHS) is a heritable disorder associated with excessive joint range of motion and pain in the absence of inflammatory joint disease. It is a relatively common cause of musculoskeletal pain but is generally understood to be under-recognised and poorly managed in clinical practice. This study therefore aimed to identify how JHS is diagnosed, managed and assessed in routine physiotherapy practice. A survey tool was developed from similar physiotherapy surveys of musculoskeletal practice, a review of the literature, and consultation with researchers and clinicians. Paper copies of the final survey were sent to 201 randomly selected secondary care organisations across the UK and an electronic version was advertised through physiotherapy professional networks. A total of 66 responses (80% women) were received from physiotherapists with a wide range of clinical experience. Only 32% of respondents reported that they had received formal training in JHS management. The Brighton diagnostic criteria for JHS were not often used (only 31% of respondents). The stated aims of physiotherapy and the specific interventions employed seem well matched, with a focus on advice, education, exercise and self management. Although pain relief was not reported as a high priority in terms of treatment aims, pain was most often assessed as an outcome, suggesting a mismatch between what clinicians aim to achieve and what they measure. The results suggest that reported management strategies are broadly appropriate to long term musculoskeletal conditions but additional training specific to JHS may be required, particularly in diagnosis and assessment.

## INTRODUCTION

Joint Hypermobility Syndrome (JHS) is a heritable connective tissue disorder associated with excessive joint range of motion, often at multiple joints, in the presence of pain (Ferrell et al. 2004). JHS is reported to be under-recognised, poorly understood and poorly managed in clinical practice (Hakim and Grahame 2004, Grahame 2000). However it is a common source of musculoskeletal pain, with symptomatic joint hypermobility reported to affect approximately 5% of women and 0.6% of men (Simpson 2006). It should be acknowledged, however, that there is currently a lack of good quality epidemiological evidence for the true prevalence of JHS in the general population. When compared with healthy controls, JHS has been shown to have a significant impact on a wide range of outcomes such as exercise endurance, gait, pain, proprioception, strength, function and quality of life in both children (Engelbert et al. 2006, Fatoye et al. 2009, Fatoye et al. 2011, Fatoye et al. 2012) and adults (Hall et al. 1995, Mallik et al. 1994, Sahin et al. 2008a, Sahin et al. 2008b).

Physiotherapy is considered the mainstay of treatment (Hakim and Grahame 2004, Keer and Grahame 2003, Simmonds and Keer 2007, Simmonds and Keer 2008, Tinkle 2008, Keer and Simmonds 2011) and professionals within a number of centres in the United Kingdom (UK) have developed a specialist interest in treating people with JHS. The development of such specialist services is to be welcomed but we currently know very little about how JHS patients are diagnosed, assessed and managed by physiotherapists in routine clinical practice. This knowledge is necessary to inform the future development of education and research in this area.

The revised Brighton 1998 criteria are now recommended for the diagnosis of JHS (Grahame et al. 2000), although a range of other diagnostic criteria have been used historically. A key component of the Brighton criteria is the Beighton score, a nine-point score of joint mobility which has been in clinical usage for many years (Beighton et al. 1973). One point is awarded for being able to place the hands flat on the floor whilst keeping the knees straight (assessing lumbar spine mobility). One point is also awarded for each hypermobile peripheral joint as

follows: 10° knee hyperextension; 10° elbow hyperextension; 90° extension of the 5<sup>th</sup> finger metacarpophalangeal joint; and opposition of the thumb to touch the forearm (points are awarded for the left and right limbs as appropriate). The Brighton criteria incorporate a number of other clinical features to confirm a diagnosis of JHS and exclude other differential diagnoses. Greater consistency in the criteria being used for diagnosis is required to facilitate research and clinical practice in this area. It would therefore be helpful to determine to what extent the Brighton criteria have been adopted in routine physiotherapy practice and whether other historical diagnostic criteria persist.

Keer and Simmonds (2011) reported that pain relief and preventing recurrence of joint pain are the main aims of treatment, with exercise key to achieving these aims. They reported research evidence supporting the importance of interventions targeting posture, proprioception, strength and motor control, in conjunction with education, physical activity and fitness. A recent systematic review of evidence for the effectiveness of exercise for JHS (Palmer et al. 2014) concluded that, whilst there is some evidence that people with JHS who receive exercise interventions improve over time, there is little convincing evidence for the effectiveness of different forms of exercise or for exercise being more effective than a control condition. Greater knowledge of what treatment interventions are being implemented in routine physiotherapy practice, and how these relate to recommendations from experts in the field and to existing research evidence will help to underpin future research regarding optimal interventions.

Outcome assessment is key to identifying the success (or otherwise) of treatment. As evidenced by our recent systematic review of the effects of exercise in JHS (Palmer et al. 2014), a wide range of objective and patient reported outcome measures have been used in previous research. These include specific measures of pain at rest and on movement; function; balance; quality of life; strength; proprioception; and range of motion. It would be helpful to know what specific outcome measures are being used in clinical practice, how these relate to those used in previous research, and how they reflect the aims of physiotherapy.

To address these questions a UK-wide survey was undertaken to identify 'usual' physiotherapy practice related to the diagnosis, management and assessment of adults with JHS.

## **METHODS**

### **Participants**

The aim was to generate data from a wide range of musculoskeletal practice settings. A paper version of the survey was distributed to physiotherapists from a random selection of National Health Service secondary care organisations across the UK. It was decided not to include primary care organisations due to major ongoing changes in the organisation and delivery of primary care services in England at the time of the survey. The paper survey was supplemented by an online version distributed through 'interactiveCSP', an online resource hosted by the Chartered Society of Physiotherapy (CSP). This strategy aimed to recruit physiotherapists from a slightly wider range of practice settings, including primary care and private practice.

### **Procedures**

The survey was designed following close consideration of previous surveys of physiotherapy practice in a range of other musculoskeletal conditions (French 2007, Walsh and Hurley 2009, Hanchard et al. 2011, Artz et al. 2013). The specific aims of the current survey were considered and key features of the previous tools were selected and adapted to address those aims. A wide range of textbooks, scholarly articles, reviews and original research related to the diagnosis, assessment and management of JHS were also consulted to inform the selection of specific survey items. A draft survey was developed on this basis and this was distributed to and commented upon by members of the research team. Subsequent drafts of the survey were also distributed to physiotherapists in two local rheumatology physiotherapy services. A total of four drafts of the survey were developed and refined in this way, before the final fifth version was agreed upon. The final version of the survey addressed the following main areas:

- Participant characteristics
- Description of the physiotherapy service
- Diagnostic criteria used

- Aims of physiotherapy
- Specific interventions used
- Outcome measures used

The precise wording of individual questions is included in the presentation of the results.

All secondary care NHS organisations in each UK nation (England, Northern Ireland, Scotland and Wales) were identified from publicly available lists. A representative number (based on population of each of the 4 UK home nations) was then randomly selected from this list. A total of 201 paper copies of the survey were distributed to the 'Lead Physiotherapist (Musculoskeletal)' at each organisation as follows: England = 168, Northern Ireland = 6, Scotland = 17 and Wales = 10. Although surveys were addressed to lead physiotherapists, the accompanying letter contained the following statement: *"If you are not personally interested in taking part, please feel free to distribute this information to a colleague who you think is most relevant"*. An online version of the questionnaire was also developed using an online survey tool ([www.surveymonkey.com](http://www.surveymonkey.com)). News items were posted to two separate online discussion networks on 'interactiveCSP'. The total number of CSP members subscribed to each of the networks (as of 17<sup>th</sup> June 2013) was 'Musculoskeletal' (n=8,835) and 'Research' (n=5,021). An identical news item and link to the online survey was posted on each Network. It should be noted that many CSP members subscribe to more than one Network and the level of interaction with the online resource varies between individual members. It is therefore not possible to determine how many members were accessed through this method.

### **Data Analysis**

Data were entered into IBM SPSS Statistics 20 and analysed using descriptive statistics (the proportion of valid responses).

### **ETHICAL APPROVAL**

The project was approved by the Faculty of Health & Applied Sciences Ethics Subcommittee, University of the West of England, Bristol (HLS/13/05/67).

## **RESULTS**

A total of n=66 responses were received (n=61 paper responses and n=5 online responses). Two unopened paper questionnaires were returned to the researchers, both with incorrect addresses. Table 1 below presents the characteristics of survey participants. Participants were predominantly women, employed at Band 7 or above (equivalent to a specialist senior physiotherapist), with more than 10 years' experience. There was a relatively even split between full-time and part-time working. The vast majority had not received any formal training in JHS management and saw ten or less people with JHS in the last year. Most worked in secondary care (63%) but a sizeable proportion (44%) worked in primary care (with some working in both).

Table 2 contains information about the clinical services offered to people with JHS, as well as details of the diagnostic criteria that physiotherapists employ. Most referrals are received from general practitioners (GPs) and rheumatology consultants and most patients are seen within two months. All patients are offered one-to-one treatment, although 16% of respondents also offer a group intervention. Initial assessment is generally an hour or less, with six or fewer treatment sessions (each lasting 30 minutes or less) and treatment typically lasting a maximum of four months. The majority of participants work as part of a multidisciplinary team, with Occupational Therapy and Podiatry most commonly being available. There was no consensus on whether people with JHS should exercise in a restricted range of movement (41%) or throughout their available range (43%). The Brighton diagnostic criteria are relatively infrequently used (31%), with the Beighton score predominating (87%), although this was often used in conjunction with family history of JHS and pain. Interestingly, those who reported that they had received formal training in JHS management were more likely to use the Brighton criteria (43%) than those without training (22%).

The aims that physiotherapists report when they are managing people with JHS are presented in Table 3. Aims addressing education, self management, exercise, muscle control, posture/ergonomics and joint protection predominated. Decreasing pain was considered relatively less important. Table 4 reports the frequency with which physiotherapists use specific interventions for people with JHS, with advice, education, self-management, exercise and posture being most prevalent. 'Passive' interventions such as electrotherapy modalities, injection and acupuncture were relatively rarely employed. Finally, details of the frequency with which physiotherapists use specific outcome measures for people with JHS are detailed in Table 5. Pain scales were most common, whilst validated functional and quality of life instruments such as the EQ5D (The EuroQol Group 1990) or SF12/SF36 (Ware 2000) were less likely to be employed.

## **DISCUSSION**

The survey captured information from physiotherapists with a wide range of characteristics and experience (Table 1). 80% of participants in this survey were female and this compares very favourably with a Centre for Workforce Intelligence review of the physiotherapy workforce in England (CfWI 2010) which reported 84% female. Extrapolation of data available from graphs in that workforce review (CfWI 2010) indicated that physiotherapists within the NHS in England were employed at bands 5, 6, 7 and 8 in the approximate proportions of 20%, 34%, 33% and 12% respectively. Similarly, a recent UK-wide Chartered Society of Physiotherapy survey (CSP 2011) indicated that musculoskeletal outpatient physiotherapists were most frequently employed at band 6, followed by bands 7, 5, 8a and 8b. Data from our survey indicated that more than 77% of respondents were employed at Band 7 or above, suggesting that respondents may have been slightly more experienced than the general physiotherapy or musculoskeletal outpatient physiotherapy workforce. This is supported by the observation that more than 71% of respondents were qualified for 11 years or more. This may have inadvertently been a product of addressing the paper-based surveys to lead physiotherapists, but it might



also suggest that physiotherapists treating people with JHS are more likely to have specialist musculoskeletal skills. Although questionnaires were distributed predominantly to secondary care organisations, some 44% of respondents reported that they worked in primary care. We are therefore confident that we have gained insight into a range of musculoskeletal physiotherapy services.

Almost 80% of participants saw 10 or less JHS patients each year (Table 1) suggesting that JHS is not widely encountered by respondents to this survey. This is perhaps surprising given the high reported prevalence of symptomatic hypermobility (Simpson 2006). It is clear that high quality epidemiological evidence related to the prevalence of JHS in the general population is lacking, although the prevalence in Omani women attending musculoskeletal outpatient physiotherapy services was established to be 55% (Clark and Simmonds 2011). It is therefore likely that the condition continues to not be adequately identified in clinical practise (Hakim and Grahame 2004), supported by the observation that almost 68% of respondents reported that they had not received any formal education or training related to JHS. Referrals were received mainly from GPs or Rheumatology consultants, although the specific referral details are unknown. Patient self-referral was reported by approximately 21% of respondents which contrasts with 46% of musculoskeletal services available for self-referral across the UK (CSP 2011).

After referral, approximately 75% of physiotherapists see patients within 2 months and 95% within 4 months. This compares favourably with the 71% of patients seen in 8 weeks or less in musculoskeletal physiotherapy services across the UK (CSP 2011). 86% of physiotherapists in the present survey offer an initial assessment of at least 40 minutes, with follow-up appointments lasting 30 minutes or less in 95% of cases. 79% offer 6 sessions or less, compared to a UK average number of musculoskeletal physiotherapy sessions of 3.31 and a maximum average of 6 (CSP 2011). 72% complete their treatment within 4 months. 39% of respondents 'rarely' or 'never' offer a maintenance review, although a sizeable 30% 'always' or 'frequently' offer a review. It is not possible to know how appropriate or effective these models

of service delivery are or how much control individual therapists have in tailoring services to individual patient needs. It is likely that many service constraints are dictated by local managers in response to service demands and available resources. Nonetheless it is useful to understand the organisational context within which people with JHS are being managed.

The Brighton criteria for the diagnosis of JHS were reported as being used by only 31% of respondents. It is therefore not possible to be confident about whether patients labelled as having JHS actually meet the recommended diagnostic criteria. Some may be misdiagnosed and have other specific connective tissue diseases such as Marfan Syndrome. 87% of respondents reported using the Beighton criteria, which of course remain a key component of the Brighton criteria. Using the Beighton criteria in isolation however, means that other criteria related to arthralgia, dislocation/subluxation, soft tissue lesions, marfanoid habitus and other soft tissue signs may be ignored. Those who had received training in the management of JHS were more likely to use the Brighton criteria therefore additional awareness-raising and training are required to ensure that consistent diagnostic criteria are being used in clinical practice.

The primary aims of treatment included education, enhancing self-management and encouraging long-term exercise, each being reported by over 90% of physiotherapists as 'always' being an aim. However decreasing pain was not very high up the list of treatment aims, with only 42% 'always' considering this a key aim. This is perhaps surprising, particularly when expert opinion suggests that addressing pain is a key aim of treatment (Keer and Simmonds 2011). However, this may simply be a reflection of the structure and design of this survey, which listed a wide range of specific aims and interventions, some of which might act to reduce pain directly (such as Transcutaneous Electrical Nerve Stimulation - TENS) and others which might do so indirectly (such as exercise). It may be that respondents prioritised some of the other stated aims, with pain reduction being an implicit by-product. Alternatively, there may be a true predominance of attempts to address the impact of pain rather than reduce pain in its own right. In the future, it would be useful to explore treatment aims in more detail using alternative methodologies such as in-depth interviews. It would also be helpful to identify how the aims of

physiotherapy management match the beliefs and expectations of patients and other health professionals.

It was noticeable that the specific interventions used seemed to match very well against the stated aims of physiotherapy. Advice, education, self-management techniques, exercise/physical activity, posture re-education, pacing and goal setting were amongst the most popular interventions. Such interventions are common approaches in effective pain management and are commonly recommended in evidence based clinical guidelines for chronic pain (SIGN 2013) and other musculoskeletal conditions such as osteoarthritis (NICE 2014) and low back pain (NICE 2009). Experts in the area have recommended that education and advice for adults with JHS should include information about sustaining harmful postures and repetitive activities (Keer et al. 2003). Advice on a range of other lifestyle modifications is also advocated, including sleeping, carrying, clothing, footwear, travelling, family and children, other health issues, management of related symptoms, medication and sport and exercise (Keer et al. 2003). Passive interventions such as electrotherapy modalities, injection and acupuncture were used less often, reflecting the emphasis on self-management.

In terms of outcome measures, a pain scale was most often used yet, as noted earlier, pain reduction was not considered a main aim of physiotherapy. There therefore seems to be a mismatch between the reported main aims of treatment and the outcome measures chosen to capture changes in patients' conditions. It is likely that this is because pain scales such as the visual analogue scale are quick to administer and score (Waterfield and Sim 1995). The Chartered Society of Physiotherapy (CSP 2010) has recommended that the EQ-5D, a generic measure of health, should be used in all musculoskeletal services, along with a condition-specific outcome measure as appropriate. Only 34% reported that they 'always' used the EQ-5D, with 38% reporting that they 'never' used it with patients with JHS. Outcome assessment in JHS therefore requires additional consideration. Development of a condition-specific outcome measure would be welcome in this area.

Limitations of the present survey include the relatively small sample size. In saying that, it captured a wide range of individual experience and both primary and secondary care NHS services. Only a small number of private practitioners were included (n=2) so care must be taken in extrapolating the findings to such settings. It should also be acknowledged that the results represent perceptions of what occurs in practice, which may differ from actual practice. Implementation of strategies to enhance the response rate would have been useful and should be pursued in future research.

## **CONCLUSION**

This survey successfully captured the views of physiotherapists with a very wide range of experience, working in a range of practice settings across the UK. The results suggest that the aims of physiotherapy and management approaches seem appropriate to JHS as a long term condition. However the recommended Brighton diagnostic criteria are not often used and assessment tools do not necessarily match the treatment aims and approaches used. Additional training specific to JHS may therefore be required, particularly in diagnosis and assessment.

## **KEY WORDS**

Joint Hypermobility; Physiotherapy; Survey

## REFERENCES

- Artz N, Dixon S, Wylde V, Beswick A, Blom A, Gooberman-Hill R (2013). Physiotherapy provision following discharge after total hip and total knee replacement: a survey of current practice at high-volume NHS hospitals in England and Wales. *Musculoskeletal Care* 11: 31-38. DOI:10.1002/msc.1027.
- Beighton P, Solomon L, Soskolne CL (1973). Articular mobility in an African population. *Annals of the Rheumatic Diseases* 32(5): 413-418. DOI:10.1136/ard.32.5.413.
- Clark CJ, Simmonds JV (2011). An exploration of the prevalence of hypermobility and joint hypermobility syndrome in Omani women attending a hospital physiotherapy service. *Musculoskeletal Care* 9(1): 1-10. DOI: 10.1002/msc.184.
- Chartered Society of Physiotherapy (2010). EQ-5D-5L - Measuring change in Musculoskeletal Physiotherapy Outpatient Services <http://www.csp.org.uk/documents/eq-5d-5l-measuring-change-musculoskeletal-physiotherapy-outpatient-services> [3 January 2014].
- Chartered Society of Physiotherapy (2011). A survey of NHS Physiotherapy waiting times, workforce and caseloads in the UK 2010-2011. London: Chartered Society of Physiotherapy.
- Engelbert RH, van Bergen M, Henneken T, Helders PJ, Takken T (2006). Exercise tolerance in children and adolescents with musculoskeletal pain in joint hypermobility and joint hypomobility syndrome. *Pediatrics* 118(3): e690-696. DOI: 10.1542/peds.2005-2219.
- The EuroQol Group (1990). EuroQol-a new facility for the measurement of health-related quality of life. *Health Policy* 16(3):199-208. DOI: 10.1016/0168-8510(90)90421-9.
- Fatoye F, Palmer S, Macmillan F, Rowe P, van der Linden M (2009). Proprioception and muscle torque deficits in children with hypermobility syndrome. *Rheumatology* 48(2): 152-157. DOI: 10.1093/rheumatology/ken435.
- Fatoye F, Palmer S, Macmillan F, Rowe P, Van der Linden M (2011). Gait kinematics and passive knee joint range of motion in children with hypermobility syndrome. *Gait and Posture* 33: 447-451. DOI: 10.1016/j.gaitpost.2010.12.022.
- Fatoye F, Palmer S, Macmillan F, Rowe P, Van der Linden M (2012). Pain intensity and quality of life perception in children with hypermobility syndrome. *Rheumatology International* 32(5): 1277-1284. DOI: 10.1007/s00296-010-1729-2.

Ferrell WR, Tennant N, Sturrock RD, Ashton L, Creed G, Brydson G, Rafferty D (2004). Amelioration of symptoms by enhancement of proprioception in patients with joint hypermobility syndrome. *Arthritis and Rheumatism* 50(10): 3323-3328. DOI: 10.1002/art.20582.

French HP (2007). Physiotherapy management of osteoarthritis of the hip: a survey of current practice in acute hospitals and private practice in the Republic of Ireland. *Physiotherapy* 93: 253-260. DOI:10.1016/j.physio.2006.12.008.

Grahame R (2000). Hypermobility-not a circus act. *International Journal of Clinical Practice* 54(5): 314-315.

Grahame R, Bird HA, Child A (2000). The revised (Brighton 1998) criteria for the diagnosis of benign joint hypermobility syndrome (BJHS). *The Journal of Rheumatology* 27: 1777-1779.

Hakim AJ, Grahame R (2004). Non-musculoskeletal symptoms in joint hypermobility syndrome. Indirect evidence for autonomic dysfunction? *Rheumatology* 43(9):1194-1195. DOI: 10.1093/rheumatology/keh279.

Hall MG, Ferrell WR, Sturrock RD, Hamblen DL, Baxendale RH (1995). The effect of the hypermobility syndrome on knee joint proprioception. *British Journal of Rheumatology* 34: 121-125.

Hanchard NCA, Goodchild L, Thompson J, O'Brien T, Davison D, Richardson C (2011). A questionnaire survey of UK physiotherapists on the diagnosis and management of contracted (frozen) shoulder. *Physiotherapy* 97: 115-125. DOI: 10.1016/j.physio.2010.08.012

Keer R, Grahame R (2003). *Hypermobility syndrome. Recognition and management for physiotherapists*. Edinburgh: Butterworth Heinemann.

Keer R, Edwards-Fowler A, Mansi E (2003). Management of the hypermobile adult. In Keer R, Grahame R (eds) *Hypermobility syndrome. Recognition and management for physiotherapists*. Edinburgh: Butterworth Heinemann.

Keer R, Simmonds J (2011). Joint protection and physical rehabilitation of the adult with hypermobility. *Current Opinion in Rheumatology* 23: 131-136. DOI: 10.1097/BOR.0b013e328342d3af.

Mallik AK, Ferrell WR, McDonald A (1994). Impaired proprioceptive acuity at the proximal interphalangeal joint in patients with the hypermobility syndrome. *British Journal of Rheumatology* 33: 631-637.

National Institute for Health and Clinical Excellence (2014). Osteoarthritis. Care and management in adults. Clinical guideline CG177. <http://www.nice.org.uk/guidance/CG177> [18 September 2014].

National Institute for Health and Clinical Excellence (2009). Low back pain: early management of persistent non-specific low back pain. Clinical guideline 88. <http://www.nice.org.uk/guidance/CG88> [18 September 2014].

Palmer S, Bailey S, Barker L, Barney L, Elliott A (2014). The effectiveness of therapeutic exercise for joint hypermobility syndrome: a systematic review. *Physiotherapy*, 100: 220-227. DOI: 10.1016/j.physio.2013.09.002.

Sahin N, Baskent A, Ugurlu H, Berker E (2008a). Isokinetic evaluation of knee extensor/flexor muscle strength in patients with hypermobility syndrome. *Rheumatology International* 28: 643-648. DOI: 10.1007/s00296-007-0493-4.

Sahin N, Baskent A, Cakmak A, Salli A, Ugurlu H, Berker E (2008b). Evaluation of knee proprioception and effects of proprioception exercise in patients with benign joint hypermobility syndrome. *Rheumatology International* 28: 995-1000. DOI: 10.1007/s00296-008-0566-z.

Scottish Intercollegiate Guidelines Network (2013). Management of chronic pain (SIGN publication no. 136). Edinburgh: SIGN <http://www.sign.ac.uk> [18 September 2014].

Simmonds JV, Keer RJ (2007). Hypermobility and hypermobility syndrome. *Manual Therapy* 12(2): 298-309. DOI:10.1016/j.math.2007.05.001.

Simmonds JV, Keer RJ (2008). Hypermobility and the hypermobility syndrome. Part 2: Assessment and management of hypermobility syndrome: illustrated via case studies. *Manual Therapy* 13(2): e1–11. DOI: 10.1016/j.math.2007.11.001.

Simpson MR (2006). Benign joint hypermobility syndrome: evaluation, diagnosis, and management. *Journal of the American Osteopathic Association*, 106(9): 531-6.

Tinkle BT (2008). Issues and management of joint hypermobility. A guide for the Ehlers-Danlos Syndrome Hypermobility Type and the Hypermobility Syndrome. USA: Left Paw Press.

Walsh NE, Hurley MV (2009). Evidence based guidelines and current practice for physiotherapy management of knee osteoarthritis. *Musculoskeletal Care* 7(1): 45-56. DOI: 10.1002/msc.144.

Ware JE (2000). SF-36 Health Survey Update. *Spine* 25(24): 3130-3139. DOI: 10.1097/00007632-200012150-00008.

Waterfield J, Sim J (1995). Clinical assessment of pain by the visual analogue scale. *British Journal of Therapy and Rehabilitation* 3(2): 94-97.



**Table 1. Participant characteristics.** \*Multiple answers could be selected for these questions therefore total responses may exceed 100%.

<b>Characteristic (number of valid responses)</b>	<b>Response Options</b>	<b>% of valid responses</b>
“Sex” (n=66)	Male	19.7
	Female	80.3
“Working status” (n=66)	Full-time	53.0
	Part-time	47.0
“UK employment grade” (n=66)	Band 5	4.5
	Band 6	18.2
	Band 7	54.5
	Band 8a	15.2
	Band 8b	6.1
	N/A	0.0
“Number of years qualified” (n=66)	0-2	1.5
	3-5	10.6
	6-10	16.7
	11-15	24.2
	16-20	13.6
	>21	33.3
“Estimate the number of individuals with JHS that you have treated in the last year” (n=53)	None	1.9
	1-5	56.6
	6-10	20.8
	11-15	7.5
	16-20	7.5
	>21	28.3
“Have you received formal training in JHS management?” (n=65)	No	67.7
	Yes	32.3
“In which settings do you manage individuals with JHS?” (n=59)*	Primary care	44.1
	Secondary care	62.7
	Private practice	3.4

**Table 2. Information about clinical services and criteria used to diagnose JHS.** \*Multiple answers could be selected for these questions therefore total responses may exceed 100%.

Question (number of valid responses)	Response Options	% of valid responses
"How long (on average) do individuals with JHS wait to see a physiotherapist in your service?" (n=64)	<1 week 1-2 weeks 3-4 weeks 1-2 months 3-4 months 5-6 months >6 months Don't know	1.6 7.8 23.4 42.2 20.3 0.0 1.6 3.1
"Where do you receive JHS referrals from?" (n=61)*	GP Orthopaedic consultant Patients (self-referral) Rheumatology consultant Other	77.0 45.9 21.3 77.0 13.1
"On what basis do you offer physiotherapy to individuals with JHS?" (n=61)*	Individual Group	100.0 16.4
"What is the duration (on average) of the first assessment?" (n=66)	10 minutes 20 minutes 30 minutes 40 minutes 50 minutes 60 minutes >60 minutes Don't know	0.0 0.0 13.6 40.9 12.1 31.8 1.5 0.0
"What is the duration (on average) of each treatment session?" (n=66)	10 minutes 20 minutes 30 minutes 40 minutes 50 minutes 60 minutes >60 minutes Don't know	3.0 16.7 75.8 4.5 0.0 0.0 0.0 0.0
"How many sessions (on average) do you offer (including the first assessment)?" (n=66)	1 2 3 4 5 6 >6 Don't know	0.0 1.5 9.1 21.2 4.5 42.4 12.1 9.1
"Over what duration (on average) do you treat each individual with JHS?" (n=65)	<1 week 1-2 weeks 3-4 weeks 1-2 months 3-4 months 5-6 months >6 months Don't know	0.0 1.5 10.8 23.1 36.9 15.4 9.2 3.1

“Do you work as part of a multidisciplinary team?” (n=65)	No	36.9
	Yes	63.1
“What other health services can you refer individuals with JHS to?” (n=56)*	Clinical psychologist	17.9
	Occupational therapist	78.6
	Pain clinic	51.8
	Pain management programme	53.6
	Podiatrist	75.0
	Other	8.9
“Do you provide a follow-up maintenance review for individuals with JHS?” (n=64)	Always	10.9
	Frequently	18.8
	Sometimes	31.3
	Rarely	20.3
	Never	18.8
	N/A	0.0
“If you use exercise, do you encourage individuals with JHS to exercise through...” (n=58)	Their full range of motion	43.1
	A restricted range of motion	41.4
	I don't specify	15.5
“What formal diagnostic tests/criteria do you use to confirm a diagnosis of JHS?” (n=61)*	Beighton	86.9
	Brighton	31.1
	Bulbena	0.0
	Carter & Wilkinson	0.0
	Contompasis	0.0
	Other	0.0
	No formal test/criteria	13.1
“What additional criteria do you use to confirm a diagnosis of JHS?” (n=53)*	Pain	64.2
	Family history of JHS	81.1
	Other	5.7

**Table 3. Aims of physiotherapy.** Figures are presented as a % of valid responses. Responses are presented in rank order based on frequency. N/A = Not Applicable.

“What do you consider to be the aims of physiotherapy for JHS?” (number of valid responses)	Always	Frequently	Sometimes	Rarely	Never	N/A
Education (n=64)	96.9	3.1	0.0	0.0	0.0	0.0
Enhance self-management (n=65)	93.8	4.6	1.5	0.0	0.0	0.0
Encourage long-term exercise (n=65)	90.8	9.2	0.0	0.0	0.0	0.0
Improve muscle control (n=65)	76.9	21.5	1.5	0.0	0.0	0.0
Improve posture/ergonomics (n=65)	73.8	23.1	3.1	0.0	0.0	0.0
Teach joint protection (n=63)	71.4	25.4	1.6	0.0	0.0	1.6
Improve function (n=65)	67.7	23.1	9.2	0.0	0.0	0.0
Increase strength (n=63)	55.6	39.7	4.8	0.0	0.0	0.0
Reduce fear avoidance (n=65)	52.3	35.4	10.8	1.5	0.0	0.0
Improve balance (n=64)	50.0	28.1	21.9	0.0	0.0	0.0
Improve endurance (n=65)	47.7	33.8	18.5	0.0	0.0	0.0
Decrease pain (n=65)	41.5	36.9	20.0	1.5	0.0	0.0
Improve mobility (n=63)	33.3	39.7	15.9	7.9	3.2	0.0
Prevent surgery (n=62)	24.2	19.4	30.6	17.7	4.8	3.2
Increase range of movement (n=63)	7.9	25.4	31.7	20.6	12.7	1.6
Swelling (n=62)	4.8	19.4	48.4	27.4	0.0	0.0
Other (n=0)	0.0	0.0	0.0	0.0	0.0	0.0

**Table 4. Specific interventions used for JHS.** Figures are presented as a % of valid responses. Responses are presented in rank order based on frequency. N/A = Not Applicable.

“What interventions do you use for JHS?” (number of valid responses)	Always	Frequently	Sometimes	Rarely	Never	N/A
Advice (n=65)	96.9	3.1	0.0	0.0	0.0	0.0
Education (n=65)	96.9	3.1	0.0	0.0	0.0	0.0
Self-management (n=64)	89.1	4.7	3.1	1.6	1.6	0.0
Exercise (muscle control) (n=65)	67.7	30.8	1.5	0.0	0.0	0.0
Posture re-education (n=64)	62.5	31.3	6.3	0.0	0.0	0.0
Exercise (proprioception) (n=65)	61.5	27.7	9.2	1.5	0.0	0.0
Goal setting (n=64)	60.9	14.1	21.9	3.1	0.0	0.0
Physical activity (n=64)	59.4	31.3	7.8	1.6	0.0	0.0
Pacing (n=65)	52.3	36.9	10.8	0.0	0.0	0.0
Exercise (functional) (n=63)	50.8	39.7	9.5	0.0	0.0	0.0
Exercise (strengthening) (n=64)	50.0	31.3	15.6	3.1	0.0	0.0
Movement re-education (n=64)	34.4	46.9	18.8	0.0	0.0	0.0
Exercise (cardiovascular) (n=64)	34.4	32.8	23.4	7.8	1.6	0.0
Ergonomics (n=62)	33.9	43.5	14.5	4.8	3.2	0.0
Exercise (range of movement) (n=63)	33.3	23.8	22.2	14.3	4.8	1.6
Gait re-education (n=64)	25.0	32.8	37.5	1.6	3.1	0.0
Health behaviour change techniques (n=64)	25.0	29.7	28.1	12.5	4.7	0.0
Pilates (n=64)	23.4	46.9	14.1	3.1	10.9	1.6
Hands-on soft tissue techniques (n=63)	19.0	12.7	39.7	23.8	3.2	1.6
Stretching (n=63)	15.9	19.0	47.6	9.5	6.3	1.6
Electrotherapy (n=44)	6.8	2.3	11.4	47.7	25.0	6.8
Ultrasound (n=52)	9.6	5.8	28.8	30.8	21.2	3.8
Combination therapy (n=48)	6.3	2.1	8.3	16.7	56.3	10.4
Laser (n=48)	6.3	0.0	8.3	12.5	64.6	8.3
TENS (n=54)	5.6	3.7	40.7	22.2	25.9	1.9
Interferential (n=49)	4.1	2.0	6.1	12.2	67.3	8.2
Electroacupuncture (n=50)	4.0	0.0	10.0	14.0	58.0	14.0
Shortwave diathermy (n=51)	3.9	2.0	9.8	11.8	62.7	9.8
Manual joint mobilisations (n=64)	6.3	10.9	26.6	34.4	20.3	1.6
Taping (n=63)	6.3	7.9	55.6	23.8	6.3	0.0
Walking aids (n=61)	3.3	1.6	54.1	32.8	6.6	1.6
Tubigrip (n=61)	3.3	1.6	26.2	36.1	31.1	1.6

Hydrotherapy (n=63)	3.2	23.8	33.3	12.7	19.0	7.9
Heat (n=64)	3.1	20.3	37.5	26.6	9.4	3.1
Ice (n=64)	1.6	6.3	46.9	25.0	18.8	1.6
Injection (n=64)	1.6	0.0	12.5	35.9	35.9	14.1
Other (n=4)	0.0	50.0	0.0	25.0	0.0	25.0
Acupuncture (n=62)	0.0	9.7	43.5	29.0	14.5	3.2

**Table 5. Outcome assessment used for JHS.** Figures are presented as a % of valid responses. Responses are presented in rank order based on frequency. N/A = Not Applicable.

"What outcome measures do you use for individuals with JHS?" (number of valid responses)	Always	Frequently	Sometimes	Rarely	Never	N/A
Pain scale (n=59)	57.6	25.4	6.8	3.4	6.8	0.0
Patient agreed (n=51)	52.9	13.7	9.8	2.0	19.6	2.0
Strength (n=47)	46.8	21.3	8.5	6.4	14.9	2.1
Functional test (n=50)	38.0	22.0	4.0	4.0	30.0	2.0
Range of movement (n=45)	37.8	17.8	22.2	0.0	22.2	0.0
EQ5D (n=53)	34.0	11.3	11.3	3.8	37.7	1.9
Other (n=20)	20.0	10.0	10.0	5.0	45.0	10.0
Proprioception (n=44)	18.2	31.8	13.6	2.3	29.5	4.5
MYMOP (n=47)	14.9	19.1	21.3	4.3	38.3	2.1
SF12/SF36 (n=43)	11.6	7.0	9.3	4.7	62.8	4.7
WOMAC (n=40)	2.5	5.0	5.0	2.5	82.5	2.5
AIMS2 (n=40)	2.5	2.5	5.0	2.5	82.5	5.0
Health Assessment Questionnaire (HAQ) (n=42)	2.4	2.4	11.9	9.5	71.4	2.4
Timed Up and Go (n=42)	2.4	2.4	9.5	11.9	71.4	2.4
Hospital Anxiety & Depression Scale (HADS) (n=47)	2.1	6.4	19.1	17.0	53.2	2.1
Illness Behaviour Questionnaire (n=39)	0.0	2.6	2.6	7.7	84.6	2.6