ELSEVIER

Physiotherapy

Physiotherapy 103 (2017) 186–192

Development and initial validation of the Bristol Impact of Hypermobility questionnaire

S. Palmer [a,*], F. Cramp [a], R. Lewis [b], G. Gould [c], E.M. Clark [c]

[a] *Department of Allied Health Professions, University of the West of England, Bristol, UK*
[b] *Department of Physiotherapy, North Bristol NHS Trust, Bristol, UK*
[c] *Musculoskeletal Research Unit, University of Bristol, Bristol, UK*

Abstract

Objectives Stage 1 – to identify the impact of joint hypermobility syndrome (JHS) on adults; Stage 2 – to develop a questionnaire to assess the impact of JHS; and Stage 3 – to undertake item reduction and establish the questionnaire's concurrent validity.
Design A mixed methods study employing qualitative focus groups and interviews (Stage 1); a working group of patients, clinicians and researchers, and 'think aloud' interviews (Stage 2); and quantitative analysis of questionnaire responses (Stage 3).
Setting Stages 1 and 2 took place in one secondary care hospital in the UK. Members of a UK-wide patient organisation were recruited in Stage 3.
Participants In total, 15, four and 615 participants took part in Stages 1, 2 and 3, respectively. Inclusion criteria were: age ≥18 years; diagnosis of JHS; no other conditions affecting physical function; able to give informed consent; and able to understand and communicate in English.
Interventions None.
Main outcome measures The development of a questionnaire to assess the impact of JHS.
Results Stage 1 identified a wide range of impairments, activity limitations and participation restrictions In Stage 2, a draft questionnaire was developed and refined following 'think aloud' analysis, leaving 94 scored items. In Stage 3, items were removed on the basis of low severity and/or high correlation with other items. The final Bristol Impact of Hypermobility (BIoH) questionnaire had 55 scored items, and correlated well with the physical component score of the Short Form 36 health questionnaire ($r = -0.725$).
Conclusions The BIoH questionnaire demonstrated good concurrent validity. Further psychometric properties need to be established.

Keywords: Hypermobility; Joint; Joint laxity; Familial; Questionnaires; Interview; Focus groups; Validity of results

Introduction

Joint hypermobility syndrome (JHS) is a heritable connective tissue disorder characterised by excessive joint range of motion and pain [1]. It has been reported to affect up to 5% of women and 0.6% of men [2], although there is a lack of good-quality epidemiological evidence for the true prevalence of JHS in the general population. The prevalence in musculoskeletal practice contexts is likely to be very high, however, with 30% of those referred to a musculoskeletal triage clinic in the UK meeting the Brighton diagnostic criteria [3,4].

JHS is associated with a wide range of problems including pain; fatigue; proprioception deficits; and repeated cycles of injury, anxiety and catastrophising [5]. It may also be associated with a range of autonomic and gastrointestinal symptoms, and functional difficulties indicative of developmental coordination disorder/dyspraxia [6]. Empirical data have shown that, compared with healthy controls, JHS has a significant impact on outcomes such as exercise endurance, gait, pain, proprioception, strength, function and quality of

* Corresponding author. Address: Department of Allied Health Professions, University of the West of England, Blackberry Hill, Bristol BS16 1DD, UK. Tel.: +44 0117 3288919.
E-mail address: Shea.Palmer@uwe.ac.uk (S. Palmer).

http://dx.doi.org/10.1016/j.physio.2016.04.002
0031-9406/

life in both children [7–10] and adults [11–14]. A recent systematic review and meta-analysis confirmed the impact of JHS on a range of psychological variables such as fear, agoraphobia, anxiety, depression and panic disorders [15].

Physiotherapy, particularly exercise, is a mainstay of treatment for JHS, although recent systematic reviews highlighted the lack of research evidence [16,17]. The trials in adults included in those reviews used a range of patient-reported outcome measures (PROMs), including the Short Form 36 (SF-36) [18], the Arthritis Impact Measurement Scales 2 (AIMS-2) [13] and a questionnaire developed by Barton and Bird [19]. Of those, only the SF-36 captured improvements following exercise [18]. Only one of the five AIMS-2 subscales changed with exercise [13], and no changes were evident in Barton and Bird's questionnaire [19]. Therefore, if exercise is effective (which has yet to be demonstrated convincingly [16]), only the SF-36 seemed to demonstrate sufficient measurement sensitivity. Closer inspection of these PROMs identified a lack of face, content and construct validity [20] for many issues reported by people with JHS [5]. For example, Barton and Bird's questionnaire [19] focused on lower limb activity (such as ascending and descending stairs, squatting, standing up and walking), failing to reflect upper limb functional difficulties. Neither the process of development nor the psychometric properties of the questionnaire were reported. A recent survey of physiotherapy practice in the UK [21] highlighted a lack of congruence between the aims of physiotherapy management for JHS and the tools used to assess the effectiveness of management. There is, therefore, a need to develop a condition-specific, psychometrically sound, outcome measure to underpin future research and clinical practice in this area.

This project had a number of related aims.

- Stage 1 – to identify the impact of JHS on adults with the condition to inform initial patient-specific questionnaire items.
- Stage 2 – to develop a questionnaire to assess the personal impact of JHS.
- Stage 3 – to reduce the number of questionnaire items and establish the concurrent validity of the new questionnaire against the SF-36.

Method

Ethical approval was obtained from the South West 5 NHS Research Ethics Committee. The research was conducted in three stages.

- Stage 1 – identification of questionnaire items. Methods: focus groups and telephone interviews with people with JHS.
- Stage 2 – development of the initial questionnaire. Methods: working group of patient research partners and researchers; 'think aloud' evaluation.
- Stage 3 – item reduction and validation of the questionnaire. Methods: administration of the initial questionnaire and SF-36 to members of the Hypermobility Syndromes Association (HMSA), a UK-based patient organisation; item removal; assessment of the concurrent validity of the final questionnaire items against the SF-36; production of the final questionnaire.

Participants

Inclusion criteria for Stages 1 to 3 were: diagnosed with JHS; age ≥18 years; no other formally diagnosed conditions affecting physical function (such as inflammatory arthritis, osteoarthritis or neurological conditions); able to give informed consent; and able to understand and communicate in English. All five members of the research team were recruited in Stage 2.

The sources of recruitment at each stage were as follows.

- Stages 1 and 2 – patients who met the Brighton criteria [3] for JHS (confirmed by a physiotherapist) who had been seen by the physiotherapy service at North Bristol NHS Trust in the previous 2 years were sent an invitation letter, participant information sheet and a reply slip. All participants completed informed signed consent forms. Two patient research partners (people with JHS who advised on the design and conduct of all aspects of the research, including the wording of patient information sheets and consent forms, and sat as equal members of a study steering group), and one further person with JHS who contributed to the working group during Stage 2 were recruited from the same cohort.
- Stage 3 – adult members of HMSA were sent an invitation letter, participant information sheet and a copy of the questionnaires. Diagnosis of JHS was self-declared. Completion and return of the questionnaires was taken as implied consent.

Procedure

Stage 1

Two focus groups of people with JHS were conducted to explore the impact of the condition. An option to undertake a telephone interview was provided for those who were unable or unwilling to attend a focus group. A loose topic guide was used to steer the focus group and interview discussions. The same researcher (GG) conducted all focus groups and interviews, with another researcher (SP) taking notes during the focus groups to aid transcription. Focus groups and interviews were audio-recorded, transcribed verbatim and anonymised. Open coding of the transcripts was used to identify individual questionnaire items, and codes were discussed in detail and verified by two researchers (GG and SP). Thematic analysis of the data did not progress beyond this first level of coding as the aim was limited to identification of individual items.