



Original article

A qualitative evaluation of the appropriateness, validity, acceptability, feasibility and interpretability of the Bristol Impact of Hypermobility (BioH) questionnaire



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ABSTRACT

Background: The Bristol Impact of Hypermobility (BioH) questionnaire is a condition-specific patient-reported outcome measure developed for adults with Joint Hypermobility Syndrome (JHS). It has previously demonstrated strong concurrent validity with the Short-Form 36 health questionnaire and excellent test-retest reliability.

Objectives: This study aimed to evaluate its appropriateness, validity, acceptability, feasibility and interpretability.

Design: A qualitative evaluation using semi-structured telephone interviews, incorporating a 'think aloud' exercise and additional prompts.

Method: Adults with JHS (n = 11) were recruited through a patient organisation and physiotherapists with a professional interest in JHS (n = 9) were recruited through the same organisation and an online professional network. Interviews were transcribed and data synthesised using a framework matrix.

Findings: Patients and physiotherapists commented positively on the appropriateness, validity, acceptability and feasibility of the BioH questionnaire. Physiotherapists assessed the interpretability of the questionnaire and commented that, whilst further information might be captured, the value of that information might be limited. The questionnaire was considered comprehensive with only a very limited number of potential missing areas related to use of hand-held technology, hair washing/drying and intimacy. Interestingly, keyboard use and hair washing were excluded during initial questionnaire development as they were rated as relatively unimportant. Intimacy was not considered a comfortable addition for all participants.

Conclusions: Patients and physiotherapists regarded the BioH questionnaire as a welcome addition to the toolkit available to assess those with JHS. It was broadly accepted as reflecting the experience of people with JHS in sufficient detail to support management.

1. Introduction

Joint Hypermobility Syndrome (JHS) is a heritable connective tissue disorder characterised by excessive range of movement in multiple joints and pain (Grahame, 2003). There is a lack of epidemiological data, although the prevalence in musculoskeletal services is high, with 30% of referrals to one musculoskeletal triage service in the UK (Connolly et al., 2015) meeting the diagnostic criteria (Grahame et al., 2000). JHS is associated with pain, fatigue, proprioception deficits and repeated cycles of injury (Terry et al., 2015). There is also evidence of

anxiety and catastrophising (Terry et al., 2015), fear, agoraphobia, depression and panic disorders (Smith et al., 2014b). It should be noted that the diagnostic criteria and associated nosology have recently been revised but the current research will use the term JHS as it pre-dated the new terms of 'hypermobile Ehlers-Danlos Syndrome (hEDS)' (Malfait et al., 2017) and 'Hypermobility Spectrum Disorder (HSD)' (Castori et al., 2017).

Physiotherapy is the main treatment for JHS, particularly exercise to enhance physical function. Two systematic reviews have, however, highlighted a lack of evidence to support therapy (Palmer et al., 2014;

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Smith et al., 2014a). A lack of congruence between the aims of physiotherapy and the outcome measures used to evaluate effectiveness has also been identified (Palmer et al., 2015). A condition-specific outcome measure has therefore been developed with patients to more accurately reflect the wide-ranging impact of JHS (Palmer et al., 2017a). The 'Bristol Impact of Hypermobility (BioH)' questionnaire gives a maximum score of 360, with higher scores representing more severe impact. It addresses items such as pain, fatigue, physical function, anxiety, planning and management, and strength and weakness (Palmer et al., 2017a).

The BioH questionnaire has already undergone some evaluation of its psychometric properties. It correlates strongly with the physical component score of the Short Form 36 (SF-36) questionnaire ($r = -0.725$, $n = 615$) (Palmer et al., 2017a). High correlation coefficients were also observed for the majority of the physical component score subscales (physical function, role physical and bodily pain all $r > -0.7$, $p < 0.001$). The only SF-36 mental component score subscale that had a strong correlation with the BioH questionnaire values was social functioning ($r = -0.717$, $p < 0.001$). The overall mental component score and all other SF-36 subscales demonstrated statistically significant correlations (all $p < 0.001$) but with more moderate coefficient values ($r = -0.447$ to -0.624) (Palmer et al., 2017a). The BioH questionnaire has also demonstrated excellent test-retest reliability (ICC = 0.923, $n = 233$), performing better than the SF-36 in this regard (ICC = 0.887 and 0.778 for the physical and mental component scores respectively) (Palmer et al., 2017b). The smallest detectable change in the BioH score is 42 points, equivalent to a 19% change from baseline (as compared to a 25% and 37% change for the SF-36 physical and mental component scores respectively) (Palmer et al., 2017b). Finally the BioH has been shown to be more closely related to patients' global ratings of change ($r = -0.493$, $p < 0.001$, $n = 363$) than the SF-36 ($r = 0.186$ and 0.203 for the physical and mental component scores respectively, both $p < 0.001$ and $n = 363$) (Palmer et al., 2017b).

Important aspects related to validity, reliability and sensitivity to change have thus been established for the BioH questionnaire and it seems to perform very well in these aspects. However, other psychometric properties must be established before the BioH questionnaire can be confidently used to support JHS research and clinical practice.

This investigation therefore aimed to qualitatively evaluate specific properties of the BioH questionnaire, namely appropriateness, validity, acceptability, feasibility and interpretability. These concepts are defined in Table 1 and represent five of the eight criteria for evaluating patient-reported outcome measures for use in clinical trials (Fitzpatrick et al., 1998). Other criteria related to reliability, responsiveness and precision will continue to be addressed in future work.

2. Methods

The study received a favourable opinion from the Faculty of Health & Applied Sciences Ethics Sub-Committee at the University of the West of England, Bristol (HAS/15/01/99).

Table 1

Definitions of the criteria for evaluating patient-reported outcome measures explored in this research (Fitzpatrick et al., 1998).

Criteria	Definition
'Appropriateness'	Is the content of the instrument appropriate to the questions which the clinical trial is intended to address?
'Validity'	Does the instrument measure what it claims to measure?
'Acceptability'	Is the instrument acceptable to patients?
'Feasibility'	Is the instrument easy to administer and process?
'Interpretability'	How interpretable are the scores of an instrument?

2.1. Study design

Semi-structured telephone interviews using a 'think aloud' technique (also known as 'cognitive interviewing' (Drennan, 2003)) captured patients' and physiotherapists' views on the questionnaire.

2.2. Recruitment

2.2.1. Adults with JHS

Patients were purposively selected from those who took part in an associated test-retest reliability study (Palmer et al., 2017b). Members of the Hypermobility Syndromes Association (HMSA), a United Kingdom (UK) patient organisation, were approached. Inclusion criteria were:

- ≥ 16 years old.
- Fulfil two or more JHS screening questions (Hakim and Grahame, 2003).
- No other formally diagnosed conditions affecting physical function.
- Able to give informed consent.
- Able to understand and communicate in English.

Recruitment packs were distributed to all HMSA members happy to be contacted about research ($n = 1080$). Packs included a participant information sheet, consent form, BioH and SF36 questionnaires (used for the associated test-retest reliability study (Palmer et al., 2017b)). Potential participants were separately asked to consider taking part in qualitative interviews, the results of which are the focus of this manuscript. The prospective target sample was $n = 10$ which was considered sufficient to identify an appropriate range of opinions (Sandelowski, 1995). From the positive responses, patients were purposively selected on the basis of gender, age and BioH score.

2.3. Physiotherapists

Physiotherapists with an interest in JHS were also approached through the HMSA, who distributed recruitment packs (containing an information sheet and response slip) to all physiotherapists on their professional database ($n = 25$). An invitation was also placed on the interactive Chartered Society of Physiotherapy (iCSP) online professional network, with a request to contact the lead researcher (SM) for further information if they were interested. The prospective target sample was again $n = 10$ (Sandelowski, 1995). It was anticipated that physiotherapists would be purposively selected by gender and years qualified however, due to the small number of responses, all were interviewed.

2.4. Interviews

Interviews were arranged by telephone and confirmed by e-mail. The aim was to learn what stakeholders thought about the BioH questionnaire. To achieve this, relevant 'manufactured data' (Silverman, 2013) was captured through interviews. Written informed consent was provided by participants in advance and verbal consent was confirmed at the beginning of each interview. A copy of the BioH questionnaire was sent to participants in preparation for the interview. The questionnaire is available as additional online material in the original report of its development and validation (Palmer et al., 2017a). Patient and physiotherapist interviews were undertaken concurrently. The interviewer (SM) was a very experienced postdoctoral researcher with extensive qualitative research experience, although this was her first involvement with JHS research. As such, she had no pre-conceptions.

2.5. Environment

All one-to one interviews were undertaken over the telephone

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