Hypermobility: Perspectives on Physiotherapy (HPoP) – Study Protocol

Project title: Designing a comprehensive physiotherapy intervention for adults with joint hypermobility: focus groups

Short title: Hypermobility: Perspectives on Physiotherapy (HPoP)

Sponsor: North Bristol NHS Trust

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Study Sites (NHS):
North Bristol NHS Trust (for recruitment of NHS patients and clinicians and conduct of focus groups)
Royal National Hospital for Rheumatic Diseases NHS Foundation Trust, Bath (for recruitment of NHS patients and clinicians and conduct of focus groups)

Study Sites (other):
University of Hertfordshire (for recruitment of people with joint hypermobility and clinicians and conduct of focus groups)
Bournemouth University (for recruitment of people with joint hypermobility and clinicians and conduct of focus groups)
University of the West of England (for data analysis and storage)
University of Bristol (for data analysis and storage)

Local Collaborators (NHS):
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Abbreviations:
BU = Bournemouth University
HMSA = Hypermobility Syndrome Association
JHS = Joint Hypermobility Syndrome
NBT = North Bristol NHS Trust
RCT = Randomised Controlled Trial
RNHRD = Royal National Hospital for Rheumatic Diseases NHS Foundation Trust
UoB = University of Bristol
UoH = University of Hertfordshire
UWE = University of the West of England, Bristol

Research objective

This study is part of a larger research project which aims to develop and pilot a comprehensive physiotherapy intervention for adults with joint hypermobility syndrome (JHS) and then to test this in a feasibility randomised controlled trial (RCT). The present study will address the first of these objectives, specifically to:

- Develop a comprehensive physiotherapy intervention for adults with JHS informed by patient and clinician focus groups

Background

Joint Hypermobility Syndrome (JHS) (commonly people with this condition are described as being 'double-jointed') is characterised by excessive joint range of motion and pain (Grahame 2003). It affects up to 5% of women and 0.6% of men (Simpson 2006), although the true prevalence is difficult to determine due to historical variation in the criteria for diagnosis. The Brighton criteria (Grahame et al 2000) are now widely used for diagnosis. The condition is associated with a wide range of problems including pain; decreased muscle strength; reduced awareness of where the limbs are in space; changes in the pattern of walking; anxiety; reduced fitness; reduced function; and reduced quality of life (Grahame 2003).

Most of the identified problems are clearly related to physical function. Physiotherapy (particularly exercise) is therefore a mainstay of treatment for JHS and there seems to be a growing interest in this area of clinical practice. The research evidence is at a very early stage of development, however, with a recent systematic literature review conducted by this research team (publication in preparation) identifying only three exercise studies in adults which met the inclusion criteria (Barton and Bird 1996, Ferrell et al 2004, Sahin et al 2008).

These studies seem to suggest that patients with JHS improve over time with exercise but it is important to note that no study to date has included an appropriate control condition against which exercise was compared. Although Sahin et al (2008) included a control group, there was no statistical analysis of between group differences reported and fundamental methodological details are unclear. Improvements in outcomes could therefore be explained by natural history of the condition, positive interactions with the therapists, or other unknown factors. The true effectiveness of physiotherapy (including exercise) in JHS therefore remains unknown and an appropriately controlled study is urgently required.

It is clear from the literature (Grahame 2003) and from previous focus groups conducted by our research group that people with JHS encounter wide-ranging difficulties. Physiotherapy interventions therefore need to address these complex problems but there is little consensus on best practice. The proposed study will

therefore conduct focus groups with both patients and clinicians to establish perspectives on physiotherapy treatment. This information, in conjunction with knowledge from the available literature, will inform development of a comprehensive physiotherapy intervention package for adults with JHS which will be tested in a later feasibility RCT.

Research methods

In order to examine the views and experiences of physiotherapy for JHS, we will conduct a series of focus groups with patients and clinicians. Qualitative methods have been chosen as the most appropriate means of gathering data regarding beliefs, experiences and perceptions of physiotherapy interventions (Dowswell et al 2002, Galvin et al 2009). Qualitative methods are valuable in the pre-trial development phase to both help develop and refine the trial and improve our understanding of the experiences of patients receiving, and staff delivering, an intervention (Campbell et al 2000, Donovan et al 2002, Lewin et al 2009). Such use of qualitative methods in randomised controlled trials, specifically as part of pre-intervention development and post hoc interpretation, is well established (Finch 2003, Flottorp 2003, Sandelowski 1996, Toroyan 2004) and recommended (MRC 2008). Focus groups permit sharing and comparing of ideas amongst group members, which then facilitates the evaluation and interpretation of those ideas and the exploration of areas of consensus and disagreement.

People with JHS and health care professionals with a special interest in managing patients with JHS will be invited to take part in a series of focus groups at four specialist centres across the UK. Separate focus groups will be conducted with patients and then with health care professionals (eight focus groups in total). The professional groups are likely to comprise mainly physiotherapists but might also include occupational therapists and rheumatologists. Each focus group will comprise of between 6-8 participants, will be facilitated by two members of the research team and, with informed consent, audio recorded. Topic guides will be used to facilitate the focus groups which will explore:

1. what is considered ‘usual care’
2. the content and delivery of an education package;
3. the content and delivery of an exercise intervention;
4. mechanisms to support patients and enhance long-term engagement;
5. most appropriate outcomes for the intervention;
6. content of the control arm for the randomised controlled feasibility trial;
7. content and delivery of a training package for physiotherapists.

They will be asked to discuss what is considered to be ‘standard care’ for JHS as there is little known about practice variability across the UK. This will help to inform the proposed control intervention for the feasibility RCT and any future full RCT.

Discussion will take place within the context of a number of guiding principles. These are related to the resource context within which most physiotherapy services operate but mirror best practice as conducted at North Bristol NHS Trust (NBT). Firstly, the intervention will be one-to-one as the needs of individual patients are varied. Secondly, it should be easy to implement in any outpatient department (this is therefore likely to exclude resource-intensive interventions such as hydrotherapy). It is anticipated that a ‘menu’ of interventions will be developed, with the entry level tailored to individuals patients based on their readiness to change and functional

capacities. Thirdly, it should include a maximum of six treatment sessions over 4 months.

The project steering group, incorporating service users, will develop the intervention package based on the results of the focus groups and other published material, with ongoing input from focus group members should they wish to be involved. A training package for physiotherapists will also be developed.

Recruitment

NHS site recruitment:

People with JHS: At the Royal National Hospital for Rheumatoic Diseases (RNHRD) and NBT, people with JHS will be recruited through mailing of information packs to all patients with JHS who have been seen by the physiotherapy service within the previous 12 months. Appropriate patients will be identified by the clinical teams from treatment records. The packs will include a response slip which potential participants will return to the research team if they are interested in being involved. Potential participants will then be contacted to arrange a mutually convenient time to conduct the focus group.

Clinicians: At the RNHRD in Bath and NBT, clinicians will be recruited through mailing of information packs to clinicians with an interest in joint hypermobility. Appropriate individuals will be identified by Sin-ti Towlson (RNHRD) and Rachel Lewis (NBT) who are both Clinical Specialist Physiotherapists in Rheumatology. The packs will include a response slip which potential participants will return to the research team if they are interested in being involved. Potential participants will then be contacted to arrange a mutually convenient time to conduct the focus group.

University site recruitment:

People with JHS: At the University of Hertfordshire (UoH) and Bournemouth University (BU), people with JHS will be recruited through mailing of information packs to local members of the Hypermobility Syndrome Association (HMSA) and local patients who have previously expressed an interest in assisting with research activity. Appropriate individuals will be identified by Dr Jane Simmonds who is a medical advisor for the HMSA and the Physiotherapy Research Lead at the University of Hertfordshire, Carol Clark who is a Lecturer in Physiotherapy and joint hypermobility researcher at Bournemouth University, and Donna Wicks who is the Senior Medical Liaison Officer at the HMSA. The packs will include a response slip which potential participants will return to the research team if they are interested in being involved. Potential participants will then be contacted to arrange a mutually convenient time to conduct the focus group.

Clinicians: At the University of Hertfordshire and Bournemouth University, clinicians will be recruited through mailing of information packs to academic staff, postgraduate students and local clinicians with an interest in joint hypermobility. Appropriate individuals at the University of Hertfordshire will be identified by Dr Jane Simmonds who is a medical advisor for the Hypermobility Syndrome Association (HMSA) and the Physiotherapy Research Lead at the University of Hertfordshire. Appropriate individuals at Bournemouth University will be identified by Carol Clark who is a Lecturer in Physiotherapy and joint hypermobility researcher at Bournemouth University. The packs will include a response slip which potential participants will return to the research team if they are interested in being involved. Potential participants will then be contacted to arrange a mutually convenient time to conduct the focus group.

Inclusion/exclusion criteria:

People with JHS: Inclusion criteria: More than 18 years old; able to give informed consent; able to understand and communicate in English; physiotherapy treatment for JHS received within the previous 12 months. Exclusion criteria: Failure to meet the inclusion criteria; other known musculoskeletal pathology causing pain, particularly osteoarthritis and inflammatory musculoskeletal disease such as rheumatoid arthritis.

Clinicans: Inclusion criteria: More than 18 years old; able to give informed consent; able to understand and communicate in English; actively involved in the assessment and management of people with JHS. Exclusion criteria: Failure to meet the inclusion criteria.

Participant sampling:

In the event of more people than required volunteering, participants will be purposively sampled to attempt to capture maximum variation in views and experiences in order that they adequately reflect those of a range of patients and clinicians. Patients will be purposively sampled in relation to socio-demographic variables (age, gender). Physiotherapists will be purposively sampled in relation to length of time since qualification and gender.

Proposed sample size:

Sample size will be determined by the need to achieve data saturation, such that no new themes are emerging from the data by the end of data collection (Sandelowski, 1995). For the focus groups this is likely to include up to 32 patients and 32 staff.

Focus group conduct

Topic guides will be used in order to assist questioning during focus groups. The topic guides, devised to guide, but not dictate data collection, will incorporate considerable flexibility to allow participants to introduce new issues unanticipated by the researchers. Topic guides will be modified as necessary throughout the course of the study to reflect findings as they emerge. The researcher will use open-ended questioning techniques to elicit participants' own experiences and views and participants will be asked to provide examples. Focus groups are expected to last a maximum of 2 hours.

Ethical arrangements

Risks and anticipated benefits for trial participants:

The only disadvantage is likely to be the time it takes for the discussion group. It will be made clear that taking part will not affect, in any way, patient care or staff employment. Although participants will not benefit directly, taking part will help the researchers to understand participants' views about physiotherapy for the treatment of hypermobility in order to develop a treatment package. What is learned from the research may therefore help the future treatment of people with joint hypermobility.

Informing potential participants of possible benefits and known risks:

All trial participants will receive an information sheet and will be provided with an opportunity to discuss any aspects of this.

Obtaining informed consent from participants:

Fully informed written consent will be sought from all participants according to established principles of Good Clinical Practice (GCP). The chief investigator will be responsible for informed consent. Where fully informed consent is not possible, this will act as an exclusion criterion.

Proposed time period for retention of relevant trial documentation:

Researchers employed on the project will be responsible for data collection, recording and quality. This will be overseen by the Chief Investigator, Dr Shea Palmer.

Identifiable clinical data will remain in the physiotherapy departments at NBT and RNHRD. Consent forms, audio recordings and anonymous completed questionnaires will be kept in a locked filing cabinet at the University of the West of England, Bristol. Anonymised electronic transcripts will be kept on password protected computers at the University of the West of England, Bristol (Dr Shea Palmer) and the academic rheumatology department, Clinical Sciences at North Bristol (Dr Jeremy Horwood). Data will be collected and retained in accordance with the Data Protection Act (1998). Back-up copies of electronic data will be made regularly onto a CD, and also kept on the University of West of England, Bristol mainframe. All source documents will be retained for a period of 5 years following the end of the study.

Direct access to the source data and documents will be permitted for monitoring, audits, Research Ethics Committees and review. All trial documents will be made available on request for monitoring and audit by NBT, RNHRD and the relevant Research Ethics Committee.

Proposed action to comply with 'The Medicines for Human Use (Clinical Trials) Regulations 2004':

This trial will not be subject to these regulations as it does not involve medicinal products.

However the study will be monitored and audited in accordance with NBT and RNHRD policies. The project steering group will consist of an independent chair, two research partners and members of the research team. This group will meet four times per year to assess the study.

Adverse events are not envisaged due to the nature of the interventions but any such events will be recorded in accordance with NBT and RNHRD Research Related Adverse Event Reporting Policies. This is an NHS-sponsored research study. For NHS sponsored research HSG(96)48 reference no. 2 refers. If there is negligent harm during the clinical trial when the NHS body owes a duty of care to the person harmed, NHS Indemnity covers NHS staff, medical academic staff with honorary contracts, and those conducting the trial. NHS Indemnity does not offer no-fault compensation and is unable to agree in advance to pay compensation for non-negligent harm. Ex-gratia payments may be considered in the case of a claim.

Qualitative Data Analysis

With informed consent from participants, data will be audio-recorded, fully transcribed and anonymised, checked for accuracy and then imported into the software package Atlas.ti (Muhr 2004). Analysis will begin shortly after data collection starts, will be ongoing and iterative. Analysis will inform further data collection: for instance, analytic insights from data gathered in earlier interviews will shape the questions covered during later data collection.

Thematic analysis (Braun and Clarke 2006) will be used to scrutinise the data in order to identify and analyse patterns and themes of particular salience for participants and across the dataset using constant comparison techniques (Glaser and Strauss 1967, Charmay 2006). Firstly, the transcripts will be read several times, to gain familiarisation with the data and initial ideas noted. The transcripts will then be examined on a line-by-line basis with Inductive codes being assigned to the segments of the data that provide insight into the participants' views and understanding of their experiences. An initial coding frame will be developed and new data will be compared initially to previous data, and then to the properties of emerging categories that contain the main themes. The process of constant comparison will allow for the generation of new themes, re-classify themes and incorporating themes within other themes (Glaser and Strauss 1967, Charmay 2006) and the coding frame will be modified, if needed, as analysis develops. The data will be scrutinised for negative cases and reasons for the deviance will be explored by comparison with the whole dataset. Analysis of the focus group data will also give attention to the form of the data and the processes by which the group constructed their views (for example areas of consensus and disagreement).

A subset of transcripts will be independently double-coded by other members of the research team and compared; any discrepancies will be discussed and resolved in order to achieve a coding consensus.

Research Governance

This study will be performed subject to Research Ethics Committee (REC) approval, including any provisions of Site Specific Assessments (SSA) and local Research and Development Department approval. This study will be conducted in accordance with the Research Governance Framework for Health and Social Care and Good Clinical Practice.

Service users

Extensive consultation has been conducted with patient research partners who are members of an ongoing hypermobility research project steering group. Our current steering group includes a person with joint hypermobility who acts as an advisor to the Hypermobility Syndrome Association and is professionally qualified as an occupational therapist. Our patient research partners have enthusiastically endorsed the proposed research concept and design. The 'Patient Experience Partnership in Research' group at North Bristol NHS Trust Musculoskeletal Research Unit have also commented on the research design and are fully supportive of all aspects of the proposed study. Service users will be actively engaged in all aspects of the proposed study. Two service users will be invited to input to all aspects of the conduct of the study as full members of a steering group. A lay summary of the research will be given to the HMSA for further dissemination. Service user time and expenses have been fully costed according to INVOLVE guidance (INVOLVE 2010).

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Project timetable and milestones

2012									**Sep**	**Oct**	**Nov**	**Dec**
Ethics and R&D approval				X	X	X	X	X				
Recruit focus groups									X	X		
Conduct focus groups										X	X	X
Analyse focus groups & develop intervention											X	X
2013	**Jan**	**Feb**	**Mar**	**Apr**	**May**	**Jun**	**Jul**	**Aug**	**Sep**	**Oct**	**Nov**	**Dec**
Develop intervention (cont.)	X	X	X									