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Multidisciplinary intervention for treatment of benign joint hypermobility syndrome in childhood

Submission date
18/01/2017

Recruitment status
No longer recruiting

Registration date
09/03/2017

Overall study status
Completed

Last Edited
09/01/2019

Condition category
Musculoskeletal Diseases

[ ] Prospectively registered
[ ] Protocol
[ ] Statistical analysis plan
[X] Results
[ ] Individual participant data

Plain English summary of protocol

Background and study aims
Children with hypermobile joints (joints with a greater than normal range of motion) are more likely to have joint and muscle symptoms including growing pains and pain when exercising, which can become disabling in the long term and have an impact on the child's family, social life and schooling. Hypermobility occurs in up to 30% of children aged between 5 and 17 and accounts for a large number of referrals to paediatric rheumatology clinics, where it is called Benign Joint Hypermobility Syndrome (BJHS). It is often poorly recognised and takes a long time to diagnose. Effective treatment involves lifestyle changes, altering the patient's exercise habits, joint protection, and improving body mechanics. More evidence is required of how well these treatment strategies work and their costs (clinical and cost effectiveness). Although multidisciplinary treatment involving physiotherapists, occupational therapists, and the provision of education to parents, schools and the community are all thought to be important, the extent to which these approaches are used in clinical practice varies widely. The aim of this study is to assess the clinical and cost effectiveness of a structured multidisciplinary team approach for the treatment of childhood BJHS.

Who can participate?
Children aged between 5–16 years old with pain due to BJHS

What does the study involve?
All participants and their parents/guardians are invited to attend an appointment with a physiotherapist at the hospital, who does a physical assessment of the child and completes some questionnaires with both the child and parent. This appointment takes around 1 hour. Participants are randomly allocated into one of two groups: standard treatment or structured treatment. If the participant is in the standard treatment group the child does not receive any further treatment. He/she has already had an appointment with the paediatrician and has been given information and advice about BJHS and referred for orthotic support if necessary, but if the child has any problems and needs to see the doctor again this is arranged. This is what would usually happen with children diagnosed with BJHS. If the participant is in the structured treatment group, they take part in a structured treatment programme. The four treatment visits

include exercises and advice and take around 30 to 60 minutes each. Participants in both groups attend two more appointments with the physiotherapist 3 months and 1 year later to repeat the physical assessment and questionnaires. These appointments also take around 1 hour each time.

What are the possible benefits and risks of participating?
It is hoped that the multidisciplinary programme reduces pain and improves physical activity and participation. Improving the treatment of children with BJHS may improve not only their health but also their wellbeing and social and educational development. However, this is not guaranteed and the treatment may be of no benefit. This is the first formal study of treatment in BJHS, and the results may lead to better and more effective ways of managing this condition, raise awareness of BJHS in hospital and general practice, and help with the planning of hospital and community services to support children with this common and disabling condition. None of the treatments present a physical risk to patients, but it is possible that the exercise may make symptoms worse in some children. However, they are closely supervised and the programme of treatment is prepared by an experienced therapist and tailored to their needs. Attendance for follow up may represent a burden. However, the schedule is very similar to treatment schedules commonly used with children with many physical conditions and is generally well tolerated. Travel costs are covered.

Where is the study run from?
Norfolk and Norwich University Hospital NHS Foundation Trust (UK)

When is the study starting and how long is it expected to run for?
September 2010 to June 2014

Who is funding the study?
NIHR Research for Patient Benefit Programme (UK)

Who is the main contact?
Prof. Alexander Macgregor
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Contact information

Type(s)
Public

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Additional identifiers

EudraCT/CTIS number

IRAS number

ClinicalTrials.gov number

Secondary identifying numbers
CPMS ID: 9366

Study information

Scientific Title
The efficacy and cost effectiveness of a multidisciplinary intervention strategy for the treatment of benign joint hypermobility syndrome in childhood

Acronym
BENDY

Study objectives
This study aims to determine the clinical and cost effectiveness of a multidisciplinary care package in children diagnosed with benign joint hypermobility syndrome (BJH). Specifically, the aim is to conduct a clinical trial to compare a structured programme of treatment delivered through a multidisciplinary team of clinicians, physiotherapists and occupational therapists with the usual care comprising a single interview with a clinician.

Ethics approval required
Old ethics approval format

Ethics approval(s)
Norfolk Research Ethics Committee, 23/11/2009, ref: 09/H0310/80

Study design
Randomised controlled single-site study

Primary study design
Interventional

Secondary study design
Randomised controlled trial

Study setting(s)
Hospital

Study type(s)
Treatment

Participant information sheet
See additional files

Health condition(s) or problem(s) studied
Benign joint hypermobility syndrome

Interventions
Those consenting to take part will be invited for a screening assessment undertaken by the trial physiotherapist to identify their eligibility for inclusion. Written consent will be sought from eligible children and their parents. Those consenting to take part will be randomised to receive either multidisciplinary care or standard care. Minimisation will be used to ensure balance among age and gender strata.

The multidisciplinary intervention will comprise an appointment with a physiotherapist for assessment and basic advice; a programme of 6 physiotherapy sessions to improve core stability and work on strengthening exercises; gait analysis and provision of foot orthoses as necessary; an OT assessment at hospital clinic and at home with provision of appliances as necessary; an OT school visit to discuss diagnosis with teachers and provide appliances as necessary. Those with problems in their hands will also receive an OT directed exercise programme.

The groups will be reviewed at 3 and 12 months to assess which treatment approach was of greatest benefit in helping their pain and improving their physical functioning and quality of life. Data will also be collected on function, social activities and schooling. Data will also be collected that will allow a health economic evaluation.

Intervention Type
Other

Primary outcome measure
Level of pain and physical functioning, assessed by the Childhood Health Assessment Questionnaire (CHAQ) at baseline, 3 months and 12 months

Secondary outcome measures
Measured at baseline, 3 months and 12 months:
1. Health-related quality of life, assessed using the CHU 9D (9 dimensional Child Health Utility)
2. Number of primary care and hospital attendances and school attendance, measured using the BJH resource use questionnaire
3. Grip strength, assessed using a hand-held dynamometer
4. Pain, assessed using the VAS/Faces Pain Score
5. Function and gross and fine motor skills, assessed using the Movement Assessment Battery for Children
6. Participation in sport, measured using a questionnaire

Overall study start date
20/09/2010

Completion date
19/06/2014

Eligibility

Key inclusion criteria
1. Children aged between 5–16 years old
2. Satisfy the Brighton criteria for classification of BJS

3. Reporting musculoskeletal pain in one or more areas of the body for a duration of greater than three months
4. Written informed consent

Participant type(s)
Patient

Age group
Child

Lower age limit
5 Years

Upper age limit
16 Years

Sex
Both

Target number of participants
120

Key exclusion criteria
1. Inflammatory joint disease
2. Identifiable heritable disorder of connective tissue
3. Presence of other chronic illness

Date of first enrolment
21/10/2010

Date of final enrolment
31/12/2012

Locations

Countries of recruitment
England

United Kingdom

Study participating centre
Norfolk and Norwich University Hospital NHS Foundation Trust
United Kingdom
NR4 7UY

Sponsor information

Organisation
Norfolk and Norwich University Hospital NHS Foundation Trust

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Sponsor type
Hospital/treatment centre

ROR
https://ror.org/01wspv808

Funder(s)

Funder type
Government

Funder Name
NIHR Research for Patient Benefit Programme

Results and Publications

Publication and dissemination plan
Planned publication in a high-impact peer-reviewed journal. In October 2014 a letter was posted to all participants with a document summarising the results of the study.

Intention to publish date
09/03/2018

Individual participant data (IPD) sharing plan
The datasets generated during and/or analysed during the current study are/will be available upon request from Prof. Alexander Macgregor (a.macgregor@uea.ac.uk).

IPD sharing plan summary
Available on request

Study outputs

Output type	Details	Date created	Date added	Peer reviewed?	Patient-facing?

Participant information sheet		18/10/2010	09/03/2017	No	Yes
Participant information sheet		02/01/2009	09/03/2017	No	Yes
Participant information sheet		18/10/2010	09/03/2017	No	Yes
Participant information sheet	version V4.0	18/10/2010	09/03/2017	No	Yes
Results article	results	01/12/2019		Yes	No