CHILDHOOD DISABILITY RESEARCH PRIORITY SETTING PARTNERSHIP

PROTOCOL (Version 2.1 September, 2012)

Purpose

The purpose of this protocol is to set out the aims, objectives and commitments of the Childhood Disability Research Priority Setting Partnership (PSP) and the basic roles and responsibilities of the partners therein.

Steering Group

The Childhood Disability Research PSP will be led and managed by the following:

Family representatives:

- Mary Busk, National Network of Parent Carer Forums
- Anna Walker, National Network of Parent Carer Forums

Third Sector representative:

- Amanda Allard, Council for Disabled Children

Clinical representative/s:

- Chris Morris, Allied Health Professional & Senior Research Fellow in Child Health, University of Exeter
- Doug Simkiss, Consultant Paediatrician & Associate Professor in Child Health, University of Warwick

The Partnership and the priority setting process will be supported and guided by:

- Katherine Cowan, The James Lind Alliance (JLA), Chair of the Childhood Disability Research PSP
- Mark Fenton, UK Database of Uncertainties about the Effects of Treatments (UK DUETs)

The Steering Group includes representation of patient/carer groups and clinicians. The Steering Group will agree the resources, including time and expertise that they will be able to contribute to each stage of the process. The JLA will also advise on this. Consideration is being given about how to meaningfully and usefully involve young people affected by neurodisability in the Childhood Disability Research PSP, perhaps through a consultation group with representation on the Steering Group and/or in key stages of the project.

Background to the Childhood Disability Research PSP

Patients and members of the public are encouraged to engage in research activities. Public and Patient Involvement (PPI) is mandatory for research that is funded by the National Institute for Health Research and is both philosophically correct and believed to confer pragmatic benefits that improve research.

Key issues in applied health research are (i) what research questions are addressed and investigated, and (ii) who decides which research questions are most important. In the past it was researchers who decided the research topics; increasingly, there is a role for patients and clinicians to influence research agendas.

The JLA is a project which is funded by the National Institute of Health Research with support from the Medical Research Council. Its aim is to provide an infrastructure and process to help patients and clinicians work together to agree which are the most important treatment uncertainties affecting their particular interest, in order to influence the prioritisation of future research in that area. The JLA defines an uncertainty as a "known unknown" – in this case relating to the effects of treatment.

The British Academy of Childhood Disability Research (BACD) is an organisation principally for professionals working with young people affected by neurodisability. The BACD operates as an affiliate group of the British Association of Community Child Health, a specialty group of the Royal College of Paediatrics and Child Health. The BACD has a Strategic Research Group (SRG, www.bacdis.org.uk/research). One of the four key aims of the SRG is to:

- To encourage and assist people in the field, including families, young people and clinicians, to identify research priorities.

The JLA methodology provides a means for the BACD SRG to fulfil this aim.

A proposal for a Childhood Disability Research PSP was considered by the BACD Executive Committee in March 2012. The committee has made available a fixed sum of up to £20K from the Paul Polani Fund to establish and run a Childhood Disability Research PSP following the James Lind Alliance (JLA) methods and focusing on neurodisability.

Aims and objectives of the Childhood Disability Research PSP

The aim of the Childhood Disability Research PSP is to identify the unanswered questions about the effectiveness of interventions for children and young people affected by neurodisability from patient and clinical perspectives, and then to prioritise those that patients and clinicians agree are the most important.

The objectives of the Childhood Disability Research PSP are to:

- work with patients, their parents and primary carers, and clinicians to identify uncertainties about the effectiveness of treatments for childhood disability
- to agree by consensus a prioritised list of those uncertainties, for research
- to publicise the results of the PSP and process
- to take the results to research commissioning bodies to be considered for funding

Scope

Any ways to improve the health [1] and/or wellbeing [2] of children with neurodisability [3] about which there is uncertainty of the effectiveness [4] of the intervention, therapy or procedure [5] (see notes below).

1. Health is defined by the 'components of health' described by the World Health Organization in the International Classification of Functioning Disability and Health (figure 1); namely body functions & structures, activities and participation, further details are provided in Appendix 1.
2. Wellbeing is defined by how people feel about their life and their ability to reach their aspirations.
3. For the purposes of this project: "Neurodisability includes a group of chronic conditions with a broad range of severity and complexity, some of which can vary over time. Neurodisability is a consequence of impairment of the brain, central or peripheral nervous system that creates activity limitations. This may result in physical difficulties (such as cerebral palsy), learning difficulties (such as intellectual disorders), and social/communication difficulties (such as in autism), or other medical conditions (such as the problems associated with epilepsy). Sensory, behavioural and emotional difficulties are all included if they are a consequence of neurological impairment. Some children affected by neurodisability are diagnosed with specific syndromes and conditions, whilst others are not given a named diagnosis."
   [Taken from www.pencru.org/project_chums.php with permission]

4. Uncertainty of the effectiveness is verified systematically by reference to published systematic reviews and other publications.

5. Interventions, therapy or procedures [technically any environmental factor in the ICF language] that can be expected to impact on and improve health.

Uncertainties raised should identify

- The population of children, either generic or specific to a diagnosis or type of impairment, and/or a particular age group if appropriate.
- Description of the type of intervention, therapy or procedure.
- The likely positive health outcome.

The scope incorporates an applied health research model; therefore there is an expectation that, if shown to be effective in research, the intervention or procedure could be expected to benefit the health and wellbeing of children with neurodisability within 3-5 years of the results.

Partners

Organisations and individuals will be invited to take part in the PSP, which represent the following groups:

- children and young people affected by neurodisability
- parents and primary carers of young people affected by neurodisability
- professionals with experience of working with children and young people affected by neurodisability

It is important that all organisations which can reach and advocate for these groups should be invited to become involved in the PSP. The JLA will take responsibility for ensuring the various stakeholder groups are able to participate equally to the process.

Organisations wishing to participate in the PSP are required to affiliate to the JLA in order to demonstrate their commitment to the aims and values of the JLA. Details on the affiliation procedure can be found at www.lindalliance.org.

Exclusion criteria

Some organisations may be judged by the JLA or the Steering Group to have conflicts of interest. These may be perceived to adversely affect those organisations' views, causing unacceptable bias. As this is likely to affect the ultimate findings of the PSP, those organisations will not be invited to participate. It is possible, however, that interested parties may participate in a purely observational capacity when the Steering Group considers it may be helpful.

METHODS

This section describes a schedule of proposed stages through which the PSP aims to fulfil its objectives. The process is iterative and dependent on the active participation and contribution of different groups. The methods adopted in any stage will be agreed through consultation between the partners, guided by the PSP's aims and objectives. More details and examples can be found at www.JLAguidebook.org.

1. Identification and invitation of potential partners

Potential partner organisations will be identified through a process of peer knowledge and consultation, through the Steering Group members' networks and through the JLA's existing register of affiliates. The Steering Group will assemble lists of:

- Relevant professional societies.
- Relevant childhood disability and condition-specific organisations supporting children affected by neurodisability and their families.
- Funders of childhood disability research.

Potential partners will be contacted and informed of the establishment and aims of the Childhood Disability Research PSP and invited to participate in awareness raising activities.

The JLA will draft and sign the invitations which will be distributed by the BACD administrator Kelly Robinson.

2. Stakeholder awareness raising activities

The stakeholder awareness raising programme will have several key objectives:

- to create awareness of the Childhood Disability Research PSP
- to outline the proposed plan and processes for the PSP
- to identify potential family and professional partner organisations which will commit to the PSP and identify individuals who will be those organisations' representatives and the PSP's principal contacts
- to establish principles upon which an open, inclusive and transparent mechanism can be based for contributing to, reporting and recording the work and progress of the PSP

The administrative process for all stakeholder awareness raising activities will be managed by the Steering Group in coordination with the JLA.

Organisations which have decided to participate in the PSP will be asked to complete a declaration of interests, including disclosing relationships with the pharmaceutical industry or other commercial enterprises.

3. Identifying uncertainties

Partners will advertise the opportunity to propose questions and uncertainties of practical clinical importance to their members and associates relating to the treatment and management of childhood disability. A fixed period of 3-6 months will be given to complete this exercise (most likely November 2012 to April 2013).

The methods may be designed according to the nature and membership of each partner organisation, but must be as transparent, inclusive and representative as practicable. Potential methods include membership meetings, email consultation, postal or web-based questionnaires, internet message boards and focus group work.

Existing sources of information about treatment uncertainties for patients and clinicians will be searched. These can include question-answering services for patients and carers and for clinicians; research recommendations in systematic reviews and NICE and/or other clinical guidelines; protocols for systematic reviews being prepared, and registers of ongoing research. Doug Simkiss will coordinate these activities supported by postgraduate students.

The centralised collection and cataloguing of uncertainties will be managed by the BACD administrator, Kelly Robinson, using survey monkey.

4. Refining questions and uncertainties

The consultation process will produce “raw” questions about the effectiveness of treatments and interventions. These raw questions will be assembled and categorised and refined by Chris Morris & Doug Simkiss into “collated indicative questions” which are clear, addressable by research and understandable to all. Similar or duplicate questions will be combined where appropriate.

The existing literature will be researched by members of the BACD Strategic Research Group and their teams to assess to what extent these questions have, or have not, been answered by previous research; i.e. are they ‘uncertainties’?

Sometimes, uncertainties are expressed that can in fact be resolved with reference to existing research evidence - ie they are "unrecognised knowns" and not uncertainties. If a question about treatment effects can be answered with existing information but this is not known, it suggests that information is not being communicated effectively to those who need it. The Steering Group will assemble lists of these “unrecognised knowns” and seek to publicise them online and through the associated PSP dissemination activities.

Uncertainties relating to treatment which are not adequately addressed by previous research will be agreed by the BACD Strategic Research Group and PSP Steering Group. These will then be collated and entered into a Childhood Disability Research section within the UK Database of Uncertainties about the Effects of Treatments (UK DUETs - www.library.nhs.uk/duets) under the supervision of Mark Fenton. Alternative arrangements will be made for non-treatment uncertainties. This will ensure that the uncertainties have been actually checked and confirmed to be uncertainties. This is the responsibility of the BACD Strategic Research Group and PSP Steering Group.

5. Prioritisation – interim and final stages

The aim of the final stage of the priority setting process is to prioritise through consensus the identified uncertainties relating to the treatment or management of childhood Disability Research. This will be carried out by members of the Steering Group and the wider partnership representing patients and clinicians.

The interim stage, to proceed from a long list of uncertainties to a shorter list (e.g. up to 20), will be carried out over email, whereby organisations consult their membership and ask for a top 15-20 most important uncertainties, ranked or unranked.

The final stage, to reach, for example, 10 prioritised uncertainties, will be conducted at a face-to-face meeting, using group discussions and plenary sessions.

The methods used for this prioritisation process will be determined by consultation with the partner organisations and with the advice of the JLA. Methods which have been identified as potentially useful in this process include: adapted Delphi techniques; expert panels or nominal group techniques; consensus development conference; electronic nominal group and online voting; interactive research agenda setting and focus groups.

The JLA will facilitate this process and ensure transparency, accountability and fairness.

Findings and research

It is anticipated that the findings of the Childhood Disability Research PSP will be reported to funding and research agenda setting organisations such as the NIHR HTA and HS&DR programmes as well as the major research funding charities. Steering Group and BACD Strategic Research Group members and partners will develop the prioritised uncertainties into research questions, and to work to establish the research needs of those unanswered questions to use when approaching potential funders, or when allocating funding for research themselves.

Publicity

As well as alerting funders, partners and Steering Group members are encouraged to publish the findings of the Childhood Disability Research PSP using both internal and external communication mechanisms. The JLA may also capture and publicise the results, through descriptive reports of the process itself. This exercise will be distinct from the production of an academic paper, which the partners are also encouraged to do. However, production of an academic paper should not take precedence over publicising of the final results.

APPENDIX 1 - WAYS OF THINKING ABOUT HEALTH

The following are 'components of health' as defined by the World Health Organization in the International Classification of Functioning Disability and Health.

BODY FUNCTIONS & STRUCTURES

- Mental functions
- Nervous system
- Sensory functions & pain
- Ear & eye
- Voice and speech
- Cardiovascular, haematological, immunological and respiratory systems
- Immunological & respiratory systems
- Digestive, metabolic & endocrine systems
- Genitourinary & reproductive systems
- Neuromusculoskeletal & movement
- Skin & related structures
- Movement

ACTIVITY & PARTICIPATION

- General tasks & demands
- Communication
- Mobility
- Self care
- Domestic life
- Interpersonal interactions & relationships
- Major life areas
- Community, social & civic life
- Any other activity & participation

These 'components of health' are described in a little more detail on the following pages.

BODY FUNCTIONS & STRUCTURES

Mental functions - both global mental functions, such as consciousness, energy and drive, and specific mental functions, such as memory, language and calculation mental functions

Nervous system – brain, spinal cord and nervous system

Ear & eye structures, sensory functions and pain - senses, seeing, hearing, tasting and so on, as well as the sensation of pain.

Voice & speech - functions of producing sounds and speech

Cardiovascular, haematological, immunological systems - functions involved in the cardiovascular system (functions of the heart and blood vessels), the haematological and immunological systems (functions of blood production and immunity), and the respiratory system (functions of respiration and exercise tolerance).

Immunological & respiratory systems - functions involved in the cardiovascular system (functions of the heart and blood vessels), the haematological and immunological systems (functions of blood production and immunity), and the respiratory system (functions of respiration and exercise tolerance).

Digestive, metabolic & endocrine systems - functions of ingestion, digestion and elimination, as well as functions involved in metabolism and the endocrine glands and the growth maintenance functions

Genitourinary & reproductive systems - functions of urination and the reproductive functions, including sexual and procreative functions.

Neuromusculoskeletal & movement - functions of movement and mobility, including functions of joints, bones, reflexes and muscles.

Skin & related structures - functions of skin, nails and hair

Movement - moving by changing body position or location or by transferring from one place to another, gait functions including walking, running or climbing; also carrying or manipulating objects.

ACTIVITY & PARTICIPATION

General tasks and demands - carrying out single or multiple tasks, organizing routines and handling stress. These items can be used in conjunction with more specific tasks or actions to identify the underlying features of the execution of tasks under different circumstances.

Communication - general and specific features of communicating by language, signs and symbols, including receiving and producing messages, carrying on conversations, and using communication devices and techniques.

Mobility - moving by changing body position or location or by transferring from one place to another, by carrying, moving or manipulating objects, by walking, running or climbing, and by using various forms of transportation.

Self care - caring for oneself, washing and drying oneself, caring for one's body and body parts, dressing, eating and drinking, and looking after one's health.

Domestic life - carrying out domestic and everyday actions and tasks. Areas of domestic life include acquiring a place to live, food, clothing and other necessities, household cleaning and repairing, caring for personal and other household objects, and assisting others

Interpersonal interactions & relationships - carrying out the actions and tasks required for basic and complex interactions with people (strangers, friends, relatives, family members and lovers) in a contextually and socially appropriate manner.

Major life areas - carrying out the tasks and actions required to engage in education, work and employment and to conduct economic transactions.

Community, social & civic life - the actions and tasks required to engage in organized social life outside the family, in community, social and civic areas of life.

ENVIRONMENTAL FACTORS

These are generally the things we can use to modify health, i.e.

1. Products And Technology
Any product, instrument, equipment or technology adapted or specially designed for improving the functioning of a disabled person, including food, drugs and equipment.

2. Natural Environment And Human-Made Changes To Environment
The natural or physical environment, including aspects of the environment that have been modified by people.

3. Support And Relationships
People or animals that provide practical physical or emotional support, nurturing, protection, assistance and relationships to other persons, in their home, place of work, school or at play or elsewhere.

4. Attitudes
Attitudes are observable consequences of customs, practices, ideologies, values, norms, factual beliefs and religious beliefs. Attitudes influence individual behaviour and social life at all levels.

5. Services, Systems And Policies
- Services are structured programmes designed to meet the needs of individuals. Services may be public, private or voluntary.
- Systems are administrative mechanisms established to control services.
- Policies govern and regulate the systems that organise, control and monitor services.