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# Informing the NHS Outcomes Framework: what outcomes of NHS care should be measured for children with neurodisability?

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## Informing the NHS Outcomes Framework: what outcomes of NHS care should be measured for children with neurodisability?

### AIMS AND OBJECTIVES

This research will determine the outcomes of NHS care which should be measured for children with neurodisability and the extent to which they can be measured by existing patient-reported outcome measures (PROMs). To address this aim the study has the following objectives:

(i) To identify key healthcare outcomes, beyond measures of morbidity and mortality, that are regarded as important by children with neurodisability and parents;

(ii) To ascertain what outcomes of services health professionals think are important for this group and to assess the extent to which they agree with families' views;

(iii) To seek consensus between families and professionals on what health outcomes are important and assess the usefulness of candidate generic PROMs for routine use in the NHS.

(iv) To identify generic PROMs which have been used with children with neurodisability and identify which best map onto outcomes identified as most important by families and professionals.

(v) To evaluate evidence of the psychometric performance of these PROMs when used with children with neurodisability.

(vi) To make recommendations about the use of generic PROMs to measure healthcare outcomes for children with neurodisability.

### BACKGROUND

Estimates of the prevalence of childhood disability in the UK vary from 5-18% of children depending on the definition or indicator of disability. [1] Most commonly an estimate of 1 in 20 children is cited. [2] 'Neurodisability' is an umbrella term for conditions associated with impairment of the nervous system and includes conditions such as cerebral palsy, autism and epilepsy; it is not uncommon for neurological impairments to co-occur. Aside from asthma, neurodisability probably represents the largest proportion of significant childhood disability. [3] Although neurodisability represents a heterogeneous group of conditions, they share much in common in terms of healthcare needs, and neurodisability has recently been recognised as a paediatric subspecialty for training purposes. Children with neurodisability have a range of impairments some of which are relatively minor but many of these children have multiple and complex needs. As a consequence they are among the most frequent and intensive users of the NHS requiring care and support from health services across primary and community care, hospital services, and specialist centres. Although largely unable to cure the neurological damage/chronic impairments these children have, health services aim to maximise functioning, and maintain/improve the health and wellbeing of these children, most of whom can be expected to survive into adulthood. [4] Funding and provision of health services for disabled children are recognised to be highly variable and it is acknowledged that children "do not always get the attention and care from healthcare services that they need." [5, 6] Disabled children are known to face significant disadvantage and a number of initiatives have sought to improve health and social care provision e.g. National Service Framework, Aiming High for Disabled Children, Centre for Excellence and Outcomes in Children and Young People's Services (C4EO). The Every Child Matters Outcome Framework has provided a useful means to develop indicators assessing educational and social

care outcomes for children and, with adjustments, is appropriate and meaningful for disabled children. [7] However, it has been difficult to assess the impact of NHS care for disabled children as there is no overall measure of health outcomes. Kennedy recommended the need to identify a common vision between families and professionals for what services are seeking to achieve. [6] Hence identifying an outcome indicator of NHS performance for children with neurodisability would be timely and serve this purpose. Outcomes of a health condition or injury can be considered within the bio-psychosocial framework expressed through the World Health Organisation's International Classification of Functioning, Disability and Health (ICF). [8] The ICF classifies components of health and functioning, as body structures and functions, and activities and participation. Thus a disease or injury may lead to impairments of body structure or function, limitation in activities and/or restriction in participation. These impairments, limitations and restrictions are collectively referred to as disability within the ICF, and scales which measure such components are often referred to as measuring Health Status or Health Related Quality of Life (HRQoL). In the ICF the relationships between these components are mediated by environmental and personal factors. A key environmental factor is the role played by healthcare and the NHS.

The NHS Outcomes Framework is part of a strategy that aims to deliver 'the outcomes that matter most to people'. [9] Domain 2 of this framework will detail indicators of the 'quality of life of people with long term conditions'. Although much of the detail is still to be determined and will evolve over the coming years. [9] Proposed indicators include patient-reported outcome measures (PROMs) which, in practice, are largely targeted at the components of the ICF under the rubric of HRQoL. There is a wide range of generic and condition-specific PROMs for children. [10] Rather than aiming to have a PROM for every diagnosis, the Royal College of Paediatrics & Child Health have proposed, pragmatically, that there should be a single PROM "for a basket of conditions". [11] Structured reviews have identified generic and condition specific PROMs that can be used with children; [e.g. 10] and conceptual issues pertaining to the content validity of these for children with neurodisability have been discussed. [12, 13] However no reviews have comprehensively appraised published research about the psychometric performance of generic PROMs when used with this group of children. In the context of neurodisability, it is often difficult for healthcare to make changes in chronic impairments of 'body functions and structures' through. Consequently there may be a greater likelihood of health and social interventions maintaining or improving activity and/or the participation of children with neurodisability. Clearly the constructs assessed using PROMs should be those most appropriate to assessing the likely impacts of NHS care and must be credible to children and families. [14, 15] In related work, the life priorities of children with cerebral palsy were found to be broadly represented in the KIDSCREEN HRQoL questionnaire. [16] Identifying a single PROM for a "basket of conditions" requires first identifying and agreeing the construct of interest and then gathering evidence of psychometric robustness. [14] Ethically, children's own self reported health should be gathered. [14] Although there has been wide recognition that children's voices should be heard, this is often not the case. [17] In particular, the voices of disabled children are frequently overlooked. Chronological age is not a clear criterion for judging when children are capable to self-report their health by completing a questionnaire, although children aged eight years and above are widely believed to be competent. [14] Parent/carer proxy-reports

are the only way to assess outcomes for children cognitively unable to self-report but these do represent a different perspective to the child's own view. However, as it is parents who typically seek NHS care on their child's behalf they too should have an opportunity to report. Ideally, both children's and parents' reports should be collected so that both perspectives are represented. [14]

**NEED**

The identification of suitable outcome measures will improve the evaluation of integrated NHS care for the large number of children affected by neurodisability, and has the potential to encourage the provision of effective health and social interventions and improve the efficiency of services. *Health need*: The proposed work seeks to contribute to positively improving children's health outcomes by providing a high quality means for measuring them. In this context, identifying a common purpose for NHS services and appropriate outcome measures will help to ensure NHS resources are deployed to maximize effectiveness, in an efficient manner. [6] *Expressed need*: There are a number of strategic developments that express the need for the proposed study. The NHS Outcomes Framework is evolving; [8] currently the DH is running a competition inviting suggestions for potential indicators. [18] The response of the Royal College of Paediatrics & Child Health to the consultation on the NHS Outcomes Framework suggested that there should be a single PROM "for a basket of conditions", [10] represented in the current proposal by grouping children with neurodisability. *Sustained interest and intent*: There is a clear direction of travel whereby PROMs look set to be one of the key performance indicators in the UK and other health systems. [15]

*Capacity to generate new knowledge:* The Kennedy Report clearly stated the need for a common vision to be identified for children's services; [6] this is addressed as a central part of the proposed work. A number of studies have reviewed aspects of PROMs in the context of children with neurodisability; [12, 13] none have sought to address the research objectives in this proposal. *Organisational focus consistent with HSR mission*: Currently, identifying health outcomes is a major focus for the NHS; the proposed work will be an important research contribution and return on investment. The findings will have implications for children with some of the most common conditions who are frequent and intensive users of the NHS, and to whom they look to improve their health and wellbeing. The findings may have relevance outside the conditions grouped under neurodisability (i.e. a bigger basket of conditions), but this is not addressed in the present application. The team comprises applicants from centres around the UK and has substantial expertise and experience in research related to childhood disability and health outcomes. One benefit of conducting the proposed work will be to foster collaboration for further work in the future on outcomes in child health. Collaboration with the Council for Disabled Children and the British Academy of Childhood Disability ensure national representation in the project. *Generalisable findings and prospects for change*: The findings of this research have major policy and practical applicability to the NHS, both in terms of identifying a common vision between families and professionals as to the overall goal of services for children with neurodisability, and to identify a PROM (or more than one) for this large group of intensive users of NHS resources which could be considered for the NHS Outcome Framework portfolio. *Building on existing work*: There is a substantive programme of methodological and applied research regarding PROMs funded by NIHR, MRC and the DH. Much of the work has focused

on adults and less on children. A report commissioned by the DH, drafted by the lead applicant, concerned the feasibility of using PROMs with children routinely in the NHS. [14] A key recommendation was that, given the variability of the constructs assessed by different PROMs for children, it is crucial to define the purpose of measurement.

## METHODS

Three inter-related streams of work will deliver the objectives; initials of the relevant applicants in brackets.

**Stream 1: Structured literature review** (CM, JTC, CG, AT, CJ, KA)
Stream 1 is a structured review of PROMs conducted by a research fellow supported by the academic team at Peninsula Medical School, with additional input from methodological experts. The local team are part of the NIHR-funded Peninsula Collaboration for Leadership in Applied Health Research and Care (PenCLAHRC) and the team includes researchers with expertise in systematic reviews. N.B. This project was considered by the PenCLAHRC stakeholder prioritisation group and adopted onto PenCLAHRC research portfolio, providing access to these resources to support the project team. The review will identify candidate generic PROMs, and evaluate evidence to support psychometric performance of each PROM.

*Search strategy:* Candidate generic PROMs will be identified through multiple bibliographic databases, published reviews, and libraries of PROMs (e.g. ProQolid) and by contacting experts. A key data source will be the Oxford PROM bibliographic database, which comprises over 30,000 records relating specifically to PROMs. The content is based on systematic searches of published literature using a specially developed search strategy to identify PROMs across a broad spectrum of academic publications. The content is considered comprehensive up to December 2005. The Oxford PROM search strategy for identifying PROMs will be combined with filters to focus on children and neurodisability; i.e. using MeSH and thesaurus terms. The search will run across multiple databases (e.g. MEDLINE, EMBASE, CINAHL, PsycINFO, AMED, and the NHS Database of Economic Evaluations to identify utility measures) to identify citations of generic PROMs used with children since 2006. Development of the search strategies and management of the process will be supported by PenCLAHRC Evidence Synthesis Team & Information Specialists. The detailed strategies will be retained and recorded. The search results will be interrogated to ensure that key 'marker' papers are returned and if this is not the case, a clear explanation for the omission can be offered. Supplementary hand searching will include key methodology journals (e.g. Quality of Life Research), and paediatric & childhood disability journals. Search results will be kept in reference management software and the dates of searches recorded so they may be updated as required.
*Identification of PROMs*: The first stage of the review will be screening the references for names of generic PROMs, and then scoping the quantity of evidence likely to be available for each instrument. Inclusion criteria for candidate PROMs are that they measure health outcomes for children less than 18 years, are not condition-specific, and English language version have been evaluated. We have not stipulated a lower age restriction deliberately. Very young children will be unable to self-report, but children with neurodisability are frequently intensive users of NHS services following diagnosis (usually in infancy) and beginning school; thus it would not seem wise to

exclude this group at this stage. A review of PROMs for children in published in 2008 identified 30 generic instruments. [11] This provides a guide to the number of relevant PROMs likely to be found in the search; although we recognise that not all instruments might meet the eligibility criteria, and new instruments may have been developed.

*Defining the constructs measured*: The constructs assessed by each of the candidate generic PROMs to be included will be transcribed into lay terms to inform the qualitative work with families and professionals in Streams 2 & 3. We will contact the developers of the PROMs and collaborate with our parent co-applicant and children and families from the Cerebra Research Unit Family Faculty & Council for Disabled Children to ensure lay summaries are meaningful and accurately reflect the constructs, through piloting. In addition, the items/domains from each PROM will be classified according to the taxonomy of the ICF.

*Further searches*: Specific searches will then be developed for applications of the identified candidate PROMs with three exemplar conditions representing varying manifestations of neurodisability: (i) cerebral palsy (neuro-motor disorder); (ii) epilepsy (neurological), (iii) autism (neuro-psychiatric disorder).

*Review strategy:* The titles & abstracts of articles will be screened independently by two reviewers and those thought likely by either reviewer to yield evidence of the psychometric performance of candidate generic PROMs will be retrieved as full text. To be included there must be reference to a generic PROM, use with children aged under 18 years, and published in English language. Studies will be categorised as being development papers, specifically evaluating the measurement properties of PROMs, or alternatively papers where the PROMs have been used in trials or observational studies and performance indicators are reported incidentally. The Consensus-based Standards for the selection of health Measurement Instruments (COSMIN) checklist will be used to appraise the methodological quality of those development papers that specifically evaluate instrument properties. [19] Criteria for assessing the psychometric performance of candidate instruments include: reliability, validity, responsiveness, precision, interpretability, acceptability and feasibility. [20] We will use these and criteria proposed with explicit criteria for rating the level of evidence to support quality of PROMs as either positive, indeterminate, negative, or no information. [21] These analyses will be conducted for each of the four exemplar conditions, and also aggregated for all neurodisability. The criterion of appropriateness, [20] in this instance for children with neurodisability, is addressed in the other work streams. However the review will record in what age groups each PROM has been administered, and with which other diagnoses. Standardised data extraction forms will be modified from previous work and piloted. Data extraction will be undertaken largely by the research fellow; quality checks by a second reviewer will be undertaken on 10% of the appraised papers. Inconsistencies will be noted and resolved by discussion. The number of inconsistencies will guide decisions about the need for further duplicate extraction. A PRISMA-style flowchart will be produced detailing the study selection process and the reason for exclusion of each full-text paper assessed. The review will produce a report of the evidence to support the use each candidate generic PROM to measure health outcomes of children with neurodisability.

**Stream 2: Qualitative research with families** (CM, AA, BB, KA, VS)
Stream 2 involves qualitative research with children with neurodisability and parents using a mix of focus groups and individual interviews to identify their perspectives on important healthcare outcomes, and the extent to which candidate generic PROMs represent these health outcomes. *Recruitment & sampling*: The Council for Disabled Children (CDC) is the leading national policy and practice improvement organisation for disabled children. CDC maintains a Making Ourselves Heard network which provides an expedient sampling frame for this aspect of the work. The network comprises of 271 contacts across England including major providers of services to disabled children from within the voluntary sector such as Mencap, Action for Children, Barnardo's and RNID and local authority leads from within both youth services and disabled children's teams. This provides access to children with a range of ages and conditions. CDC also works closely with the National Network of Parent Carer Forums which is the umbrella body for all 152 local parent forums and thus again provides access to parents, across the country, with children of all ages and spanning the spectrum of neurodisability, willing to provide their views on topics of importance to disabled children. We propose running 14 focus groups, 8 with children and young people, and 6 with parents. In addition 15 individual interviews will enable the inclusion in the project of children and parents who may not be able to participate fully in a group. Child and parent groups will be assembled in consultation with the CDC Making Ourselves Heard Network and National Network of Parent Carer Forums. The groups will be purposively assembled to have a common sense of identity. Groups will comprise children, or parents of children, with similar functioning or diagnoses. There will be groups for children, aged 8 to 13 years (although they may not be cognitively functioning at the level of age-equivalent peers), and adolescents or parents of adolescents aged from 13 to 18 years as there are often specific issues for this age group. There is considerable healthcare input for young children around the time of starting school; therefore we will also convene a separate group of parents of very young children, aged around 3 to 7 years, who would not be able to self-report their own health outcomes. For qualitative studies, sampling is ideally judged to be complete when theoretical saturation has been reached. [22] In practice it can be difficult to demonstrate this point and it cannot be precisely specified in advance as it depends on the variability in the sample and the properties of the data. However we anticipate that our sample will balance the need to: (i) include young people with a range of neurodisabilities, such as communication difficulties, learning disability and physical disability; (ii) where possible, include families from different socioeconomic and ethnic backgrounds, and children of a range of ages; (iii) allow us to conduct a rigorous data analysis which is at the same time achievable within a feasible and reasonable timeframe and budget. Our sampling strategy will be periodically reviewed in the light of the ongoing data analysis and modified if necessary to develop and test our emerging findings.

*Data collection & analysis*: There are three components to the qualitative research. First, we want to identify, broadly, what outcomes children and parents expect from the combined resources of the NHS; second we will present lay interpretations of the constructs assessed by the candidate generic PROMs, with example items, to determine whether these instruments measure outcomes children and parents value. Third, we will consider pragmatic approaches which might motivate children to want, and be able, to complete PROM questionnaires, such as novel technology. Data collection with children and young people will employ a suite of tools to ensure the

widest possible participation and understanding. Preparatory pilot work will be undertaken with members of the Cerebra Research Unit Family Faculty to explore different ways of raising issues in a way that is meaningful and the questions which the focus group or interview will explore [see sections on service user/public involvement and research stream 1]. At the beginning of each session, introductory work will orientate the group members to the topic, establishing a level of understanding and support needs. During the focus groups flash cards using pictures and visual clues will be used to help generate discussion. The researchers will also design activity based exercises to help children and young people explore the topic in ways which are meaningful to them. These methods are regularly employed by the CDC in their research. The lead applicant has successfully used life-mapping and story-telling as a means for children and parents to project their issues onto an imaginary character to de-personalise the issues being discussed. [23] Discussions will be audio-recorded and transcribed. Paper based exercises will be transcribed and activity based exercises will be observed and written up. For interviews with children or young people with severe communication impairments bespoke approaches using the child's own communication system will be identified with their carer. For example, interviews might be observed by an interview assistant taking notes, or a commentary might be recorded if there may be little or no spoken content. The qualitative component addresses focused policy relevant questions and is not seeking to elicit a deep understanding of lived experience. The Framework Approach [24] was developed as a systematic and rigorous methodology for such applied qualitative research and will be used for analysis. In the Framework Approach data collection is more structured and analysis is more explicit and more transparent than some approaches to qualitative data analysis. [25] The analysis in this project is therefore informed by the three components set out above. Framework analysis involves five distinct stages: (i) familiarisation with the data immersion in the raw data (listening to recordings and reading transcripts) to gain an overview of the whole; (ii) identifying a thematic framework – identifying the key concepts and issues both a priori and those emerging from the data of individual respondents and recurring concepts; (iii) indexing – applying the framework to the transcripts, annotating the transcripts with identification codes referring to themes and subthemes; (iv) charting –extracting data from its original context, summarising and grouping it in chart form according to the thematic reference (v) mapping and interpretation – reviewing the charts and research notes to compare and contrast, search for patterns and connections and provide explanations for the findings. Some issues will emerge as more salient than others and the interpretation of findings is influenced by the original research objectives as well as the themes emerging directly from the data. AA will lead on the analysis; all members of the research team for this stream will be involved in the analysis. AA will read all transcripts, and all transcripts will be read by more than one member of the research team. All members of the research team will be involved in developing the thematic framework. Initially the analysis will be exploratory in nature, but then become more confirmatory as we interpret findings from health professionals (see Stream 3), with particular attention to accounting for deviant cases. AA will index and chart the material and 25% of material will also be indexed and charted by VS. This allows for checks to be made for comprehensiveness of data extraction, and consistency in the application of the index. The charts will be read by the team. During the mapping and interpretation stage, all members of the research team will discuss the developing analysis. Differences in interpretation will be resolved through

group discussion. A detailed record will be kept of the analysis process, including definitions of the themes and concepts and their application.

**Stream 3: Professional Reference group & Delphi survey** (CM, JW, RT, SL)
Stream 3 involves seeking the views of a multidisciplinary group of NHS professionals about appropriate health outcomes and PROMs; the method will be an online Delphi survey. [26] The work will be coordinated from Exeter by the research fellow and lead applicant (who is also an allied health professional) and three Consultant Paediatricians.
*Recruitment & sampling*: The British Academy of Childhood Disability (BACD) is a multidisciplinary organisation and subgroup of the Royal College of Paediatrics & Child Health (RCPCH). BACD Strategic Research Group (SRG) supports high quality childhood disability research and, following discussion at a meeting in March 2011, is enthusiastic to support this proposal. BACD SRG members recently completed a national survey of child development teams (CDTs) throughout the UK (94% response) and also a survey of the neurodisability lead professionals across the country. The resulting database is securely kept at the RCPCH. BACD intends the database to be a resource which allows clinicians to undertake research, audit and activities which help us bring about improvements in the interests of children with neurodisability, their families and clinicians. It is updated periodically (last updated 2010). Using this list of contacts will enable us to sample the opinions of a wide range of NHS professionals working with disabled children. On our behalf, BACD will email a sample of leads on the list to provide broad geographical coverage and mix of centres, further sampling will be possible depending on the response. The neurodisability leads of the CDTs will be asked to forward the invitation to take part to their colleagues across a wide range of professionals who are working with children with neurodisability. Interested professionals who wish to take part will be asked to provide details of their profession and email address to the research team in Exeter. This group will form the baseline population for the Delphi survey. Sampling will be purposive; if necessary we will seek professions who might not be represented by contacting the relevant special interest groups of other societies.

*Data collection & analysis*: The online Delphi survey (using SurveyMonkeyTM) will invite the professionals to rate their agreement with several statements based on the findings emerging from the literature review and qualitative research with families (Streams 1 & 2). We envisage 3-5 rounds of the survey over the course of the study following recommended guidelines. [27] The surveys will be staged to happen after the exploratory qualitative stage with families, and then again after the confirmatory stage. Similar to the work with families, the surveys will seek agreement, broadly, on what the combined resources of the NHS are seeking to achieve for children with neurodisability, and whether the constructs assessed by candidate generic PROMs are perceived to be appropriate. The statements presented in the Delphi survey will be constructed by the team in collaboration with members of the BACD SRG and representatives of other stakeholders to ensure the statements are considered meaningful. For each statement a five point scale will enable respondents to rate their level of agreement (strongly disagree-disagree-no opinion-agree-strongly agree). We will use a criteria of 67% approval (agree or strongly agree) among participating professionals for each statement; if any statement receives lower agreement then it will be revised and represented in a subsequent round.

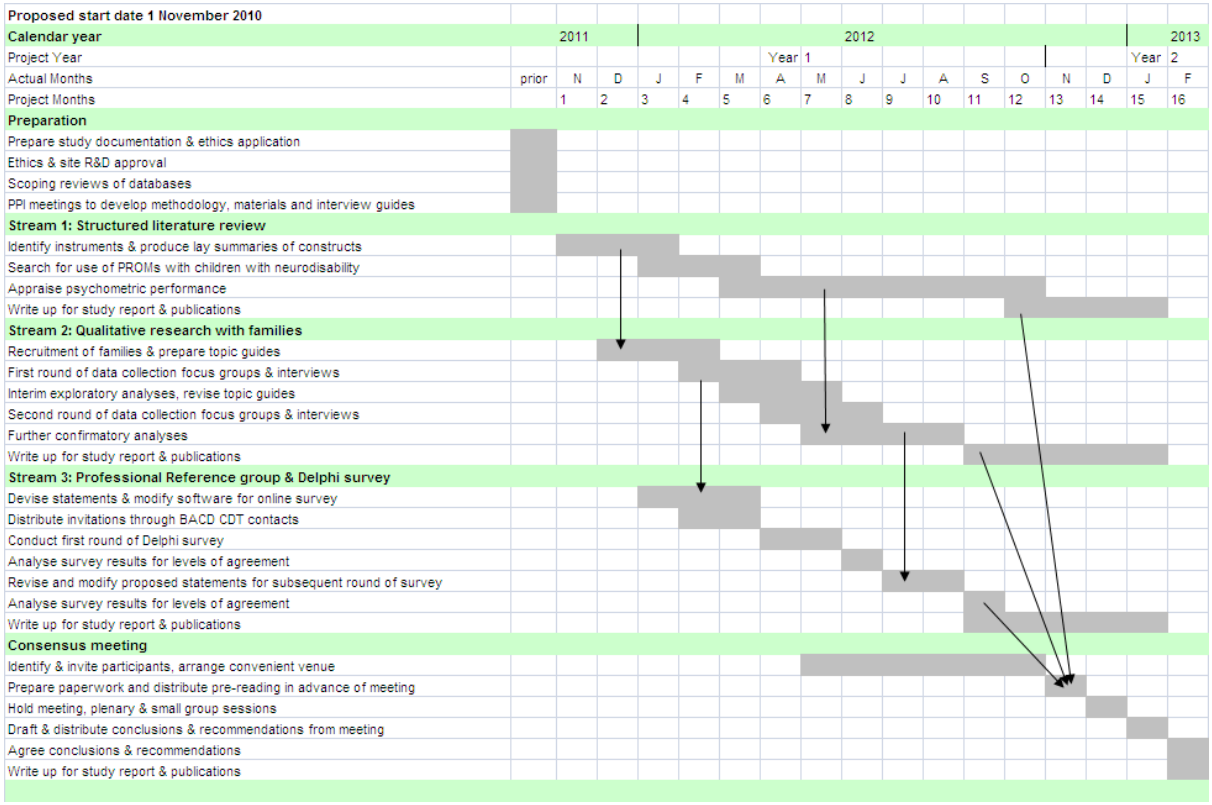
**Consensus meeting** (All)
The final stage of the research is a one-day consensus meeting where the findings of the three streams of research will be presented to, and considered by, representatives of the stakeholder groups. The meeting will seek to converge on a common view on the types of outcomes of NHS care that should be assessed, and make recommendations about the best available PROMs to measure these outcomes. All the research team will contribute to planning the meeting as decisions will be made about what information is presented. The consensus meeting will involve around 20 participants at a location that provides the easiest public transport to minimise travel costs. Participants will include two young people, from among the Young Ambassadors of the Council for Disabled Children, parent representatives, and health professionals. A summary of progress of the study will be sent in advance of the meeting with a structured agenda so that participants are acquainted with the purpose of the meeting and status of the project before they arrive. The meeting will begin with a plenary presentation of the findings of the study. Participants will then break into small groups to discuss key points identified in advance. The meeting will reconvene in a plenary session to discuss and integrate the small group recommendations. The meeting will finish with discussion of the ways in which the findings can be reported to a wide range of audiences. All sessions will be audio-recorded. Following the meeting the recommendations will be sent to a nominal group of the participants for comment and then circulated to all the participants for final agreement. Potential outcomes range from identification of one of more PROMs that measure a commonly held view of appropriate NHS outcomes, to families and professionals holding such disparate views that a common consensus on what NHS outcomes are important cannot be reached. The findings, whatever they may be, will be a valuable basis to inform further applied research and policymaking.

**CONTRIBUTION TO COLLECTIVE RESEARCH EFFORT AND RESEARCH UTILISATION**
*Policy-relevant output*: The proposed research has the potential to significantly inform NHS policy and practice by bringing together health professionals and families to identify a common vision of what outcomes of NHS care should be measured for a large group of intensive users of the NHS. The output also addresses a key recommendation of the Kennedy Report on overcoming barriers to improving provision of services by the NHS. To achieve these impacts the findings will be presented to the Department of Health, through PenCLAHRC and the NHS Outcome Framework programme. In addition, we will engage with NHS commissioners for childhood disability services early in the process, once the new commissioning arrangements for this group of children become clearer.

*Academic output*: A number of scientific outputs are expected from the work that will be presented at national and international conferences and written up for submission to peer-reviewed publications. The appropriate academic forums for presenting the work span health services research, childhood disability and measurement audiences. The structured review in Stream 1 is a stand alone output; the work in Streams 2 & 3 and the consensus meeting could be published as a single major output. There are also the methodological challenges of conducting the process, and especially with disabled children. Hence there is considerable scope for involving families in the design of the

study; reflection of this aspect of the work will contribute knowledge about public and patient involvement (PPI).

## PLAN OF INVESTIGATION AND TIMETABLE




## APPROVAL BY ETHICS COMMITTEES

Aside from the literature review, this study requires an ethics committee to approve the qualitative and Delphi survey work with families and professionals in Streams 2 & 3, and for the consensus meeting. However we do not envisage any major obstacles to gaining ethics approval providing our proposed arrangements for gaining informed consent, protection of vulnerable participants (children) and anonymity of contributions are assured. Given the national coverage of study participants we will seek approval through the National Research Ethics Service. Seed funding through the Peninsula CLAHRC is enabling the team to conduct some preparatory and preliminary piloting of the methods. The preparatory work will include submitting an application for ethics approval, which we presume will be approved in advance of the decision on this application. Thus, given a positive opinion on funding from the NIHR HSR Programme, we will be able to proceed with the study in accordance with the proposed timeline.

## SERVICE USERS/PUBLIC INVOLVEMENT

The Cerebra Research Unit at Peninsula Medical School involves families of disabled children in all their activities through a Family Faculty. The proposed research was strongly endorsed by parents at Advisory Group meetings of the Cerebra Research Unit. We have also developed the proposed work in conjunction with the Council for Disabled Children (CDC), part of the National Children's Bureau. Both groups are represented as co-applicants. We are able to draw on the combined resources of the groups to ensure that all parts of the work that involve children and parents are well

planned in terms of feasibility, managing expectations and to ensure that information is pitched at an appropriate level. We are beginning preparatory piloting work (supported by PenCLAHRC) in advance of the decision on this application. This piloting is collaborative work with disabled children and parents to find ways to explain what PROMs are and to describe the constructs assessed by relevant measures. This piloting, with meaningful involvement of families, adds to the expediency with which the work can be delivered.

**PLANNED OR ACTIVE RELATED RESEARCH GRANTS**

The lead applicant manages the Cerebra Research Unit at Peninsula Medical School; this is an academic childhood disability research unit that receives core funding from the charity *Cerebra* to support involving families in developing research questions. NIHR funded PenCLAHRC, under the direction of Professor Logan, is supporting preparatory work so that the proposed work streams can begin expediently should the application be approved; these activities include preparing documentation, ethics approval, piloting etc.

**HISTORY OF PAST OR EXISTING NIHR PROGRAMME RESEARCH**

The lead applicant has never held an NIHR programme contract which has been terminated, extended in time or in terms of funding. Five of the academic applicants from the Peninsula Medical School are under the NIHR funded PenCLAHRC umbrella, directed by Professor Logan. Professors Jenkinson, Tennant and Logan, and Dr Green are, and have been, involved in various NIHR funded studies.

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