EMPoWER: Early Mobility and POwered Wheelchair Evidence Review

Full Protocol

Full title of project:

Cost-effectiveness of earlier provision of powered mobility interventions for children with mobility limitations: a systematic review

Chief Investigator:

Nathan Bray[1]

Co-investigators:

Fiona Beyer[2]; Dawn Craig[2]; Aimee Grayston[3]; Rhiannon Tudor Edwards[1]; Jennifer McAnuff[2]; Jane Noyes[4]; Louise Tanner[2]; Lorna Tuersley[1]; Niina Kolehmainen[2,5]

Affiliations:

[1]Centre for Health Economics and Medicines Evaluation, Bangor University

[2]Institute of Health and Society, Newcastle University

[3]NHS Leeds Community Healthcare Children's Services

[4]School of Social Sciences, Bangor University

[5]Newcastle upon Tyne NHS Hospitals Trust

Funding: EMPoWER is an NIHR HTA funded evidence synthesis project (NETSCC ID: 17/70/01).

Contact details:

Nathan Bray

Centre for Health Economics and Medicines Evaluation

Ardudwy, Bangor University, Gwynedd, LL57 2PZ

Tel: +447792670053 / Email: n.bray@bangor.ac.uk

BACKGROUND AND RATIONALE

Non-communicable illness and disability explains most of health expenditure in Western countries. The UK has an estimated 952,700 disabled children (0-18 years), with mobility limitations among their most common difficulties (Department of Health, 2013). The impact of health services for children with mobility limitations is currently significantly hindered by lack of effective interventions (Novak et al, 2013; Scianni et al, 2009; McPherson et al, 2014; Tatla et al, 2013; Barton et al, 2015) and the patients, policymakers, and healthcare providers all agree that improving interventions is an urgent priority (Morris et al, 2014; 2015).

One of the most promising, emerging interventions is the early provision of powered mobility for young children (<5 years) with mobility limitations to promote self-directed mobility. Early powered mobility interventions aim to enable self-directed movement in children who are otherwise unable to do so, and through that to trigger positive developmental changes similar to crawling (Uchiyama et al, 2008; Deitz et al, 2002; Livingstone and Field, 2014; 2015; Jones et al 2003). The idea of early powered mobility as an intervention is based on the observation that acquisition of self-directed mobility (e.g. through rolling, crawling, bottom-shuffling) results in a major step-change in children’s engagement with the world, and through that in their perceptual, cognitive, social and physical development. Children with mobility limitations have less self-directed movement than their typically developing peers, with fewer opportunities for exploration of the world around them (Deitz et al, 2002; Jones et al, 2003; Jones et al, 2012; Tefft et al, 1999; Tefft et al, 2011). This places the children at greater risk of secondary disabilities (Tefft et al, 1999; Huang et al, 2014) in terms of life skills (motor, cognitive, social abilities related to daily tasks), general independence and autonomy, and participation in daily life across home, education and leisure. These secondary disabilities are, in turn, negatively related to the child’s long-term health, development and social integration, as well as having a negative impact on parental physical and mental health, parental productivity, and wider society. It is thought that early powered mobility could prevent these secondary disabilities by enabling the self-directed mobility and exploration.

Provision of mobility equipment to children is not new. For example, the UK health services provides mobility equipment for around 70,000 children (NHS Improving Quality, 2014). However, the current provision of powered mobility interventions commonly focuses on children over 5 years, with the assumption that children under the age of 5 do not benefit from it—at least not sufficiently for it to be cost-effective. However, if the hypotheses about the role of early self-directed mobility in prevention of secondary problems is true, then the current provision may be a missed opportunity to yield the best returns for public resource across the life-course (Edwards et al, 2016). Research has already shown that provision of early powered mobility is feasible and acceptable (Tefft et al, 1999; Huang et al, 2014; Jones et al, 2012), including children as young as 7-11 months (Lynch et al, 2009; Ragonesi and Galloway, 2012). The key remaining question is whether the early provision is incrementally more effective and cost-effective than later provision (i.e. from age 5 onwards).

There is currently no evidence-based national guidance, or other up-to-date evidence summary, for decision makers on what is the optimal time point for powered mobility provision for children, how different intervention components relate to cost-effectiveness, how variability in children's conditions and diagnoses impact effectiveness, and what outcomes could be used to monitor benefits of provision. In the absence of such guidance or summary, powered mobility provision is highly variable across services and regions, exacerbating the socioeconomic, health and well-being inequalities already experienced by children with mobility limitations and by their families.

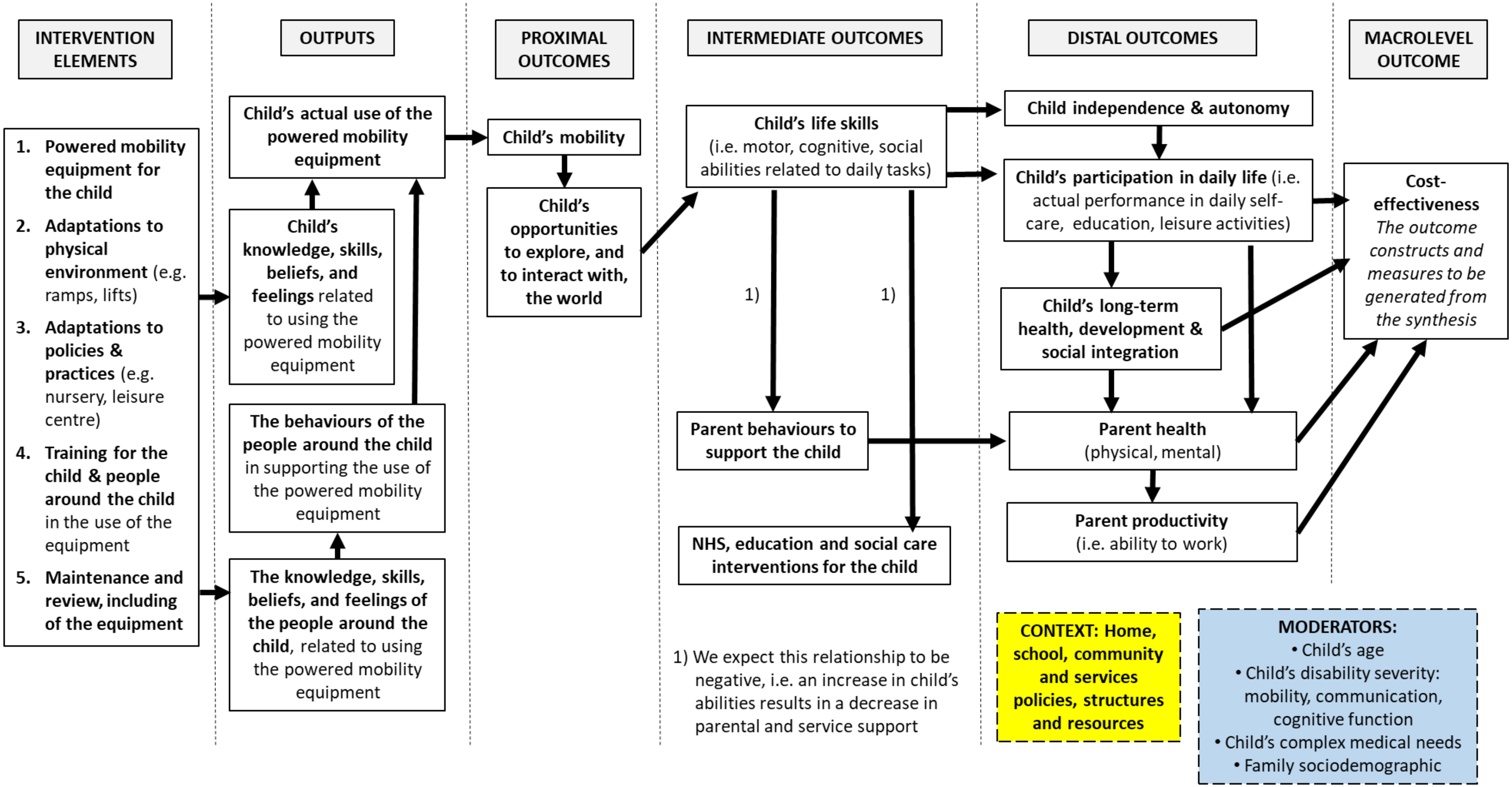
This research will synthesise existing evidence about the relative benefits and cost-effectiveness of early powered mobility (<5 years) compared to later powered mobility (≥5 years), and model the potential longer-term costs and benefits of early powered mobility. The results will provide health commissioners, wheelchair services, policymakers, and families access to timely evidence to facilitate informed decisions about how best to use resources to support disabled children and to promote their long-term health and well-being.

Logic model to underpin this research

From scoping of the literature and from expertise within our team, including the wider project advisory group (see Project Management and Patient and Public Involvement), we have developed an upfront logic model (following published guidance and examples: Anderson et al, 2011; Kneale et al, 2015) that provides a simplified representation of the proposed key outcomes and related change processes for powered mobility interventions (Figure 1). These proposals are outlined below, and will be investigated through this research.

The literature and expert opinion proposes that provision of powered mobility interventions for young children will reduce the negative effects of mobility limitations by enabling self-directed mobility, which provides opportunities for exploration and the development of the child's life skills (Jones et al, 2003; Logan et al, 2015; Huang et al, 2014, Jones et al, 2003), independence (Bottos et al, 2001, Jones et al, 2003), autonomy, and participation in daily life (Furumasu et al, 2008; Livingstone and Field, 2015). A further proposal is that these will collectively enhance the child's long-term health, development and social integration (Logan et al, 2015, Furumasu et al, 2008; Livingstone and Field, 2015), and that subsequent developmental gains will reduce the child's need for parental support (Jones et al, 2012) and related parental stress (Tefft et al 2011). Child and parent gains may together reduce the burden on public services across health, education and social care.

Fig1. Initial logic model to be used as framework for the evidence synthesis of the cost-effectiveness of powered mobility for children with mobility limitations



RESEARCH QUESTION, AIMS AND OBJECTIVES

Research question: *"Is the earlier provision of powered mobility to very young children more cost-effective than currently more common provision to children aged 5 and over?"*

Our aim is to examine and model the relative effectiveness and cost-effectiveness of powered mobility interventions for young children (<5 years) with mobility limitations compared to the more common practice of powered mobility provision for children aged ≥5 years. We will meet the aim through the following objectives:

1. To identify and synthesise quantitative, qualitative and mixed-method evidence to determine:
   1.1. The effectiveness and cost-effectiveness of powered mobility interventions for children with mobility limitations, and the wider impacts to health services and society.
   1.2. The acceptability, feasibility and anticipated outcomes of relevant interventions from multi-stakeholder perspectives (children, parents, service providers, commissioners etc.).
   1.3. The long-term implications of self-directed or independent mobility for very young children (<5 years) compared to older children (≥5 years).
2. To examine the economic costs and benefits of powered mobility interventions for children by:
   2.1. Building tariffs of NHS and non-NHS costs for powered mobility interventions (equipment, training and support, and any other components) for children with mobility limitations using a multi-perspective disaggregated cost-consequence framework.
   2.2. Modelling the relative cost-effectiveness of powered mobility equipment for very young children (<5 years) compared to standard NHS practice (≥5 years).

The key outputs will be an overview of the current evidence of powered mobility interventions for children, a summative logic model of effectiveness and cost-effectiveness for planning and evaluating future interventions, and recommendations for powered mobility provision and for future research. EMPoWER is an NIHR HTA funded evidence synthesis project (NETSCC ID: 17/70/01).

METHODS

Design

We will undertake a mixed-method evidence synthesis (Grant and Booth, 2009) (quantitative, qualitative, mixed-method and economic evidence), followed by economic modelling that incorporates those evidence and other existing economic data from NHS and third sector organisations. The evidence synthesis will follow the basic principles for conducting effectiveness and cost-effectiveness reviews (see Centre for Reviews and Dissemination (CRD) guidance (CRD, 2009)), with each stage of the review using additional up-to-date relevant methods and tools, with an explicit protocol registered with PROSPERO. We anticipate that a Markov model will be used to model cost-effectiveness, using a range of data sources to define model parameters. The initial programme theory in the logic model (above, Figure 1) will be used as a conceptual starting point and will be cumulatively developed throughout the review processes to produce a definitive logic model to inform the economic modelling.

Setting/context

The immediate provider setting is predominantly, but not exclusively, allied health and wheelchair services which provide mobility interventions for children, and any third sector providers that these services work with for powered mobility provision. The wider setting includes family contexts and nursery, education, communities, and social care. We will include international evidence from health services which are broadly delivered in a similar high income context to the UK NHS as well as evidence specific to the UK National Health Service (NHS).

Target population

The target population will be children with mobility limitations, described in terms of the child's ability to move around. We anticipate that studies will use standardised classification systems (e.g. the Gross Motor Function Classification System), codes from the WHO International Classification of Health, Functioning, and Disability, or clinical observations. Medical diagnosis (e.g. cerebral palsy, developmental delay) will be extracted but not used as inclusion criterion. This use of ability-based rather than diagnostic criteria reflects the service provision and realities of these children, and supports external validity and relevance to NHS provision (Stein et al, 1982; Varni et al, 1999). For the purpose of comparison, the population will be stratified by age (<5 years and ≥5 years).

Health technologies being assessed

Powered mobility interventions are complex interventions with several elements and synergistic outcomes and benefits. Powered mobility use takes place in, and is influenced by, the child's physical and social environment (Livingstone and Field, 2015), and the exact features and delivery of the intervention elements varies depending on the child's age and/or developmental stage and impairments. Provision of powered mobility often involves a combination of allied health (e.g. physiotherapy, occupational therapy) and wheelchair services that come together for the provision of multifaceted intervention packages.

For the purpose of this review, we will consider early powered mobility to consist of five elements:

1. The powered mobility equipment: for example powered wheelchair, ride-on device or toy car, to enable the child to move around, including any related method of control (e.g. switches)
2. Adaptations to physical environment: for example ramps and lifts, to enable the physical use of the equipment
3. Adaptations to policies and practices: for example safety rules and activity processes, to provide a socially facilitative and safe environment for the use of the equipment
4. Training and other behaviour change techniques: for example goal setting and self-monitoring, provided to the child and the people around the child to maximise the likelihood that the equipment is used regularly and appropriately
5. Maintenance and reviewing: for example maintenance of the equipment and related adaptations and use, in the longer term.

There is currently no agreed, standardised descriptions for any of these five elements. Based on our previous reviews of childhood disability interventions, we anticipate the literature to include a range of interventions, described to variable degrees of specificity. To allow meaningful comparisons and a robust economic model, we will extract the intervention details using the Template for Intervention Description and Replication (TIDieR) (Hoffmann et al, 2014), using a process we have previously applied in a systematic review of childhood disability interventions (Kolehmainen et al, 2018).

For the present review and synthesis, the active intervention is provision of powered mobility for children under 5 years, and the comparator is provision for children 5 years and over. To enable this comparison, the data will be extracted separately for the two groups where possible.

Evidence synthesis

Search strategy

The search strategy will be designed and run in collaboration with an experienced information specialist. We will search the following bibliographic databases: CINAHL, MEDLINE, PubMed, Pedro, OTSeeker, ASSIA, PsycINFO, Science Citation Index, Social Science Citation Index, Conference Proceedings Citation Index (Science and Social Science), Cochrane CENTRAL, CDSR, DARE, NHS EED, and HTA. The searches will incorporate both thesaurus-controlled subject heading terms and text words or phrases. Sensitivity will be prioritised over specificity.

The search will combine terms describing children with terms about mobility. The searches will not be restricted by outcome, disability, activity limitation or medical diagnosis (these will be incorporated at screening, see below); or by study design, language, or publication year.

To reduce publication bias, the electronic searches will be supplemented by: reference-list searching; hand-searching a sample of selected, highly relevant journals; searching any highly relevant conference proceedings that are not indexed on the databases above; and identification of relevant grey literature. Grey literature will include OpenGrey along with resources such as government policy documents, third sector organisation reports and unpublished research—much of which will be sourced from within our project advisory group that includes expert individuals, research groups, and organisations who have agreed to support this research and have knowledge of unpublished data. The project advisory group will also direct us to seminal papers, particularly related to evidence for the long-term implications of independent mobility.

Three search facets will be developed (‘children’, ‘powered mobility’, ‘mobility’) and used to identify relevant evidence. Each search facet will consist of relevant subject headings and text words. An experienced information specialist (co-investigator, Fiona Beyer) will develop, refine and apply the search strategy. For an illustrative example of keywords and search strategy, please see Appendix 1.

Screening for inclusion

Titles and abstracts will be screened for inclusion by two independent reviewers, of which at least one will be a topic expert. Papers will be included if: (i) the study **P**articipants are children with mobility limitations, defined using explicit criteria (see below); AND (ii) the **I**ntervention described in the paper involves at least one of the five intervention elements described above; AND (iii) the **O**utcome (or, the ‘phenomenon of interest’) is related to the child, their family, health or social care, or education. Within this, ‘mobility limitations’ will be defined as impairments in the functions of movement and mobility, including: functions of joints, bones, reflexes and muscles; control of voluntary and involuntary movements; gait, muscle tone and power, and joint mobility. Conditions where movement problems are part of the diagnostic criteria or condition definition (e.g. cerebral palsy, muscular dystrophy) will be considered to meet the population criterion; judgements about this will be made using explicit rules developed in a previous intervention review in this population (Kolehmainen et al, 2018) (Table 1). The project advisory group will provide assistance in refining the inclusion and exclusion criteria as needed.

Table 1. Decision rules that will be used to make judgements about population inclusion

Diagnoses and condition groups	**Decision**	**Rationale**
▪ Cerebral palsy ▪ Developmental coordination disorder, dyspraxia, other similar sensory-motor disorders ▪ Brain injury (traumatic, acquired) ▪ Spinal cord injury ▪ Hypermobility syndrome ▪ Muscular dystrophy	Always include in the review	Movement problems are part of the diagnostic criteria or condition definition.
▪ Global developmental delay, intellectual disability ▪ Rare developmental syndromes ▪ Attention disorders ▪ Autistic spectrum disorders ▪ Pre-term birth, low-weight birth ▪ Chronic disorders in movement-related body structures (e.g. juvenile idiopathic arthritis)	Included if movement problems explicitly mentioned as part of the eligibility criteria or rationale	While the diagnostic criteria do not necessitate movement problems, co-morbidity is common.
▪ Obesity, respiratory conditions, cancer, enuresis, diabetes, colic, cardiac problems, burns, HIV, chronic fatigue syndrome / myalgic encephalomyelitis ▪ Short-term impairments in body structures (e.g. fractures)	Excluded from the review	Movement problems are not inherently part of the diagnostic criteria or condition definition, or are short-term.

Papers will be excluded if it is not possible to identify outcomes (either measured or described qualitatively); OR the intervention is solely non-powered mobility (e.g. manual wheelchair); OR the paper is not based on empirical data (e.g. opinion pieces).

All studies judged as relevant based on title and abstract will be retained and obtained as full texts. Any discrepancies in decisions about eligibility will be discussed until a consensus is reached, including a discussion among the wider team as necessary. Software packages EndNote and Rayyan (Ouzzani et al, 2016) will be used to manage literature and document the review process.

The project advisory group, and in particular the parent advisors and expert clinicians, will play a key role in finalising and implementing the inclusion and exclusion criteria; for example, in meaningfully defining 'mobility limitations' in very young children, interpreting papers where mobility limitations are not explicitly reported, and checking whether the reasons for excluding papers, particularly on the basis of population or outcome, are understandable and acceptable.

Quality appraisal

Assessment of study methodological strengths and limitations will focus on risk of bias in randomised controlled trials and non-randomised studies; risk of confounding in non-randomised studies; and risks to rigour in qualitative studies. The assessments will be used to feed into the development and interpretation of results and may be used to inform sensitivity and subgroup analyses, but will not be used to exclude studies. The assessment will consist of two stages.

First, all included studies will be classified on the basis of their specific design features, e.g. method for assigning intervention, as opposed to primary authors' reported design labels, which are often inconsistently applied (Reeves et al, 2011; CRD, 2009). This will support interpretation of results by highlighting particular risks associated with specific design features. The classification will be conducted by one reviewer, with arbitration from another as required, using a published algorithm adapted for the purposes of the proposed synthesis (Seo et al, 2016; Hartling et al, 2010; Viswanathan et al, 2013).

Second, the studies will be assessed for methodological strengths and limitations. We will select the most appropriate tool(s) based on the papers included. As guiding principles, we will use the established Cochrane Collaboration's tool for assessing risk of bias in randomised controlled trials (Higgins and Altman, 2011), and the ROBINS-I (Risk Of Bias In Non-randomised studies – of Interventions) (Sterne et al, 2016) for non-randomised studies. For qualitative studies we will follow Cochrane Qualitative and Implementation Methods Group Guidance and use the Critical Appraisal

Skills Programme (CASP; http://www.casp-uk.net/casp-tools-checklists) qualitative tool. For mixed-method studies we will use the Mixed Methods Appraisal Tool (MMAT) (Pluye et al, 2011). Grey literature studies will be assessed using the appropriate method-specific tool. Two reviewers will pilot the quality assessment procedures and independently assess each included study, with a third reviewer arbitrating disagreements as required to reach consensus and with input from a topic expert. The software(s) to be used will depend on the tools chosen.

After the analysis and synthesis, assessments of primary studies obtained from stages one and two will subsequently feed into the Grading of Recommendations, Assessment, Development and Evaluation (GRADE) approach to assess how much confidence can be placed in the results of the effects of interventions (Guyatt et al, 2008). For synthesised qualitative findings, the Confidence in the Evidence from Reviews of Qualitative research (CERQual) will be used (Lewin et al, 2015). This incorporates four components: methodological strengths and limitations of studies contributing to the finding; relevance of evidence to the review question; coherence of findings; and adequacy of data. A quantitative and qualitative summary of findings table will be produced to display GRADE assessments.

Data extraction

A bespoke data extraction form will be developed based on a previous similar review in this population (Kolehmainen et al, 2018); piloted on a small number of included studies; and revised as necessary. Numeric and textual data will be extracted on: the first author; year of publication; the participants; country of data collection; the intervention label(s) as provided by the authors; the intervention characteristics (using the TIDieR (Hoffmann et al, 2014)); outcome(s) and benefits targeted or reported by participants (including long-term implications), and any related measure(s); hypothesised change processes (‘mechanism’, ‘process outcomes’) and any related measure(s); evidence about feasibility and acceptability; and costs and any related measure(s).

For outcomes, benefits, interventions and change mechanisms both numeric and textual data will be extracted, and the source of data (e.g. objective measures, participant accounts, author narrative) recorded. Based on our previous reviews, we anticipate that of the data to be extracted the following will require substantial discussion and interpretation due to limitations in reporting: the outcomes and related measures, intervention characteristics, and hypothesised change processes. For these, explicit decision rules similar to previous reviews will be used to ensure consistency and transparency.

To extract the data, two independent reviewers (at least one of whom will be a topic expert) will: read the title, abstract, introduction and methods of the included full-texts one at a time; identify the relevant excerpts of text (ranging from a single number and word to strings of numbers and paragraphs) containing information about any of the data aspects above; and will copy, verbatim, the extracts from the full-texts on to a data file. The data extraction will be carried out in batches where the reviewers will independently extract data for 2-5 papers and then come together to check consistency.

The data extraction will result in two streams of data: the numeric (quantitative) and the textual (qualitative). Across these, we anticipate the data to include evidence from different study designs on effectiveness and perceived benefits; cost-effectiveness and perceived costs; and feasibility and acceptability of provision and use.

Our Patient and Public Involvement (PPI) co-investigator, Ms Aimee Grayston, will contribute to each of the stages above, for example by generating search terms, observing the application of the search strategy, refining inclusion/exclusion criteria, screening a proportion of titles and abstracts, jointly assessing the quality of 1-2 included papers, and jointly extracting data from 1-2 papers. This will ensure the wider research team capitalises on Ms Grayston's unique lived experience of powered mobility, and also provide a continuous check that the study makes sense in the context of her lived experience. By doing so we aim to create a habit of conveying the study in plain English, which we anticipate will strengthen the accessibility of our subsequent dissemination of study methods and findings (see Dissemination and Projected Outputs). In addition, our first 1-2 workshops with young powered mobility users (see Patient and Public Involvement) will focus on engaging the young people in the study aims, logic model, and methods, and beginning to explore creative and interesting ways of describing these to a wider audience, including decision makers.

Data analysis and synthesis

The exact analysis and synthesis techniques will depend on the data that can be extracted from the included studies. Based on the scoping searches, we anticipate using a results-based convergent approach where qualitative and quantitative data will be analysed and presented separately but integrated using further synthesis methods including the logic model, tables, and matrices. We will use an appropriate Office software for different steps according to the type of data.

Based on the scoping searches, we anticipate the numeric data to focus mainly on quantifying the effectiveness, perceived benefits, cost-effectiveness and costs; and the textual data on describing how stakeholders' conceptualise and experience outcomes, benefits and costs, as well as the interventions (including feasibility and acceptability). We will use the following key principles to guide the analysis and synthesis. The best ways to present the different results to be identified once we know the exact data available, and work closely with our PPI co-investigator and wider project advisory group to experiment with different visual, graphical, and textual methods for making the findings understandable, useable, and interesting for stakeholders.

Data about study characteristics, population, outcomes, interventions, and change mechanisms will be described using common classes (e.g. age, mobility, powered wheelchairs, parent training) derived from the included studies. Where possible, the aggregate data will be described as frequencies and illustrated e.g. in spider diagrams.

To map the preponderance of evidence, we will identify the data (qualitative and quantitative) which address the same outcomes or relationships. These may be outcomes and relationships that are already in the logic model or that extend it. We will analyse and pool different types of data as segregate streams, and subsequently match analytical approaches to the types of data, while also giving consideration to the purpose (e.g. effectiveness, cost-effectiveness, feasibility) for which the data were originally generated. Based on scoping, we anticipate that meta-analysis will not be possible. We will follow Cochrane methods to summarise numeric data in narrative summary format in a separate stream, which will then be presented graphically, e.g. in harvest plots (Ogilvie et al, 2008). The textual data, likely to consist mainly of brief quotations and original authors' narrative sentences, will be analysed and summarised using framework synthesis (Brunton et al, 2006). In this, evidence from the included studies is coded against the concepts in an *a priori* framework (i.e. the logic model, Figure 1) while new themes are also simultaneously generated through constant comparison across studies and against the framework. The new themes are then integrated in the framework and the framework further developed.

Overarching integrative synthesis

Synthesised evidence from all the respective within-data-type streams will be brought together in an overarching integrative synthesis. Data will be configured across the streams to identify complementary and contrasting findings within and across the studies. In this process, similar or relevant findings across data types and purposes will be interrogated and configured in relation to the developing logic model. Specifically, the focus will be on extending the coverage of the findings by: (i) complementing numeric data with textual data; and (ii) expanding the breadth and range of inquiry by drawing on one type of data to follow up and extend findings from another. For example, specific themes related to the feasibility of using a specific piece of powered mobility equipment can be used to explain and further interpret the corresponding numeric data that reports on the relative effectiveness of that piece of equipment. We will keep an open mind about conducting additional targeted searches for evidence to try and fill any obvious gaps or whether to use the advisory group and team expertise to come up with propositions and conjectures to explain the findings.

The logic model will be modified throughout the analysis to respond to the data and synthesised evidence, resulting in the final version that is the best fit of the initially hypothesised factors, modified factors, and new factors. We will attempt to identify theories to help populate the logic model, following Cochrane guidance (Noyes et al, 2015). The project advisory group will be instrumental in the continuous review and critique of the logic model, for example by challenging its assumptions and gaps, and enabling the research team to better understand and convey relationships between powered mobility interventions and outcomes. We will be mindful to differentiate between evidence, expert/lay opinion and proposition/conjecture when populating the logic model.

The final resulting model will illustrate the relationship between intervention effectiveness and cost-effectiveness and important modifying factors, including child age, and taking into account evidence from studies that have used external measurement as well as studies of stakeholder perspectives. Throughout, we will categorise data and findings to children <5 years and children ≥5 years for comparisons, and regularly review and critique the developing logic model and the data underpinning it.

Economic model

Model structure

If evidence of sufficient effectiveness can be identified from the evidence synthesis we will develop an economic model to estimate the incremental cost-effectiveness of early powered mobility compared to common NHS practice. We will use the synthesised evidence to define intervention scenarios (stratified by age) and undertake a number of primary cost-effectiveness analyses, using a range of outcome measures (identified from the systematic review). We will conduct a secondary cost-consequence analysis to set out the disaggregated costs and benefits.

There are two frequently used model types in decision analysis modelling: the decision tree and the Markov model (Siebert, 2003). The model structure proposed for this project is to be a Markov model (Sonnenberg & Beck, 1993) because it allows room for frequency and uncertainty in occurrences unlike the decision tree, which is useful in a “one-off” stage-by-stage analysis. Thus, the Markov model is more suited in the evaluation of fluctuating health states and chronic illness.

Our planned model will allow us to examine a hypothetical cohort of patients moving through defined Markov states and time periods. Markov modelling allows complex real-life events to be represented in a simplified health state form (Briggs & Sculpher, 1998). A Markov cohort/Monte Carlo simulation model, written in STATA (version to be determined) will be used to carry out the cost-effectiveness analyses. In a cohort simulation each cycle gives the analysis of the movement of patients collectively across the transition states. In a Monte Carlo simulation the movement of each individual across the states is summed and analysed, hence why the Monte Carlo simulation is called the individual simulation.

Parameters such as clinical outcomes, healthcare costs incurred, time duration/initiation of the intervention and wider societal costs/benefits will be included in the model construction. The modelled patients can remain in their current state, move to another health state or reach the 'absorbed' state according to certain transition probabilities (Siebert, 2003).

We will use child development literature to estimate the impact of independent mobility (initiated before/after the age of 5) on the attainment of development milestones. To examine the impact of delayed mobility across the life-course, we will use transitional probabilities (related to developmental attainment) and a range of outcomes to model progression over a range of time horizons (e.g. 0-10years; 0-18years; 0-death). Potential long-term savings in terms of health and social care expenditure will be factored into the model where possible. Sensitivity analyses will be used to account for uncertainty, for instance adjusting estimates of costs and/or outcomes to examine the effect on cost-effectiveness. Although we anticipate that a Markov model will be used to model cost-effectiveness and extrapolate end points, we are also open to other modelling approaches depending on the data.

To generate cost data we will liaise with various NHS wheelchair services, charitable organisations and our expert advisors to develop a tariff of costs for powered mobility equipment for children (stratified by age i.e. under 5 years/5 or over; and content of the intervention). We anticipate working with a range of NHS posture and mobility services across England and Wales, and will liaise with the NHS Wheelchair Managers Forum to engage with service managers. We have already engaged with the Rehabilitation Engineering and Aids to Living service at Newcastle upon Tyne Hospitals NHS Foundation Trust, and three third sector providers of early powered mobility: Whizz-Kidz (who also deliver NHS services), Queen Elizabeth's Foundation for Disabled People, and Designability, who have given preliminary approval for us to access their powered mobility intervention provision data. We will synthesise these data with existing published literature to establish estimated costs for the provision of different types of powered mobility equipment for children across the age range. We will again use the TiDIER checklist to classify the interventions.

The costing of the interventions will take into account a range of different factors, including base cost of powered mobility equipment, customisation/adaptation costs, staff/admin costs, maintenance costs and the recyclability of equipment. Furthermore, we will utilise published evidence identified in the evidence synthesis to estimate potential cost and benefits related to powered mobility interventions, for instance changes in the need for other services and treatments; physical environmental adaptation; training; and parent productivity. NHS England (2016) have published several reports about the recent Wheelchair Tariff Pilot, including information on staff time and repair/maintenance costs, which we will utilise to build the model.

Our PPI co-investigator and expert advisors will support the economic modelling by contributing expertise on potential benefits and costs of powered mobility interventions, and – importantly – ensuring the modelling methods can be conveyed in an accessible and engaging way to strategic decision makers and a lay audience. Our subsequent workshops with young powered mobility users will focus on exploring the study findings, building on the advisors' ideas for conveying the data synthesis and modelling methods, and storyboarding an animated film that will simplify and represent the study methods, and convey the key findings and recommendations.

Budget impact analysis

We will undertake a budget impact analysis (Sullivan et al, 2014) to demonstrate the potential scale of resources required to extend powered mobility provision to under 5s routinely. This analysis will be undertaken to compare the direct resource use costs and healthcare processes of the proposed earlier provision of powered mobility. The integrated cost estimate will be incorporated to compare earlier provision against the current cost of powered mobility from age 5 onwards. The totality of the budget implemented will be drawn from the cost estimate per person, estimated from published evidence and other expert sources. Hence, the budget impact analysis will be applied to produce better resource allocation guidance.

Value of information analysis

Our intention is to implement a value of information (VOI) analysis as part of the model outputs; an implicit next step to the probabilistic sensitivity analysis. A VOI analysis will help quantify the expected gain in net benefit from obtaining further information to inform a decision. Any decision based on existing information will be uncertain and, in time may turn out to be incorrect if more information becomes available. Should the wrong decision be made there will be a cost in terms of lost health benefit and wasted resources. Quantifying the value of an incorrect decision, alongside the probability of making an incorrect decision, will allow us to estimate the expected value of perfect information (EVPI). If the EVPI for a decision problem exceeds the cost of future research, additional investigation may be worthwhile.

As well as determining EVPI around the decision as a whole, VOI approaches can also be used for particular elements of the decision with the purpose of focussing research in areas where the elimination of uncertainty might have the most value. Expected value of partial perfect information

(EVPPI) analysis can be used to estimate the expected value of removing uncertainty surrounding specific parameters or groups of parameters to identify where future research should focus on identifying more precise and reliable estimates of specific pieces of information, e.g. relative effectiveness, costs or utilities. If the EVPI or EVPPI exceeds the expected costs of additional research, then it is potentially cost-effective to acquire more information by undertaking this research. This additional analysis can be very worthwhile, however to fully inform research the calculation of the expected net gain of sample information (ENGS) would be required. This can be a challenging and time-consuming process. Whilst we envisage that EVPI and EVPPI may be feasible, the final step of ENGS would only be undertaken if the model data are fully informed by evidence. If speculative, exploratory data are used to run alternative scenarios the VOI analysis can be clumsy due to extremely high levels of uncertainty. Therefore the full extent of the VOI analysis is likely to depend on the evidence base identified within this work.

DISSEMINATION AND PROJECTED OUTPUTS

This proposed research project has the potential for high impact and to inform commissioning of NHS paediatric wheelchair services and NHS provision of powered mobility equipment for children. A full report will be written for the funder, which will be published as a full and complete account of the research in the NIHR HTA Journal. A shorter report will be developed and disseminated to relevant commissioners, clinicians, practitioners and service managers, which will be designed to clearly and efficiently present the key findings relevant to decision makers. The short report will draw on recent evidence about how commissioners and managers use research (Swan et al, 2012; 2017; Edwards et al, 2013; Wilson et al, 2017), by incorporating case studies, examples, and direct quotes from expert advisors and key primary studies included in the evidence synthesis. We will aim to develop an evidence to decision framework (without offering recommendations) so that decision makers can easily make sense of the evidence. Additionally, we plan to publish a scientific paper in a peer-reviewed journal, so that the work is widely disseminated. We will aim for a REF returnable publication in a high impact journal, and will prioritise open access publication to increase the availability of the published research. An authorship and paper plan, following BMJ guidance, will be agreed by the research team and, where required, members of the project advisory group. We will seek to present the findings to a number of multidisciplinary conferences and networks, such as the International Society of Wheelchair Professionals, the Rehabilitation Engineering and Assistive Technology Society of North America (RESNA), the International Alliance of Academies of Childhood Disabilities, the NHS Wheelchair Managers Forum and the British Academy of Childhood Disability.

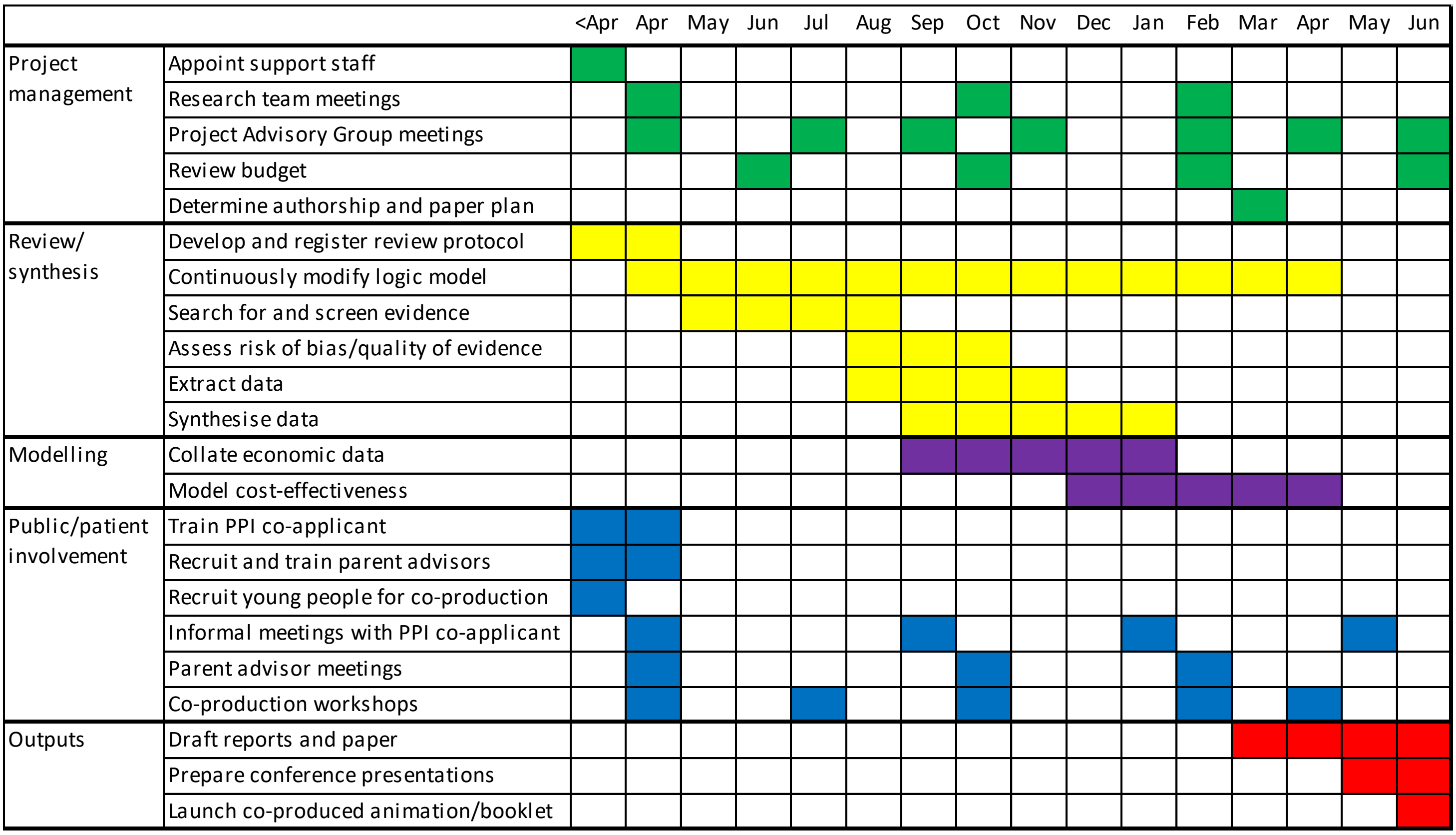
We will also target specific, key unidisciplinary networks such as the different allied health professional groups and the Health Economists' Study Group.

Within our wider dissemination strategy, a key projected output will be an animation and booklet summarising the study methods and findings, co-produced with young powered mobility users. These will be an accessible and engaging means of disseminating the study findings to children, families, clinicians, decision makers, and researchers within the NHS and the international childhood disability community. Ms Jennifer McAnuff (co-investigator) will jointly facilitate the co-production with Ms Aimee Grayston (Patient and Public Involvement co-investigator), and Ms Lucy Barker, a participatory artist who works with young people and vulnerable groups to design, conduct, and disseminate research and wider arts-based initiatives. Ms Barker has considerable experience of delivering high impact co-production projects. For example, in 2016 she co-produced an animation and booklet with young survivors of child sexual exploitation that featured on Radio 4’s Today Programme and was selected as a case study for Children in Need 2017. She is currently working with Ms McAnuff and a group of young people with disabilities to disseminate an NIHR-funded study on childhood disability participation interventions.

By synthesising evidence in this field and informing decision making, this research could lead to a fundamental change in NHS provision of powered mobility equipment for young children. If the findings demonstrate that early provision of powered mobility is cost-effective (compared to later provision) this will be a significant step towards evidence-based and transparent provision of early powered mobility for children. We are aware of the current state of literature in this field, and acknowledge that there is an absence of high quality effectiveness evidence relating to early powered mobility, and almost a total absence of economic literature (Bray et al, 2014). We plan to use pragmatic and novel approaches to collating data, using diverse sources. This research will provide a new understanding of the relative cost-effectiveness of early powered mobility and will be a foundation for further research in this field. The project is likely to lead to further primary research, for instance a clinical trial or cohort study to definitively establish the effectiveness and cost-effectiveness of early powered mobility.

PROJECT MANAGEMENT

As Chief Investigator, Dr Nathan Bray will be responsible for the overall management of this research project. At the beginning of the project the research plan and timetable, covering all key time-scales and objectives, will be agreed by the project team. The project plan and timetable will be used as a key indicator of progress during the project, as remaining within the specified time-scales will help to ensure that the budget is adhered to and the project objectives are met (see Figure 2). The core research team will teleconference on a monthly basis to discuss progress of the project. Every four months a face-to-face meeting will be arranged with all co-investigators and support staff.



		<Apr	Apr	May	Jun	Jul	Aug	Sep	Oct	Nov	Dec	Jan	Feb	Mar	Apr	May	Jun
Project management	Appoint support staff	🟩															
	Research team meetings		🟩						🟩				🟩				
	Project Advisory Group meetings		🟩			🟩		🟩		🟩			🟩		🟩		🟩
	Review budget				🟩				🟩				🟩				🟩
	Determine authorship and paper plan													🟩			
Review/ synthesis	Develop and register review protocol	🟨	🟨														
	Continuously modify logic model		🟨	🟨	🟨	🟨	🟨	🟨	🟨	🟨	🟨	🟨	🟨	🟨	🟨		
	Search for and screen evidence			🟨	🟨	🟨	🟨										
	Assess risk of bias/quality of evidence						🟨	🟨	🟨								
	Extract data						🟨	🟨	🟨	🟨							
	Synthesise data							🟨	🟨	🟨	🟨	🟨					
Modelling	Collate economic data							🟪	🟪	🟪	🟪	🟪					
	Model cost-effectiveness										🟪	🟪	🟪	🟪	🟪		
Public/patient involvement	Train PPI co-applicant	🟦	🟦														
	Recruit and train parent advisors	🟦	🟦														
	Recruit young people for co-production	🟦															
	Informal meetings with PPI co-applicant		🟦					🟦				🟦				🟦	
	Parent advisor meetings		🟦						🟦				🟦				
	Co-production workshops		🟦			🟦			🟦				🟦		🟦		
Outputs	Draft reports and paper													🟥	🟥	🟥	🟥
	Prepare conference presentations															🟥	🟥
	Launch co-produced animation/booklet																🟥

Figure 2: Plan of investigation

The two institutions, Bangor University and Newcastle University, will work closely together to ensure that the project is completed appropriately. Each institution will employ one post-doctoral research support staff member to work on the project, and they will be encouraged to work closely together and communicate on at least a weekly basis. They will be supported and supervised throughout the project by one of the co-investigators. Administrative support has also been costed into the project to help undertake administrative duties such as booking travel and processing expenses.

A budget review will be undertaken on a quarterly basis to ensure that all costs are within set limits. An appointed member of the finance team at each institution will help to monitor the budget, with

Bangor University taking overall responsibility as the lead partner. The financial budget for the project has been explicitly stated and costed appropriately. If any major funding issues are encountered we will discuss with the funder any possible solutions, for instance revisiting the allocation of funding within the workstreams. We have consulted with our local R&D department, and have been informed that this project will not require R&D or ethical approval.

We have established an international Project Advisory Group including a wide range of clinical, academic, and strategic stakeholders with expertise in powered mobility interventions, childhood disability, and NHS service delivery and commissioning. The group includes representatives from three leading UK charities with expertise in the implementation, benefits, and costs of powered mobility interventions. Designability is an assistive technology charity that has pioneered early powered mobility through its design and manufacture of the Wizzy-Bug, a powered wheelchair for children under 5 years. Queen Elizabeth's Foundation for Disabled People is a national disability charity with a nationwide loan scheme for the Bugzi, an indoor powered wheelchair for children aged 18 months and over. Whizz-Kidz provides mobility equipment for children and young people, and has a long-established therapy team with expertise in early powered mobility in particular.

In addition, we have engaged TinyTRAX, an independent designer and manufacturer of powered wheelchairs for young children in the UK. We have also engaged the Council for Disabled Children, the umbrella body for the disabled children's sector in the UK, which includes over 250 member organisations and works closely with central government to influence national policy; and the British Academy of Childhood Disability, a multidisciplinary specialty group of the Royal College of Paediatrics and Child Health, that promotes the development of quality standards and guidelines for good practice in the field of child development and disability. We will work flexibly with the Project Advisory Group by convening targeted meetings and using brief online surveys to draw on individuals' expertise at key decision-making points throughout the study. We envisage drawing on each expert advisor on 2-3 occasions, and have agreed with advisors to conduct all meetings by teleconference to maximise feasibility.

PATIENT AND PUBLIC INVOLVEMENT

Our PPI co-investigator is Ms Aimee Grayston, a young adult with significant personal experience of powered mobility in the context of her long-term physical and communication disabilities. Ms Grayston has been a volunteer for Leeds Community Healthcare NHS Trust for four years, where she leads quality improvement and patient involvement projects within Children's Services. She will provide a unique perspective on the benefits and costs of powered mobility and support the dissemination of study findings. Ms Grayston currently works with Ms Jennifer McAnuff, who will oversee the training and support she will require to carry out her role. We have used INVOLVE guidance (INVOLVE, 2016) to fully and realistically cost Ms Grayston's role as a co-investigator, including her engagement in full team meetings and further informal update meetings, attendance at a UK-based conference, and involvement in our planned co-production activities.

We will actively engage young powered mobility users as key stakeholders throughout the study. We will work with 'AniMates' to co-produce an accessible and engaging animation and booklet summarising the study methods and findings. AniMates is a group of young research advisors – three of whom use powered mobility – who work with researchers and artists to create animated films and broader artworks to shape and share research findings. We will work with AniMates across five co-production workshops throughout the study, starting these in the early stages to ensure adequate time for influencing the research team's thinking on the benefits and costs of powered mobility, and understanding study methods and findings. We will also engage young children (aged <5 years) through Designability, a UK assistive technology charity with an active online community of families using early powered mobility. We will invite them to provide visual materials about their powered mobility to include in the animation, e.g. photographs and videos of themselves using powered mobility, key words describing the benefits and costs, siblings' art work etc. Ms Jennifer McAnuff will lead the engagement of children and young people, ensure their training and support needs are met, and maintain communication and relationships with their parents. To cost young people's involvement, we have used INVOLVE and government guidance, (INVOLVE, 2016; https://www.gov.uk/child-employment/paying) and our previous experience of what has been acceptable in similar projects, including high street gift vouchers and small prizes to acknowledge their contributions, travel and carer expenses, and an event to launch the co-produced animation, celebrate their involvement, and bring the project to a close.

Our Project Advisory Group includes three parents of children who received powered mobility interventions at different ages (under and over 5 years). We have recruited the parent advisors through Designability and their role will be to help us continuously refine our logic model and advise us on how powered mobility interventions may vary, for example according to children's age or severity of disability. To ensure these advisory roles are feasible for parents, we will not ask them to travel to meetings, and have realistically costed their time and expenses (INVOLVE, 2016) for approximately three meetings via telephone, Skype, or videoconference throughout the study. Ms Jennifer McAnuff will lead on maintaining all relationships with parent advisors throughout the study, and will oversee their training and support requirements.

Our Project Advisory Group also includes representatives from three leading UK organisations who will raise awareness of the study and disseminate the findings through their impact pathways directly into the childhood disability community. Cerebra supports families of children with brain conditions such as cerebral palsy, and is particularly focused on bringing together researchers and parents to generate new insights in childhood disability. Spinal Muscular Atrophy (SMA) Support UK connects families affected by SMA with the latest research on interventions. Children with cerebral palsy or SMA constitute two significant populations of current/potential users of early powered mobility. We have also engaged the National Network of Parent Carer Forums, which comprises 152 local parent carer forums with over 80,000 parent carer members, and works closely with health authorities and clinical commissioning groups to influence strategic and operational service commissioning, design, and development.

EXPERTISE OF PROJECT TEAM

None of the co-investigators have any conflicts of interest to declare.

Nathan Bray: Dr Bray's PhD research was one of the first applications of health economics to wheelchair interventions for disabled children. He has published a mixed-method systematic review regarding the provision of wheelchairs for children. Nathan was awarded a post-doctoral fellowship by HCRW to develop a novel approach to measuring health-related quality of life and utility for people with mobility limitations. As PI, Dr Bray will lead the project and provide specific health economics expertise relating to paediatric mobility equipment provision.

Fiona Beyer: Ms Beyer is an experienced information specialist and systematic reviewer. She has previously worked for Cochrane and the Centre for Reviews and Dissemination, and is now based in the Evidence Synthesis Team at the Institute of Health and Society, Newcastle University. She is an author on several Cochrane and NIHR-funded systematic reviews.

Dawn Craig: Dr Craig is a senior Health Economist, and Deputy Director of the NIHR Innovation Observatory, and has extensive experience in systematic reviews, evidence synthesis methodologies and model based economic evaluations. She has led and contributed to many evidence syntheses projects for NIHR and other funders. Dr Craig is the Lead for the Evidence Synthesis group at the Institute of Health and Society, Newcastle University, and will support the design and conduct of the evidence synthesis.

Rhiannon Tudor Edwards: Prof Edwards, a senior health economist and co-director of the Centre for Health Economics and Medicines Evaluation, has a great deal of experience in the design, conduct and analysis of economic evaluations of complex interventions, and has worked in the area of return on investment from early years interventions. Prof Edwards will provide methodological oversight for the economic model design and analysis, and supervisory support for junior team members.

Aimee Grayston: Ms Grayston, PPI co-investigator, brings significant personal experience of using powered mobility as a young adult with complex physical and communication disabilities, and professional experience of engaging NHS decision makers in quality improvement projects. She will use her unique perspective to guide the search strategy and data extraction/interpretation and help specify benefits and costs for the economic model. She will also be involved in disseminating study findings by jointly leading a conference presentation, and supporting the co-production with young people.

Jennifer McAnuff: Ms McAnuff, HEE/NIHR Clinical Doctoral Research Fellow and Clinical Lead Occupational Therapist, brings experience in implementing allied health interventions in NHS childhood neurodisability services, conducting systematic reviews, and designing and leading public and patient involvement (PPI) activities with children, young people, and parents. She will support the evidence synthesis and lead the PPI strategy, supporting Ms Grayston (PPI co-investigator) and parent advisors to carry out their roles, and working with children, young people, and named charitable organisations to disseminate the study findings.

Jane Noyes: Professor of Health and Social Services Research and Child Health, primary researchers and systematic review methodologist. Co-Chair of the Cochrane Methods Executive, Member of the Cochrane Scientific Committee, and Lead of the Cochrane Qualitative and Implementation Methods Group. Noyes has led and contributed to several methodological developments in systematic review methodology (from methods of synthesis, classification of complex interventions and reporting guidelines, and is lead author on Cochrane’s new guidance on the use of Social Theories in systematic reviews. Prof Noyes will provide methodological oversight.

Louise Tanner: Dr Tanner has worked as a Research Associate in the Evidence Synthesis Team at the Institute of Health and Society, Newcastle University, since March 2017. Dr Tanner previously gained two BSc (Hons) degrees, in Applied Psychology and Biology, from Durham University. Her Postgraduate qualifications were gained from Newcastle University and include a PG Cert. in Public Health and Health Serviced Research; an MSc in Social Science and Health; and a PhD in recognition of a thesis entitled “What determines spatiotemporal variations in cold-weather-related mortality in England?”.

Lorna Tuersley: Dr Tuersley is experienced in patient preferences and healthcare information communication. A pharmacist, Dr Tuersley gained her BPharm at Cardiff University, with experience in hospital pharmacy followed by medical/marketing research roles in the pharmaceutical industry. She undertook a PhD in healthcare information seeking for self-care from the University of Manchester. Most recently, Dr Tuersley developed the Wheelchair outcomes Assessment Tool for Children (WATCh), which is the first patient-centred outcome measure designed specifically for paediatric wheelchair services.

Niina Kolehmainen: Dr Kolehmainen, a senior clinical lecturer and consultant allied health professional, brings experience in research on improving effectiveness, empowerment, and efficiency of NHS child health interventions. With expertise in reviewing, specifying, and developing complex NHS interventions (non-drug and behaviour change), she will lead the evidence synthesis and work together with Dr Bray to specify the benefit and cost attributes for the economic model.

REFERENCES

Anderson LM, Petticrew M, Rehfuess E, Armstrong R, Ueffing E, Baker P, Francis D, and Tugwell P. Using logic models to capture complexity in systematic reviews. Res Syn Meth 2011;2:33–42.

Barton E, Reichow B, Schnitz A, Smith I, Sherlock D. A systematic review of sensory-based treatments for children with disabilities. Res Dev Disabil 2015 2;37(0):64-80.

Bottos M, Bolcati C, Sciuto L, Ruggeri C, Feliciangeli A: Powered wheelchairs and independence in young children with tetraplegia. Developmental Medicine and Child Neurology 2001;43(11):769–777.

Bray N, Noyes J, Edwards RT, Harris N. Wheelchair interventions, services and provision for disabled children: a mixed-method systematic review and conceptual framework. BMC Health Services Research 2014;14:309.

Briggs A, Sculpher M. An introduction to Markov modelling for economic evaluation. Pharmacoeconomics 1998;13(4):397–409.

Brunton G, Oliver S, Oliver K, Lorenc T. A Synthesis of Research Addressing Children's, Young People's and Parents' Views of Walking and Cycling for Transport. London: EPPI-Centre, Social Science Research Unit, Institute of Education, University of London; 2006.

Centre for Reviews and Dissemination: Systematic Reviews: CRD’s Guidance for Undertaking Reviews in Health Care. York: University of York; 2009.

Deitz J, Swinth Y, White O. Powered mobility and preschoolers with complex developmental delays. American Journal of Occupational 2002;56(1):86-96.

Department of Health. Annual report of the Chief Medical Officer 2012. Our children deserve better: prevention pays. London: Department of Health; 2013.

Edwards C, Fox R, Gillard S, Gourlay S, Guven P, Jackson C, Chambers M, Drennan V. Explaining health managers' information seeking behaviour and use: Final report. NIHR Service delivery and organisation programme 2013. London: NIHR; 2013.

Edwards RT, Bryning L, Lloyd-Williams H. Transforming Young Lives across Wales: The Economic Argument for Investing in Early Years. Bangor: Centre for Health Economics and Medicines Evaluation; 2016.

Furumasu J, Tefft D, Guerette P. The impact of early powered mobility on young children's play and psychosocial skills. Paper presented at: International Seating Symposium. 24th Annual Meeting; 2008 March 6–8; Vancouver, Canada.

Grant MJ, Booth A. A typology of reviews: an analysis of 14 review types and associated methodologies. Health Information & Libraries Journal 2009;26:91-108.

Guyatt GH, Oxman AD, Vist GE, Kunz R, Falck-Ytter Y, Alonso-Coello P, Schünemann HJ. GRADE: an emerging consensus on rating quality of evidence and strength of recommendations. British Medical Journal 2008;336(7650):924-926.

Hartling LBK, Harvey K, Santaguida PL, Viswananthan M, Dryden DM. Developing and Testing a Tool for the Classification of Study Designs in Systematic Reviews of Interventions and Exposures. Agency for Healthcare Research and Quality, 2010 AHRQ Publication No. 11-EHC-007.

Higgins JP, Altman DG. Chapter 8: Assessing Risk of Bias in Included Studies. 2011. In: Cochrane Handbook for Systematic Reviews of Interventions Version 510 (updated March 2011). The Cochrane Collaboration. Available from: Available from www.handbook.cochrane.org.

Hoffmann TC, Glasziou PP, Boutron I, Milne R, Perera R, Moher D, Altman DG, Barbour V, Macdonald H, Johnston M, Lamb SE, Dixon-Woods M, McCulloch P, Wyatt JC, Chan A-W, Michie S. Better reporting of interventions: template for intervention description and replication (TIDieR) checklist and guide. BMJ 2014;348:g1687.

Huang H, Ragonesi CB, Stoner T, Peffley T, Galloway J. Modified Toy Cars for Mobility and Socialization: Case Report of a Child With Cerebral Palsy. Pediatric Physical Therapy 2014;26(1):76-84.

INVOLVE. Policy on payment of fees and expenses for members of the public actively involved with INVOLVE. Southampton: INVOLVE Coordinating Centre; 2016.

Jones MA, McEwen IR, Hansen L. Use of power mobility for a young child with spinal muscular atrophy. Physical Therapy 2003;83(3):253–262.

Jones MA, McEwan IR, Neas BR. Effects of Power Wheelchairs on the development and function of young children with severe motor impairment. Pediatric Physical Therapy 2012;24(2):131-140.

Kneale D, Thomas J, Harris K. Developing and Optimising the Use of Logic Models in Systematic Reviews: Exploring Practice and Good Practice in the Use of Programme Theory in Reviews. PLoS ONE 2015;10(11):e0142187.

Kolehmainen N, McAnuff J, Tissen-Budde A. Activity and participation interventions for children with movement problems: a systematic review of Rehabilitation Intervention Techniques [In Preparation; 2018].

Lewin S, Glenton C, Munthe-Kaas H, Carlsen B, Colvin CJ, Gülmezoglu M, Noyes J, Booth A, Garside R, Rashidian A. Using Qualitative Evidence in Decision Making for Health and Social Interventions: An Approach to Assess Confidence in Findings from Qualitative Evidence Syntheses (GRADE-CERQual). PLoS Med 2015;12(10): e1001895.

Livingstone R, Field D. Systematic review of power mobility outcomes for infants, children and adolescents with mobility limitations. Clin Rehabil 2014;28(10):954-964.

Livingstone R, Field D. The child and family experience of power mobility: a qualitative synthesis. Developmental Medicine and Child Neurology 2015;57(4):317-327.

Logan SW, Schreiber M, Lobo M, Pritchard B, George L, Galloway JC. Real-World Performance: Physical Activity, Play, and Object-Related Behaviors of Toddlers With and Without Disabilities. Pediatric Physical Therapy 2015;27(4):433-441.

Lynch A, Ryu JC, Agrawal S, Galloway JC. Power mobility training for a 7-month-old infant with spina bifida. Pediatr Phys Ther 2009;21:362–368.

McPherson A, Keith R, Swift J. Obesity prevention for children with physical disabilities: a scoping review of physical activity and nutrition interventions. Disabil Rehabil 2014;36(19):1573-87.

Morris C, Janssens A, Allard A, Thompson Coon J, Shilling V, Tomlinson R, et al. Informing the NHS Outcomes Framework: what outcomes of NHS care should be measured for children with neurodisability?. Health Serv Deliv Res 2014;2(15).

Morris C, Simkiss D, Busk M, Morris M, Allard A, Denness J, Janssens A, Stimson A, Coghill J, Robinson K, Fenton M, Cowan K. Setting research priorities to improve the health of children and young people with neurodisability: a British Academy of Childhood Disability-James Lind Alliance Research Priority Setting Partnership. BMJ Open 2015;5:e006233.

NHS England. Developing a Wheelchair Tariff Pilot Programme. London: NHS England; 2016.

NHS Improving Quality. Right chair, right time, right now. London: NHS Improving Quality; 2014.

Novak I, Mcintyre S, Morgan C, Campbell L, Dark L, Morton N, et al. A systematic review of interventions for children with cerebral palsy: state of the evidence. Dev Med Child Neurol 2013;55(10):885-910.

Noyes J, Hendry M, Booth A, Lewin S, Glenton C, Garside R, Chandler J. Guidance for review authors on choice and use of social theory in complex intervention reviews. Version 1. London: Cochrane; 2015.

Ogilvie D, Fayter D, Petticrew M, Sowden A, Thomas S, Whitehead M, Worthy G. The harvest plot: A method for synthesising evidence about the differential effects of interventions. BMC Medical Research Methodology 2008;8(8).

Ouzzani M, Hammady H, Fedorowicz Z, Elmagarmid A. Rayyan—a web and mobile app for systematic reviews. Systematic reviews 2016;5:210.

Pluye P, Robert E, Cargo M, Bartlett G, O'Cathain A, Griffiths F, Boardman F, Gagnon MP, Rousseau MC. Proposal: A mixed methods appraisal tool for systematic mixed studies reviews. 2011. Retrieved on [25/09/2017] from http://mixedmethodsappraisaltoolpublic.pbworks.com.

Ragonesi CB, Galloway J. Short-term, Early Intensive Power Mobility Training: Case Report of an Infant at Risk for Cerebral Palsy. Pediatric physical therapy 2012;24:141-8.

Reeves BC, Higgins JPT, Wells GA. Chapter 13: Including non-randomized studies. 2011. In: Cochrane Handbook for Systematic Reviews of Interventions Version 510 (updated March 2011). The Cochrane Collaboration. Available from www.handbook.cochrane.org

Scianni A, Butler JM, Ada L, Teixeira-Salmela LF. Muscle strengthening is not effective in children and adolescents with cerebral palsy: a systematic review. Australian Journal of Physiotherapy 2009;55(2):81-87.

Seo H-J, Kim SY, Lee YJ, Jang B-H, Park J-E, Sheen S-S, et al. A newly developed tool for classifying study designs in systematic reviews of interventions and exposures showed substantial reliability and validity. J Clin Epidemiol 2016;70:200-205.

Siebert U. When should decision-analytic modelling be used in the economic evaluation of healthcare? Eur J Health Econom 2003;4:143–150.

Sonnenberg FA, Beck JR. Markov models in medical decision making: a practical guide. Medical Decision Making 1993;13(4):322–338.

Stein R, Jessop D. A noncategorical approach to chronic childhood illness. Public Health Rep 1982;97(4):354-362.

Sterne JAC, Hernán MA, Reeves BC, Savović J, Berkman ND, Viswanathan M et al. ROBINS-I: a tool for assessing risk of bias in non-randomised studies of interventions. BMJ 2016;355:i4919.

Sullivan SD, Mauskopf JA, Augustovski F, Jaime Caro J, Lee KM, Minchin M, Orlewska E, Penna P, Rodriguez Barrios JM, Shau WY. Budget impact analysis-principles of good practice: report of the ISPOR 2012 budget impact analysis good practice ii task force. Value Health 2014;17:5–14.

Swan J, Clarke A, Nicolini D, Powell J, Scarbrough H, Roginski C, Gkeredakis E, Mills P, Taylor-Phillips S. Evidence in Management Decisions (EMD): advancing knowledge utilization in healthcare management: Final report. NIHR Service delivery and organisation programme 2012. London: NIHR; 2012.

Swan J, Gkeredakis E, Manning RM, Nicolini D, Sharp D & Powell J. Improving the capabilities of NHS organisations to use evidence: a qualitative study of redesign projects in Clinical Commissioning Groups. Health Serv Deliv Res 2017;5(18).

Tatla S, Sauve K, Virji-Babul N, Holsti L, Butler C, Van Der Loos H. Evidence for outcomes of motivational rehabilitation interventions for children and adolescents with cerebral palsy: an American Academy for Cerebral Palsy and Developmental Medicine systematic review. Dev Med Child Neurol 2013;55(7):593-601.

Tefft D, Guerette P, Furumasu J. Cognitive predictors of young children’s readiness for powered mobility. Developmental Medicine & Child Neurology 1999;41:665-670.

Tefft D, Guerette P, Furumasu J. The impact of early powered mobility on parental stress, negative emotions, and family social interactions. Physical and Occupational Therapy in Pediatrics 2011;31(1):4–15.

Uchiyama I, Anderson DI, Campos JJ, Witherington D, Frankel CB, Lejeune L, Barbu-Roth M. Locomotor Experience Affects Self and Emotion. Developmental Psychology 2008;44(5):1225–1231.

Varni J, Seid M, Rode C. The PedsQL: measurement model for the pediatric quality of life inventory. Med Care 1999;37:126-39.

Viswanathan M, Berkman N, Dryden D, Hartling L. Assessing risk of bias and confounding in observational studies of interventions or exposures: further development of the RTI item bank. Methods Research Report. Rockville, MD: Prepared by RTI–UNC Evidence-based Practice Center 2013.

Wilson PM, Farley K, Bickerdike L, Booth A, Chambers D, Lambert M, et al. Effects of a demand-led evidence briefing service on the uptake and use of research evidence by commissioners of health services: a controlled before-and-after study. Health Serv Deliv Res 2017;5(5).

APPENDIX 1. EMPoWER illustrative example keywords and search strategy

Developed by fiona.beyer@ncl.ac.uk

Suggested keywords – please edit/add as appropriate
We will search for [Facet 1] AND [Facet 2 OR Facet 3]

Brackets denote optional forms of a word.

Facet 1: children		
child(ren) or adolescent (ce) or teen(s)/(ager(s)) or youth(s) or girl(s) or boy(s) p(a)ediatric(s) or juvenile(s) or infant(s) or "young people" or "young person" or "young adult" or " young men" or "young women" or schoolchild(ren)		
Facet 2: powered mobility		
power(ed) electric(al) motorised (zed) assist(ed)	within 5 words of	(wheel)chair(s) pushchair(s) equipment car(s) scooter(s) toy(s) mobility
Facet 3: independent mobility		
independent(ly) self-directed autonomous	within 3 words of	mobile mobility move/moving walk(ing) crawl(ing)

Example strategy from CINAHL

- Note it goes backwards – line 1 at the bottom
- MH denotes thesaurus headings, TI title terms, AB abstract terms, ZG age groups

S14	S10 OR S13	(1,589)
S13	S11 OR S12	(116)
S12	MH "Wheelchairs, Powered in infancy and childhood"	(19)

S11	MH "Assistive Technology Devices in infancy and childhood")	(101)
S10	S5 AND S9	(1,589)
S9	S6 OR S7 OR S8	(8,823)
S8	TI ((independent* or self* or autonomous*) N3 (mobile or mobility or move* or moving or walk* or crawl*)) OR AB ((independent* or self* or autonomous*) N3 (mobile or mobility or move* or moving or walk* or crawl*))	(3,112)
S7	TI ((power* or electric* or motorised or motorized or assist*) N5 (chair* or wheelchair* or pushchair* or equipment* or car or cars or scooter* or toy* or mobility)) OR AB ((power* or electric* or motorised or motorized or assist*) N5 (chair* or wheelchair* or pushchair* or equipment* or car or cars or scooter* or toy* or mobility))	(1,656)
S6	(MH "Assistive Technology Devices") OR (MH "Wheelchairs, Powered")	(4,707)
S5	S1 OR S2 OR S3 OR S4	(612,322)
S4	TI (infant or infants or "young people" or "young person" or "young adult" or " young men" or "young women" or "schoolchild*") OR AB (infant or infants or "young people" or "young person" or "young adult" or " young men" or "young women" or "schoolchild*")	(73,065)
S3	TI (child* or adolescen* or teen or teens or teenager* or youth or youths or girl or girls or boy or boys or pediatric* or paediatric* or juvenil*) OR AB (child* or adolescen* or teen or teens or teenager* or youth or youths or girl or girls or boy or boys or pediatric* or paediatric* or juvenil*)	(334,011)
S2	(MH "Child") OR (MH "Child, Disabled") OR (MH "Infant+") OR (MH "Child, Preschool")	(359,737)
S1	(ZG "adolescent: 13-18 years") or (ZG "child, preschool: 2-5 years") or (ZG "child: 6-12 years") or (ZG "infant: 1-23 months")	(462,198)