Powered mobility interventions for very young children with mobility limitations to aid participation and positive development: the EMPoWER evidence synthesis

Nathan Bray,[1,2*] Niina Kolehmainen,[3,4] Jennifer McAnuff,[3] Louise Tanner,[3] Lorna Tuersley,[2] Fiona Beyer,[3] Aimee Grayston,[5] Dor Wilson,[3] Rhiannon Tudor Edwards,[1,2] Jane Noyes[1] and Dawn Craig[3]

[1]School of Health Sciences, Bangor University, Bangor, UK
[2]Centre for Health Economics and Medicines Evaluation, Bangor University, Bangor, UK
[3]Population Health Sciences Institute, Newcastle University, Newcastle upon Tyne, UK
[4]Newcastle upon Tyne Hospitals NHS Foundation Trust, Newcastle upon Tyne, UK
[5]Children's Services, Leeds Community Healthcare NHS Trust, Leeds, UK

*Corresponding author n.bray@bangor.ac.uk

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Scientific summary

The EMPoWER evidence synthesis

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Scientific summary

Background

Mobility impairment is the leading cause of disability in the UK, accounting for 49% of reported disabilities. Around 8% of children in the UK have a disability, equating to 1.1 million disabled children. Of this number, 19% are estimated to have some form of mobility limitation. The ability of public services, including health and social care and education, to improve the life of children with mobility limitations is currently significantly hindered by a lack of effective interventions. The patients, policy-makers and health-care providers all agree that improving interventions is an urgent priority.

Early provision of powered mobility for very young children (aged < 5 years) may help to enable self-directed movement in children who are limited in their movements or unable to move, and could consequently trigger positive developmental changes, similar to crawling. This, in turn, could facilitate exploration, learning and further 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. This places the children at greater risk of secondary disabilities in terms of general independence, autonomy and participation in daily life across home, education and leisure. It is thought that early powered mobility could prevent these secondary disabilities developing by enabling self-directed mobility and exploration.

Powered mobility interventions are complex interventions with several elements and synergistic outcomes and benefits. The use of powered mobility takes place in, and is influenced by, the child's physical and social environment, and the exact features and delivery of the intervention elements vary depending on a child's age and/or developmental stage and impairments.

The NHS provides mobility equipment for ≈ 60,000 children each year. However, the current provision of powered mobility interventions commonly focuses on children aged > 5 years, with the assumption that very young children do not benefit from it, at least not sufficiently for it to be cost-effective. If the hypotheses about the role of early self-directed mobility in the prevention of secondary problems are true, then current provision is likely to be a missed opportunity to improve the outcomes of children with disabilities and yield the best returns for public resource across the life course. The key remaining question is whether or not early provision (i.e. before the age of 5 years) is incrementally more effective and cost-effective than later provision (i.e. from the age of 5 years onwards).

Objectives

The research question was as follows: is the earlier provision of powered mobility to very young children more cost-effective than currently more common provision to children aged ≥ 5 years?

The aim was to examine and model the relative effectiveness and cost-effectiveness of powered mobility interventions for very young children with mobility limitations, compared with the more

common practice of powered mobility provision for children aged ≥ 5 years. To do so, the following key objectives were defined:

- to identify and synthesise quantitative, qualitative and mixed-method evidence to determine –
  - the effectiveness and cost-effectiveness of powered mobility interventions for children with mobility limitations, and the wider impacts on health services and society
  - the acceptability, feasibility and anticipated outcomes of relevant interventions from multistakeholder perspectives (e.g. children, parents, service providers and commissioners)
  - the long-term implications of self-directed or independent mobility for very young children (aged < 5 years), compared with older children (aged ≥ 5 years)
- to examine the economic costs and benefits of powered mobility interventions for children by –
  - 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 multiperspective disaggregated cost–consequence framework
  - modelling the relative cost-effectiveness of powered mobility equipment for very young children (aged < 5 years), compared with standard NHS practice (provision of such equipment for children aged ≥ 5 years).

Methods

This study was planned as a mixed-methods evidence synthesis and economic modelling study, incorporating quantitative, qualitative, mixed-methods and economic evidence. The key intended outputs were an overview of the current evidence of powered mobility interventions for children; a logic model of effectiveness and cost-effectiveness for planning and evaluating future interventions and commissioning of services; an economic model, budget impact analysis and value-of-information analysis for early powered mobility; and recommendations for powered mobility provision and for future research.

The initial programme theory, developed in the form of a novel logic model, was utilised as a conceptual starting point and was refined throughout the review processes to produce a definitive logic model. The active intervention was provision of powered mobility for children aged < 5 years, and the comparator was provision of powered mobility for children aged ≥ 5 years. The systematic review protocol was registered on PROSPERO as CRD42018096449.

Review 1: effectiveness and cost-effectiveness of powered mobility for children

We reviewed evidence relating to the effectiveness, cost-effectiveness, acceptability, feasibility and anticipated outcomes of paediatric powered mobility interventions, integrating a range of perspectives (children, parents, service providers, commissioners, society).

To identify relevant literature, we systematically searched a range of bibliographic databases [Cumulative Index to Nursing and Allied Health Literature (CINAHL), MEDLINE, EMBASE™ (Elsevier, Amsterdam, the Netherlands), Physiotherapy Evidence Database (PEDro), Occupational Therapy Systematic Evaluation of Evidence (OTseeker), Applied Social Sciences Index and Abstracts (ASSIA), PsycINFO, Science Citation Index (SCI; Clarivate Analytics, Philadelphia, PA, USA), Social Sciences Citation Index™ (SSCI; Clarivate Analytics), Conference Proceedings Citation Index – Science (CPCI-S; Clarivate Analytics), Conference Proceedings Citation Index – Social Science & Humanities (CPCI-SSH; Clarivate Analytics), Cochrane Central Register of Controlled Trials (CENTRAL), Cochrane Database of Systematic Reviews, Database of Abstracts of Reviews of Effects (DARE), NHS Economic Evaluation Database (NHS EED) and Health Technology Assessment (HTA) Database; searches covered 1946 to September 2019], reference lists of included papers, relevant journals and conference proceedings, and grey literature. For the search

terms, we used a combination of three facets: 'children', 'powered mobility' and 'independent mobility'. The searches were not restricted by outcome, disability/condition, study design, language or publication year. Papers were included if all three of the following criteria were met: (1) the study participants included children with significant mobility limitations defined using explicit criteria, (2) the intervention involved at least one of the five explicitly defined intervention elements and (3) the outcome (or the 'phenomenon of interest') was related to the child, their family, health or social care, or education. The searches were carried out in June 2018 and updated in October 2019.

Titles and abstracts were screened for inclusion by two independent reviewers, of which at least one was a topic expert; studies judged as being relevant were obtained as full texts. Methodological strengths and limitations of included studies were assessed using a range of tools, with focus on risk of bias in randomised controlled trials, risk of bias and confounding in non-randomised studies and risks to rigour in qualitative studies. A range of numeric and textual data were extracted from all included papers using a bespoke extraction form similar to previous reviews.

Two syntheses were carried out to make best use of the extracted evidence. First, we undertook a convergent mixed-methods evidence synthesis using a framework synthesis method. In this, textual and numeric data across studies were integrated to identify and develop key concepts relevant to the intervention, its outcomes, and feasibility and acceptability. We applied selected, mixed-methods-specific criteria to assess certainty of the evidence underpinning the resulting concepts. Second, we undertook a separate qualitative evidence synthesis using the Thomas and Harden (Thomas J, Harden A. Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Med Res Methodol* 2008;**8**:45) method of thematic synthesis. Findings in primary qualitative and relevant mixed-methods studies were translated and then transformed to look for new patterns and meanings that were not seen in individual primary studies. We subsequently applied the Grading of Recommendations Assessment, Development and Evaluation – Confidence in the Evidence from Reviews of Qualitative research (GRADE-CERQual) approach to assess the confidence in synthesised qualitative findings. The results from the two syntheses were subsequently compared and contrasted, and further integrated to develop a final, revised logic model for evaluations of effectiveness and cost-effectiveness of powered mobility interventions.

Review 2: long-term implications of independent mobility

A second systematic review, to identify evidence relating to the long-term implications of self-directed or independent mobility for very young children, compared with the implications for older children, used two broad facets to identify relevant papers: (1) independent mobility and (2) children or young people. The search resulted in > 47,000 potential papers; however, despite several attempts to identify relevant papers, we were unable to identify key papers relevant to the aim and had to conclude that it was not possible to systematically identify research studies related to the long-term implications of self-directed and independent mobility.

Economic modelling and budget impact analysis

The original intention was to develop an economic model to examine the long-term cost-effectiveness of early powered mobility. However, after the completion of the reviews, it became apparent that there were insufficient published data to develop a robust economic model or to carry out a value-of-information analysis.

As an alternative, we conducted a budget impact analysis (following International Society for Pharmacoeconomics and Outcomes Research guidance) to model the cost of current practice and those of hypothetical scenarios of increased access to powered mobility for children aged < 5 years. Cost data were generated from publicly available publications, and through consultation with NHS posture and mobility services, the National Wheelchair Managers Forum, charitable organisations, various wheelchair manufacturers and expert advisors to the project. A range of different direct costs were considered, including mobility equipment, accessories/modifications, repair/maintenance, training, home/vehicle adaptations and staff time.

Results

Review 1

We identified 5948 potentially relevant titles and abstracts, from which 221 were included in full-text screening. A further 16 relevant publications were identified from reference lists and 30 from the grey literature, and 50 were recommended by expert advisors; this resulted in a total of 317 publications for full-text screening. Of these, 89 publications (covering 89 studies) met the inclusion criteria.

The included studies comprised two randomised controlled trials; 18 qualitative studies; three mixed-methods studies; and 66 studies of other designs, such as observational and non-randomised trials. Nearly one-third of the studies were single subject studies, which often consisted of case reports or clinical cases.

Of the included studies, 39% reported data specifically for children aged < 5 years, and 28% for children aged ≥ 5 years. Half of the included studies described multiple diagnoses, and, overall, the studies covered a broad range of diagnoses. For the included quantitative studies, there were substantial concerns about the lack of control for potential confounding. For the qualitative and mixed-methods studies, all were judged to have used appropriate overall methodologies, but all had some form of methodological limitation.

Across the included studies, 70% investigated powered mobility equipment, 22% investigated powered mobility training and 8% investigated a combination of equipment and training. None explicitly investigated environmental or policy adaptations.

We found no conclusive evidence about the effectiveness or cost-effectiveness of powered mobility for children aged either < 5 or ≥ 5 years. However, the mixed-methods synthesis of all evidence, and an assessment of certainty of that evidence, found strong support that powered mobility interventions have a positive impact on children's movement and mobility, and moderate support for a positive impact on children's participation, play and social interactions, and on the safety outcome of accidents and pain. Limited support was found for the concepts of self-care, autonomy, choice/control, freedom and psychological consequences (i.e. confidence, motivation and cognition). Limited support was also found for the safety outcome of emotional consequences (positive and negative feelings for the parent or the child), and inconsistent support was found for children learning to drive powered mobility.

The qualitative synthesis provided further insights, particularly regarding the implementation, feasibility and acceptability of powered mobility for children (for both very young and older children). We found the acceptance of powered mobility to be a journey for both parents and children, and the child's development of powered mobility skills to be a continuum that was supported by experiential learning and play. In this journey, it appears to be important for parents and therapists to invest their time and provide the child with support for the intervention to have full impact. Similarly to the mixed-methods synthesis, the fit between the child, the equipment and the environment was found to be important, as were the outcomes related to a child's independence, freedom and self-expression.

Budget impact analysis

We estimate that, each year, £2.84M is spent on the provision of powered mobility to very young children in the UK. Of this, £1.89M is spent by the NHS, which is $< 2\%$ of the total current wheelchair service spend of the NHS. If the provision of powered mobility were to increase and the NHS were to take on third-sector provision/training, this would rise to £5.64M, or 5.2% of current wheelchair service spend, and cover 1375 very young children.

Conclusions and recommendations

The evidence supported two key, distinct ways of conceptualising the primary powered mobility outcome: movement and mobility. First is 'movement for movement's sake' (e.g. play, exploring, acting on the environment, autonomy, misbehaving, cognition and learning). This is relevant to all children, regardless of physical or cognitive ability, and appears to be the primary function of early powered mobility. In this, two further outcome chains are implied: (1) developmental benefits achieved through movement and (2) preparing children for becoming powered wheelchair users. Of these, the former is relevant to all children, whereas the latter can be difficult to prospectively judge in terms of which children this applies to.

Second is destination-focused mobility (e.g. getting from A to B, which is a key mechanism of participating at home, in education, in the community). This is essentially the conceptualised purpose of adult powered wheelchairs; although the review did find evidence of the importance of this dimension for children, it was only one of the two dimensions, and not always the most important one, especially for the very youngest of children. These two conceptualisations of powered mobility outcomes suggest that the provision of early powered mobility should not be considered through the same framework as provision of adult powered mobility, nor should it be treated as a stepping stone to powered wheelchair use. Instead, a key criterion for provision should include the potential of powered mobility to enable a child to move – for movement's sake or for the child to go from A to B – in order to promote the child's development, play, exploration and independence.

We further conclude that age is not the key factor in paediatric powered mobility provision. There are few data to support the restriction of powered mobility provision by age. Instead, the focus should be on providing powered mobility interventions in a developmentally appropriate manner, focusing on 'movement for movement's sake' in the first instance and establishing good 'fit' between the child, the powered mobility equipment and the child's environment (both physical and social). Therefore, comparing powered mobility effectiveness and cost-effectiveness before and after the age of 5 years may not be informative, because these interventions are likely to have different aims, purposes and conceptualisations.

Current NHS provision of early powered mobility covers only a limited proportion (50% at most) of very young children who could benefit from early powered mobility, with third-sector providers filling the gap in provision. Even if the NHS was to double provision of early powered mobility and take on third-sector provision, the costs would remain a small fraction of overall wheelchair service expenditure.

Study registration

This study is registered as PROSPERO CRD42018096449.

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This report

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