MORE THAN MOBILITY: A MIXED-METHOD SYSTEMATIC REVIEW OF WHEELCHAIR INTERVENTIONS FOR CHILDREN AND YOUNG PEOPLE WITH DISABILITIES

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ABSTRACT

Wheelchairs for disabled children (≤18 years) can provide health, developmental and social benefits, however not all children have access to the right equipment at the right time. We conducted the first systematic review to explore current UK policy, international effectiveness/cost-effectiveness evidence and service user perspectives in order to inform future research and service development. Fourteen policy/guidance documents and 22 studies were included. Powered wheelchairs were found to offer benefits in reduced need for caregiver assistance; improved communicative, personal-social and cognitive development; and improved mobility function and independent movement. Children and parents placed more emphasis on improving social skill and independence than functional outcomes. Policy intentions and aspirations are in line with the perspectives of children/parents, although translation of policy recommendations into practice is still lacking. There is currently a lack of high quality economic evidence. Development of outcome measures that reflect how children define effective wheelchair interventions are required. Translation of policy and guidance into practice is lacking and more effective implementation strategies are required to improve services and outcomes.

BACKGROUND

Approximately 5% of children worldwide (around 95 million children aged 14 or under) have a disability [1]. It is estimated that between 10% [2] and 15% [3] of the world's population live with some form of disability and one in ten disabled people require a wheelchair to provide essential mobility assistance [4]. Access to appropriate mobility equipment is a worldwide issue, particularly in low-income countries [5]. Disabling barriers include lack of adequate policy, services and funding [3], which limit appropriate supply of essential wheelchairs.

Independent mobility for disabled people and provision of equipment to facilitate this is considered a human right, with calls for all countries to ensure that disabled people are able to access essential equipment to promote mobility and independence [6]. Without adequate wheelchair provision many disabled people are caught in a cycle of poverty and depravation, lacking the ability to access education, work and social facilities [5]. Disabled people are more likely to be unemployed than non-disabled people, and when employed tend to earn less [3]. These issues also have national economic impacts due to loss of productivity and health service resource use [3].

In the United Kingdom (UK), around 800,000 children and young people live with a disability [7]. Of this number, It is estimated that 70,000 have unmet mobility needs [8]. Technological advances have led to improvements in the quality and diversity of equipment available for children. Many disabled children in the UK rely on the National Health Service (NHS), education and social services to provide essential mobility aids. However, advanced assistive mobility equipment is expensive and, due to budgetary constraints, can be difficult for children and young people to access.

AIMS AND OBJECTIVES

The overarching aim of this mixed-method systematic review was to explore current policy, effectiveness evidence, cost-effectiveness evidence and service user perspectives in order to develop a conceptual framework to inform future research and wheelchair service development. Four objectives were developed to inform searching, management and interpretation of evidence:

1. to determine the effectiveness and cost-effectiveness of wheelchairs for disabled children and young people
2. to better understand service user, parent and professional perspectives regarding wheelchairs for disabled children and young people
3. to explore current UK policy, not-for-profit organization (NFPO) publications and clinical guideline recommendations and intentions regarding wheelchair provision for children and young people
4. to determine if children's desired outcomes matched with existing policy aspirations and effectiveness evidence

METHODS

Design

A mixed-method systematic review design was used. Review questions and a protocol were developed to guide the review. Searches were conducted between January and

April 2012. An adapted Evidence for Policy and Practice Information and Co-ordinating Centre (EPPI-centre) methodology was utilized for evidence synthesis [9]. Evidence was streamed by evidence and methodology type and results were then synthesized across the streams in a final overarching synthesis. Thematic synthesis [10], narrative summary and narrative synthesis [11] were used to synthesise evidence.

Search Methods

Mixed-method and economic evidence searches were carried out separately to increase specificity of searches. Searched internet databases included Cochrane Collaboration Register and Library, Science Direct, CINAHL, Medline, ASSIA, PsychINFO, PubMed, Web of Science, DARE, NHS EED and HTA. All mixed-method and economic literature was from the last 15 years (February 1997 to February 2012). Reference list and hand-searching supplemented electronic searching. Grey literature was also included to limit publication bias. Studies exploring bio-mechanical impact of wheelchair interventions were excluded as the review focused on outcomes relevant to service-users and carers.

Only UK policy/NFPO literature from the last 10 years (March 2002 to March 2012) was considered for inclusion to avoid obsolete literature being included in the review. It was deemed too expansive to include all international policy literature in this review, thus the policy and NFPO literature stream was limited to UK publications.

Each study was independently critically analyzed for quality using a suitable quality appraisal tool. The general quality of studies was low to moderate. No studies were excluded based on methodological quality.

Evidence Synthesis

Evidence streamed to enable separate syntheses by evidence type. These streams were Intervention Evidence; Opinion Evidence; Policy and NFPO Literature; and Economic Evidence. As the statistical evidence could not be grouped and analyzed using meta-analysis due to heterogeneity, a narrative summary was conducted to form a structured narrative of the results. For the qualitative opinion evidence, thematic synthesis [10] was conducted in order to identify key themes of service user and professional perspectives on wheelchair provision. A final over-arching narrative synthesis framework was developed to draw together the results across the different streams of evidence. [11]

RESULTS AND NARRATIVE SUMMARY OF FINDINGS

Search outcomes

In total 4144 studies were found in the mixed-method evidence searches, of which 2393 duplicates were removed. After screening titles and abstracts, 76 full-texts were left. In total 20 were deemed eligible for inclusion: ten in the intervention evidence stream and fourteen in the opinion evidence stream (four studies were eligible for both streams of evidence). In total 389 studies were found in the economic evidence searches, of which 163 duplicates were removed. After screening titles and abstracts seven full-texts were left. In total two were deemed eligible for inclusion. A formal screening process was not required for the policy literature, as searches were conducted using search engines and searching of government and NFPO websites. In total 14 policy and NFPO reports were deemed eligible for inclusion. A total of 36 papers/publications were found eligible for inclusion in this review.

Intervention evidence

Evidence shows that wheelchairs (in particular powered wheelchairs [PWC]) are effective at improving pro-social behavior [12]; functional mobility [13-15]; developmental level in the domains of communication, cognition and personal-social [13]; receptive communication skills [14]; occupational performance [16]; child-initiated movement [17]; and play skills [12]. There was also evidence that PWC provision reduces need for caregiver assistance [13,14,18] and caregiver stress [19], and that children as young as 14 months can learn some degree of PWC driving competence [20].

Opinion evidence

The key issues associated with wheelchair services were long waiting times [21,22], poor maintenance procedures [21-23], strict eligibility criteria [23], differing opinions of needs [24,25] and lack of information [22,23,26]. Families were found to experience financial burden from having to pay for and maintain their child's essential equipment [22,23,25]. Furthermore, for parents there is lengthy process of coming to terms with their child's need for a wheelchair [24,27].

However, wheelchairs were deemed to offer many benefits, for instance provision of PWC was believed to facilitate development of independence in disabled children [16,22-24], which subsequently allows greater socialization [21]. Wheelchairs were perceived to offer a new lifestyle for children and their families [15,16,21,23,24] and offered improvements to quality of life (QoL) [19,23], ability to take part in age-appropriate activities [21], ability to take on responsibilities (like household chores) [24] and gave more freedom [25].

Structural barriers to wheelchair use included poor access to buildings [18,22,24,25], difficulty transporting equipment [18,19,21,24,25,26,27] and poor disabled parking facilities [22,25].

Policy and Guidelines

Comprehensive access to multi-disciplinary assessments of mobility needs was of high priority [28-31]. There were also recommendations for extended equipment

loan programs [31] and national consensus of eligibility criteria and outcomes [32,33]. Waiting time reductions were of highest priority for NFPOs and government bodies [28-30,34-39]. Several publications highlighted the need for equipment to be useable in all places required in order to maximize effectiveness [29,34,36]. There was also a call for assessment and provision to take into account the holistic needs of service users [29,35,36,38,40], as part of maximizing social, physical and lifestyle outcomes and promoting independence.

Economic evidence

Economic evidence was limited and of relatively low quality and relevance. Neilson et al [41] found the cost per quality-adjusted life year (QALY) (compared with a 'do nothing' scenario) for provision of a powered indoor/outdoor wheelchair ranged from £734 to £1378 (dependent on time horizon) based on a cost per wheelchair intervention ranging from £1500 to £2000. Inflation to 2011 prices [42,43] provides a cost per QALY of £1187 and £2229 (40 and 50 year time horizon respectively).

Frontier Economics [44] found that meeting unmet demand for wheelchairs within a state wheelchair service cost an extra £108,000 and provided an additional 10.7 to 14 QALYs. This resulted in a cost per QALY of between £7,700 and £9,800 to meet additional unmet demand.

Over-arching synthesis

An overarching synthesis of all evidence across the four streams identified six analytical themes. These themes encompass the findings from all of the different types of evidence and form the key recommendations from this review:

1. Higher quality wheelchair services must take into account the needs of the whole family
2. Children benefit more when psychosocial needs are considered alongside health needs
3. Children would benefit more if policy recommendations focussed on services meeting individual needs rather than following strict eligibility criteria
4. Without appropriate outcome measures holistic benefits of PWC provision cannot be evaluated
5. Children benefit more when physical outcomes of PWC use are seen as facilitators to wider holistic benefits, but lack of translation of evidence into practice hinders progress
6. Children benefit more when public buildings and spaces promote inclusion of people with disabilities

DISCUSSION

No major deviations from the protocol were noted. The major contribution to knowledge from this novel mixed-method review comes from the synthesis of diverse evidence to inform and guide future research and to build a holistic understanding of pediatric wheelchair provision and use.

Within this overall context, the most important finding is that for children and young people wheelchairs offer more than mobility; they offer enhanced independence, social integration, participation in age-appropriate activities and enhanced self-esteem. It is therefore paramount that wheelchair interventions are seen as more than mobility, and that they are seen has facilitators to a new way of life.

UK policy and NFPO recommendations are reflective of the perspectives of young wheelchair users and their families, but there is a lack of effective translation of policy and evidence into practice. Although policy recommendations do correlate with the opinion evidence, barriers to effective provision and use of wheelchairs have continued to prevail in UK NHS services over many years [28,36,37,40]. The key to improving outcomes for children and young people lies in improving service delivery, understanding what children want from their wheelchairs and translating this knowledge into practice.

The effectiveness evidence, although limited by quality, demonstrates that wheelchair interventions can have a range of positive effects beyond mobility. More evidence is required to understand how effective interventions can be achieved for all service users. This requires studies to use large sample sizes, robust methods and diverse outcome measures. The application of health economics could enable a better understanding of the cost-effectiveness of wheelchair interventions, and thus benefit service-commissioning and funding allocation, and enable these practices to be evidence-based and equitable. This would be beneficial in the UK and internationally.

Future research should focus on developing more appropriate outcome measures, health economic methods, and exploring the use of quality of life or capability measures to determine effectiveness from a more holistic perspective. Incorporation of generic preference-based measures into routine data collection would also allow local and international collection of utility data. This could in turn be used to develop cost per QALY estimates and utility changes facilitated by wheelchair interventions. Furthermore, this evidence would allow comparisons with other healthcare interventions and understanding of incremental cost-effectiveness. This would in turn encourage appropriate funding allocation and provision based on robust effectiveness evidence.

Child and parent proxy versions of validated HRQoL measures do exist, for instance the Health Utilities Index (HUI) [45]. However, their relevance for children with mobility impairments is still to be demonstrated. Some measures, such as the PedsQL, have additional bolt-ons for particular conditions (such as cerebral palsy) which take into account the condition-specific aspects of QoL [46], but these cannot be used to develop QALY estimates.

If wheelchair services in the UK and internationally were to adopt a single set of outcome measures a wealth of data could be generated which could be used to evaluate the holistic effectiveness of wheelchair interventions for children and young people. Furthermore, services could be structured around the needs of service users and parents.

Review limitations

In the spirit of transparency, it is worth considering some potential limitations. The original aim was to understand wheelchair interventions more generally, however due to the general focus in the literature on PWC interventions and participants with cerebral palsy, the findings may have greater relevance to these specific groups. There was also a lack of high quality evidence such as randomized controlled trials and economic evaluations.

CONCLUSIONS

The findings derived from international evidence are relevant across wheelchair services globally. Wheelchair services have an invaluable role in promoting equity for disabled people. If these services can address disabling barriers for children at a young age they may be able to facilitate more inclusion in education, employment and society more generally.

There are however important gaps in current knowledge, health economic methods and available outcome measures, which hinder further service development and research. Health economics has an important role in developing effective, efficient and equitable wheelchair services globally. The lack of economic evidence in this field highlights the lack of appropriate methods to measure cost-effectiveness. Establishing the cost-effectiveness of interventions is a priority to promote efficient services.

Collaboration between countries on future research would allow a wealth of data regarding intervention effectiveness and cost-effectiveness to be collected. The use of universal and validated outcome measures across countries would have a particular impact on the development of wheelchair services that promote social inclusion and independence.

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