

A Rapid Evidence synthesis of Outcomes and Care Utilisation following self-care support for children and adolescents with long term conditions (REfOCUS): Reducing care utilisation without compromising health outcomes.

Penny Bee, ^{1*} Rebecca Pedley ¹, Amber Rithalia ¹, Gerry Richardson, ² Steven Prymachuk, ¹ Susan Kirk, ¹ Peter Bower³

¹ Division of Nursing, Midwifery and Social Work, University of Manchester, Manchester Academic Health Science Centre, Manchester, UK.

² Centre for Health Economics, University of York, York, UK.

³NIHR School for Primary Care Research, University of Manchester, Manchester Academic Health Science Centre, Manchester, UK.

Important

A 'first look' scientific summary is created from the original author-supplied summary once the normal NIHR Journals Library peer and editorial review processes are complete. The summary has undergone full peer and editorial review as documented at NIHR Journals Library website and may undergo rewrite during the publication process. The order of authors was correct at editorial sign-off stage.

A final version (which has undergone a rigorous copy-edit and proofreading) will publish as part of a fuller account of the research in a forthcoming issue of the Health Services and Delivery Research journal.

Any queries about this 'first look' version of the scientific summary should be addressed to the NIHR Journals Library Editorial Office – journals.library@nihr.ac.uk

The research reported in this 'first look' scientific summary was funded by the HS&DR programme or one of its predecessor programmes (NIHR Service Delivery and Organisation programme, or Health Services Research programme) as project number 14/19/51. For more information visit <https://www.journalslibrary.nihr.ac.uk/programmes/hsdr/141951/#/>

The authors have been wholly responsible for all data collection, analysis and interpretation, and for writing up their work. The HS&DR editors have tried to ensure the accuracy of the authors' work and would like to thank the reviewers for their constructive comments however; they do not accept liability for damages or losses arising from material published in this scientific summary.

This 'first look' scientific summary presents independent research funded by the National Institute for Health Research (NIHR). The views and opinions expressed by authors in this publication are those of the authors and do not necessarily reflect those of the NHS, the NIHR, NETSCC, the HS&DR programme or the Department of Health. If there are verbatim quotations included in this publication the views and opinions expressed by the interviewees are those of the interviewees and do not necessarily reflect those of the authors, those of the NHS, the NIHR, NETSCC, the HS&DR programme or the Department of Health.

© Queen's Printer and Controller of HMSO 2017. This work was produced by Bee *et al.* under the terms of a commissioning contract issued by the Secretary of State for Health. This 'first look' scientific summary may be freely reproduced for the purposes of private research and study and extracts may be included in professional journals provided that suitable acknowledgement is made and the reproduction is not associated with any form of advertising. Applications for commercial reproduction should be addressed to: NIHR Journals Library, National Institute for Health Research, Evaluation, Trials and Studies Coordinating Centre, Alpha House, University of Southampton Science Park, Southampton SO16 7NS, UK.

Scientific Summary

Background

Commensurate with trends in the adult population, long term conditions in children and young people are increasing, and there is growing international emphasis on developing effective, efficient and person-centred models of service delivery to meet the needs of this patient group. Healthcare providers are faced with the growing need to deliver high quality services in a way that maximises available financial resources, without compromising care quality or health outcomes for children and young people. Self-care support interventions offer a potential solution to this problem and are intended to enhance the self-care capacities of children, young people and their families, while simultaneously reducing the fiscal burden facing contemporary healthcare systems.

Self-care can be defined as the actions that people take to maintain their physical and mental health; meet social and psychological needs; prevent illness or accidents and maintain their health and wellbeing. Self-care *support* refers to the role played by healthcare professionals (or other self-care support agents such as teachers or peers), in supporting the individual and/or their families to take control of a health condition through developing their confidence, knowledge and skills, and their psychological and social resources.

An implicit assumption underlying self-care support is that it can help people to avoid unnecessary crises and prevent more extensive health services utilisation by managing patients' problems more effectively. Children diagnosed with long term conditions face a lifetime of symptom management, and the extent to which they and their families negotiate this in childhood is likely to influence their longer term health outcomes, life chances and subsequent patterns of health service utilisation. Providing optimal, evidence-based support for self-care thus has the potential to make significant and sustained contributions to NHS efficiency, as well as improving quality of care and health outcomes.

Objectives

To determine which models of self-care support for long term condition management are associated with significant reductions in health services utilisation and cost, without compromising quality of life or health status outcomes for children and young people.

Methods

We conducted a systematic review with meta-analysis. We defined self-care support as 'any intervention primarily designed to develop the abilities of children and young people (and/or their adult carers) to undertake management of their long term health condition through education, training and support to develop their knowledge, skills or psychological and social resources.' Our review inclusion criteria were as follows:

- **Population:** children and young people aged 0 to < 18 years with a long term physical or mental health condition.
- **Intervention:** Self-care support delivered in a health, social care, educational or community setting.
- **Comparator:** Usual care, including more intensive usual care e.g. clinic or inpatient management.
- **Outcomes:** Quantitative measures of generic or health related quality of life or disease-specific symptom measures or events *and* health service utilisation or costs.
- **Design:** Randomised Trials, non-randomised trials, Controlled before and after studies, interrupted time series designs.

To identify relevant literature we searched multiple electronic databases: MEDLINE, EMBASE, PsycINFO, CINAHL, ISI Web of Science (including SSCI and SCIEXPANDED), NHS EED, Cochrane Library (including CDSR, DARE and CENTRAL), Health Technology Assessment database, PEDE and IDEAS. All databases were searched from inception to March 2015. No language restrictions were applied. Additional search strategies included

© Queen's Printer and Controller of HMSO 2017. This work was produced by Bee *et al.* under the terms of a commissioning contract issued by the Secretary of State for Health. This 'first look' scientific summary may be freely reproduced for the purposes of private research and study and extracts may be included in professional journals provided that suitable acknowledgement is made and the reproduction is not associated with any form of advertising. Applications for commercial reproduction should be addressed to: NIHR Journals Library, National Institute for Health Research, Evaluation, Trials and Studies Coordinating Centre, Alpha House, University of Southampton Science Park, Southampton SO16 7NS, UK.

scanning the bibliographies of all relevant retrieved articles, targeted author searches and forward citation searching.

Data were extracted on populations, interventions, study quality, and outcomes. We extracted data that allowed us to report a measure of the magnitude of effects (a standardised 'effect size') for both health outcomes and costs. We conducted meta-analyses and presented the results of the included studies according to a permutation plot, simultaneously plotting the effect of interventions on service utilisation and health. Each plot gives a visual impression of the distribution of studies across the cost-effectiveness plane, distinguishing between studies that reduce costs without compromising outcomes from those that reduce costs but also compromise outcomes, or those that compromise both outcomes and costs.

We analyzed data for included studies as a whole, and then conducted meaningful subgroup analyses for level of evidence quality (defined as the adequacy of allocation concealment), age of the children and young people, type of long term condition and the setting and type of self-care support intervention that was evaluated (i.e. intervention target, format, delivery method, and intensity).

Results

We screened 36493 unique records for eligibility; 97 studies reporting on 114 interventions were included in our review. Thirty seven trials (38% of all included studies) were judged to be high quality (low risk of bias) on the basis of adequate randomization and allocation concealment. Fourteen studies (14%) were conducted in the UK.

The vast majority of included studies recruited children and young people with asthma (n=66, 68%) or long term mental health conditions (n=18, 19%). Fewer studies included children with diabetes (n=6), other physical health conditions (n=2) and behavioural difficulties (n=5). The mean age of the children and young people participating in the primary studies was 10.12 (SD 3.9) years. Of the interventions, 4% were categorised as

pure self-care (i.e. delivered through a health technology without any additional support), 23% as facilitated self-care (<2 hours/4 sessions of support), 65% as intensively facilitated self-care (>2 hours/4 sessions support) and 8% as 'case management' (>2 hours support including input from a multidisciplinary team).

The majority of self-care support interventions targeted adult caregivers, either together or in parallel with children and young people. These interventions were most typically delivered face-to-face to individuals, or individual families, in either an outpatient setting or a patient's home.

A moderately sized evidence base enabled meaningful assessments of the effect of self-care support interventions on children's and young people's health and quality of life (77 comparisons), hospital admissions (65 comparisons) and emergency department visits (57 comparisons). Other forms of health service use (e.g. primary care visits) were inconsistently reported and were not amenable to meta-analysis. There was a comparative lack of data demonstrating the effects of self-care support on total health service costs (10 comparisons) and variability across studies reporting total cost outcomes was high.

Self-care support interventions have significant but minimal benefits for children's and young people's quality of life (ES 0.17, -0.23 to -0.11), but lack clear benefits for hospital admissions (ES -0.05, -0.12 to 0.03) and total healthcare costs (ES -0.11, -0.47 to 0.25). Minimal reductions in emergency department use were observed (ES -0.11, -0.17 to -0.04). Data on quality of life outcomes suggested the possibility of small study bias. Sensitivity analyses that restricted evidence to high quality trials confirmed that the findings were robust. Sub-group analyses revealed statistically significant, minimal reductions in emergency use for children <13 years old (ES -0.10, -0.17 to -0.04), children and young people with asthma (ES -0.12, -0.18 to -0.06), and children and young people receiving > 2 hours/4 sessions of support (ES -0.10, -0.17 to -0.03). The different effect sizes observed in these sub-group analyses will in part reflect differences in the number of studies available and the precision of pooled effects; additional evidence is required to confirm or reject these hypotheses.

Preliminary analyses suggest that the effects of self-care support on children's and young people's quality of life and emergency department visits may be optimised by interventions that include the child or young person, and deliver at least some of their content to an individual or individual family. Group-based delivery may be more advantageous in reducing hospital admissions, although effects are likely to remain small.

Self-care support interventions for children and young people can vary considerably in the extent to which they target different service utilisation behaviours and it is possible that this influence is meaningful. It is plausible for instance, that while written action plans to control asthma exacerbations may play a direct role in reducing emergency department visits, self-care support for mental health may be focused on longer term recovery and service user empowerment.

Preliminary data in our permutation plots suggests that self-care support for asthma is capable of reducing some aspects of health utilisation for children and young people, but high variability in patient outcomes means that compromises in health status cannot definitively be ruled out. Self-care support interventions that reduce health utilisation for children and young people with mental health conditions, may be less likely to compromise patient outcomes, but limited data, and pooling across different conditions, mean that these results must be treated with caution. Lack of data prevented permutation plots being generated for other long term conditions.

Conclusions

Self-care support for children and young people is advocated as a key method of increasing service efficiency, but there remains some uncertainty regarding the scale of the contribution that can be made. Current evidence suggests that self-care support interventions will have positive but minimal effects on children's and young people's quality of life, but may have limited impact on health utilisation and costs. Self-care support for children and young people is associated with significant but small reductions in emergency department use, particularly in relation to asthma. Models of self-care support that reduce utilisation do not

routinely compromise patient outcomes. However, the effects are highly variable and compromises in children's and young people's quality of life cannot be definitely ruled out.

New primary research is urgently needed to ascertain the effects of self-care support across a wider range of long term conditions, and to explore whether, and which, models of self-care support can achieve more powerful, consistent effects on health service utilisation.

Future studies should adopt clear and consistent standards of data reporting including comprehensive reporting of patient outcomes, utilisation and costs. New research should adopt innovative methods of patient recruitment to maximise intervention reach and consider the feasibility of longer term follow-up to explore potential differences in the shorter and longer term effects of self-care support for children and young people.

PROSPERO registration

CRD42014015452

Funding

Funding for this study was provided by the Health Services and Delivery Research programme of the National Institute for Health Research.