

Title

Improving participation outcomes and interventions in neurodisability: co-designing future research

Authorship details

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Short title: Improving participation interventions and outcomes.

Financial disclosure: None.

Conflicts of interest: None.

Abbreviations: UK=United Kingdom; NHS=National Health Service; CFCS=Communication Function Classification System; GMFCS=Gross Motor Function Classification System

Contributors' statement:

JMcA and NK conceived the project idea; developed and led the funding application; and managed the overall project. JMcA co-facilitated the service user involvement with children and young people; led and carried out the development of the search strategy; and led the writing of this manuscript. RB, CD, MQ and JM a made a substantial contribution to the study methodology, and all contributed to discussions about the intervention and the subsequent study design. RB also screened the abstracts for inclusion, and CD and JM facilitated the service user groups together with JMcA and NK. NK led the development of the inclusion/exclusion criteria for the search; screened all titles and papers for inclusion; led and carried out data extraction and summary. All authors reviewed and revised the manuscript for substantial content, and approved the final manuscript as submitted.

Word count: 2,013

Key words: Participation, self care, patient and public involvement, user involvement

Acknowledgements

We sincerely thank the service users and providers who took part in the project, and Dr Lindsay Pennington and Ms Jane Giles who provided speech and language therapy views to the ranking exercise (step 2). The project was funded by a Public and Patient Engagement/Involvement grant from the Faculty of Medical Sciences, Newcastle University. JMcA is funded by Health Education England/National Institute for Health Research (NIHR) under the Clinical Doctoral Research Fellowship Scheme (CDRF 2014-05-043). RB, CD, MQ, NK are funded by their respective institutes (see Affiliations). JM was funded by Research Capability Funding from the Newcastle upon Tyne NHS Hospitals Foundation Trust. The views expressed are those of the authors and not necessarily those of the National Health Service, the NIHR or the Department of Health.

Abstract

There is an urgent, agreed need to improve participation outcomes and interventions for children and young people with neurodisability. We worked together with service

users and providers to design research into participation outcomes and interventions in neurodisability. We built on existing evidence about participation outcomes and interventions, and the WHO International Classification of Functioning, Disability and Health. We: (1) specified seven participation outcome categories for measurement; (2) prioritised these for improvement: self-care, friends and social, and physical activity ranked the highest; (3) identified eleven potential intervention categories for targeting the top priority, self-care, through eight hypothesised change mechanisms; and agreed for the interventions to be delivered as a 'Menu of Interventions' for personalised self-care support; and (4) designed a before-and-after mixed methods feasibility study to evaluate the Menu with children and young people (0-12 years), and their parents and therapists.

Introduction

Participation in everyday life situations is a fundamental health outcome for all children, and a key healthcare outcome for children and young people with neurodisability.(Health 2013, Children and Young People's Health Outcomes Forum 2012, Allard *et al.* 2014, World Health Organization 2007) However, implementation of effective, participation-focused services is hindered by the lack of routine outcome measures of participation(Morris *et al.* 2014, Kolehmainen *et al.* under review) and scarcity of replicable participation interventions with evidence of effectiveness.(Novak *et al.* 2013, Kolehmainen N In preparation, Raghavendra 2013, Adair *et al.* 2015) To address these challenges, there is an urgent need for further research into participation outcomes and interventions, (Novak *et al.* 2013, CountMeIn! Network 2015) especially into occupational, physical, and speech and language therapy outcomes and interventions.(Morris *et al.* 2015)

Research into participation outcomes and interventions is limited and notoriously challenging.(Whiteneck and Dijkers 2009, Raghavendra 2013, CountMeIn! Network 2015, Adair *et al.* 2015) Participation as a concept covers a range of domains and there is little consensus over how it should be operationalised for measurement.(Whiteneck and Dijkers 2009) Similarly, participation interventions are often complex (multifaceted), with their active ingredients difficult to define.

Our aim was to work with service users and providers to explore if, by working together, we could design feasible and practicable research into participation outcomes and interventions for children and young people with neurodisability. The focus was on outcomes potentially modifiable by, and interventions possibly provided by, occupational, physical, or speech and language therapists.

Our aims were to: (1) specify participation outcomes for measurement in routine practice; (2) prioritise one participation domain for further research; (3) identify potential intervention(s) for targeting that outcome, specify the hypothesised change mechanisms, and agree ways to deliver the intervention(s); and (4) design a study to evaluate the intervention(s). We also sought to provide an exemplar of service user involvement in designing research into participation outcomes and interventions.

Methods

This was a service user involvement project using the principles of co-design, (McLaughlin 2015, The VIPER Project 2012, Kirby 2004) supplemented with a rapid review of published literature. The main outcome was participation, defined using the World Health Organization International Classification of Functioning, Disability and Health (ICF) (World Health Organization 2007) as 'involvement in life situations'. To build on existing research, (Morris *et al.* 2014) we started with seven participation domains: community leisure, friends and social activities, physical activities and sports, self-care, sleep, transition to independence and independent living, and transition to employment. These domains were selected as potentially central to the three therapies (above), and broadly related to the research team's expertise, thus could plausibly be advanced by the team. We adopted a view that participation outcomes may reflect both 'doing' and 'being', and that together they may form wider structures or pathways (e.g. 'doing sports' can contribute to 'being with friends'). (Kolehmainen and Johnston under review) We were open to changing this assumption over the project, in line with our broader principle of pragmatism. (Cherryholmes 1992)

The service user partners self-selected from two NHS organisations by responding to open invitations sent to them. Service providers were selected to represent the

three therapies, and invited to become involved. The partners involved six young people with neurodisability (referred to below as 'the young people'), and a young adult with neurodisability, four parents of young children with various neurodisabilities, five therapists and a senior therapy manager (referred to as 'the adults'). The young people ranged in age from 11 to 18 years, attended mainstream schools and colleges with varying levels of support, and experienced a range of communication (CFCS Levels I-III)(Hidecker *et al.* 2011) and mobility (GMFCS Levels III-V)(Palisano *et al.* 2008) limitations. The young adult had recently left the service through supported transition to adult services. Ethics approval was not required as the project was a service user and provider collaboration, not research with subjects. The aims were met through four steps:

1. Specify participation outcomes

With the young people, JMcA and CD facilitated discussions about experiences and opinions of the different participation outcome categories by using visual characters and prompts, scenarios, and vignettes. These have been previously shown to enable young people, including young people with communication limitations, to direct the agenda, engage with concepts, and voice opinions.(Morris *et al.* 2014, Kolehmainen N 2015, Fargas-Malet *et al.* 2010) With the adults, NK and JM provided visual summaries of the participation outcome categories and related key words from existing qualitative evidence.(Morris *et al.* 2014) The adults were encouraged to add further keywords to each outcome as they considered important, and each outcome was then further jointly discussed. The views expressed across the young people and the adults were collated and tabulated next to one another. The contents were shared back to the adult partners for further comments, and formed the basis for further exploration with the young people. The research team did not contribute views to this step, but reflected on the views provided by the partners.

2. Prioritise outcomes for further research

Based on the discussions above, the young people, the adults, and the researchers individually ranked the participation outcomes in the order of importance for improvement. Two speech and language therapists were also asked to provide

rankings to improve representation (see acknowledgements). NK collated the responses and calculated one overall ranking for service users and another for service providers and researchers. The rankings were fed back to the groups, who confirmed the prioritised outcome.

3. Identify intervention(s), specify change mechanisms, and agree delivery

We conducted a rapid scoping review of published literature on interventions for targeting the prioritised outcome (for details, see Supplementary File). The focus was on identifying systematic reviews and substantial formal evaluations of interventions for children (0-18 years) with or without disabilities for the prioritised outcome. Papers were screened against PICOT inclusion and exclusion criteria, and abstracts that clearly met the criteria AND offered positive evidence of potential interventions for the prioritised outcome were accessed for a full review. We extracted key messages about any promising interventions from the included papers, and summarised these and any change mechanisms in visual presentations (see descriptions in step 1). These visual presentations were shared with the adults at further meetings; whereas the young people focused on further developing the materials they had generated about interventions, which were in turn integrated with the visual presentations.

Throughout the process, the young people and the adults reflected on the feasibility, acceptability and desirability of the different interventions. They shared ideas about who should deliver the interventions, to whom, where and how frequently; and which age groups should be targeted. They reflected on their own experiences and preferences of different interventions, and on intervention features (ingredients and delivery) that influenced success. The discussions also covered hypothesised change mechanisms.

4. Design a study to evaluate the intervention

Based on the ideas and views generated over the two rounds of meetings (see above), and existing evidence (see above), the researchers drafted a research question, aims, and an overall design to advance the intervention(s) using the Medical Research Council guidance for complex interventions. (Craig *et al.* 2008, Medical Research Council 2000) Also, specific design questions were identified and

presented to the young people and adults, e.g. what the population criteria should be (age, disability, healthcare professional type), what outcomes should be measured and from whom, and what should the control condition be.

Results

Five of the seven outcome categories were specified, and ranked in order of priority (Table 1). In addition, transition to independence was moved to within self-care, and transition to independent living and employment were considered to be currently supported outside children's health services in the main, thus not included further. Self-care, friends and social, and physical activity and sports were ranked first, second, and third highest priorities for improvement, respectively. Self-care was further operationalised for measurement (Table 2), using the dimensions described as important by the service users.

For the rapid review of potential self-care interventions, we screened 9,190 titles, which were reduced to 181 abstracts, and then 38 full-texts (for full details, see Supplementary File). A final 13 full-texts were included; all but one (Law *et al.* 2011) had sample sizes <50 and were judged to have substantial methodological limitations (e.g. use of before-after designs). A further 9 papers on constraint-induced movement therapy (CIMT) and one on powered mobility would have been included, but were replaced with more up-to-date systematic reviews, (Sakzewski *et al.* 2014, Livingstone and Field 2014) resulting in a total of 15 included full-texts.

No interventions with conclusive evidence of effectiveness for improving self-care were identified. CIMT, (Sakzewski *et al.* 2014) goal-setting, (Donlau *et al.* 2013, Hwang *et al.* 2013, Löwing *et al.* 2009, Sorsdahl *et al.* 2010) and powered mobility (Livingstone and Field 2014) were consistently promising interventions. Few papers explicitly stated proposed change mechanisms, but hypothesised mechanisms (described as 'barriers' and 'facilitators') emerged over the discussions with the young people and the adults (Table 1). In addition, the young people and adults proposed interventions.

Across the young people and the adults the single most consistent message was that self-care interventions needed to be tailored to the preferences and circumstances of the individual child, young person and family. From this, a proposal

was made that the full list of interventions would be offered, and individual children, young people and families enabled to choose those that fitted their needs, preferences and circumstances. Other key points for intervention delivery were that any intervention should: always be considered in the context of family goals, proactively facilitate joint working across agencies, support the whole family, and seek to make use of everyday developmental opportunities. Ultimately, the intervention was agreed to be a 'Menu of Interventions', designed to support families and therapist(s) to make joint decisions about what factors ('mechanisms') to target and using which interventions.

The Menu (Figure 1) consists of eleven intervention categories developed together with the service users and from the rapid review of existing literature: set individual goals with support, modelling by similar people, monitor and compare against the target/standard, practice with feedback, grade tasks, problem solving, adapt tasks/environment, provide equipment, demonstrate and train, provide information, direct to community-based public health interventions. These intervention categories target eight mechanisms emerging from service-user discussions: children and young person (CYP) and parent motivation and determination, CYP confidence to undertake self-care, parent confidence in CYP undertaking self-care and to support CYP, CYP emotions, parent emotions, family habits and routines, CYP and parent knowledge and skills related to specific tasks, CYP mobility, and knowledge (especially parent). The Menu can be used by multiple agencies to facilitate joint working and be held by the family to support overall family control.

In light of the lack of conclusive evidence of effectiveness for any of the specific interventions included in the Menu, and lack of evidence about which of these might be most likely to be taken up or how they are currently delivered, it was agreed that an exploratory feasibility study was needed. The agreed study design is summarised in Table 2, including specification of the feasibility and acceptability outcomes.

Discussion

We worked together to identify self-care as the key priority outcome; develop a multifaceted Menu of Interventions for personalised self-care support; specify its eleven ingredients and eight mechanisms of change; and design a study to investigate it. The project partners were unlikely to represent the full range of views

in the neurodisability community. However, our intention was not to conduct a representative consensus study, but to work with service users as part of a team to design an intervention and a related research study.

We also provided an exemplar of service user involvement in designing research into participation outcomes and interventions. Involving service users is thought to have a positive impact on the quality, relevance, and acceptability of health research.(Brett *et al.* 2014, Patterson *et al.* 2014, Morris *et al.* 2015) In our project this impact was particularly evident in the rating of self-care as the highest priority. Self-care has long been considered the stock-in-trade for many therapies; however, the rapid review in the present project and previous reviews(Novak *et al.* 2013, Kolehmainen N In preparation, Adair *et al.* 2015) demonstrate that it has received limited research attention. In addition, service users' definition of self-care emphasised dimensions related to autonomy and dignity (e.g. making decisions, being listened to), which in current practice are often overlooked in favour of safety and access. The next step is to investigate the intervention – using the study we designed.

Key messages

- The paper presents an exemplar of service users, service providers and researchers co-designing research on participation outcomes and interventions in neurodisability
- The team adopted a cumulative approach to improving participation outcomes and interventions, building on existing evidence
- Service users prioritised participation in self-care, an outcome that has received limited research attention; and highlighted important dimensions of self-care that are under-explored in practice

Table 1. Summary of the important aspects of the five participation outcomes, and of the related proposed facilitators and barriers, and interventions, and priorities for improvement

	Adults (service users & providers)	Young people
<p>Self-care</p> <p><i>Priority #1</i></p>	<p>Important dimensions:</p> <p>Dignity – being listened to, and having one’s values and opinions respected</p> <p>Building the skills over time, learning, development</p> <p>Increasing independence across basic activities (dressing, personal hygiene, etc), and more complex tasks (managing money and time, snack preparation and cooking, laundry, cleaning and tidying, clothes care)</p> <p>Accessing what is needed</p> <p>Potential facilitators/barriers:</p> <p>Physical skills and mobility to do tasks</p> <p>Knowledge and skills specific to the tasks</p> <p>Potential interventions:</p> <p>Learning opportunities with appropriate support</p> <p>Individual goal-setting with appropriate support</p> <p>Equipment, adaptations</p> <p>Joint working</p>	<p>Important dimensions:</p> <p>Making decisions</p> <p>Instructing other people in tasks (e.g. instructing carer)</p> <p>Positive outcomes/goals: smelling nice</p> <p>Potential facilitators/barriers:</p> <p>Motivation, determination</p> <p>Knowledge and skills (incl. doing things safely)</p> <p>Children and young people’s (CYP’s) confidence</p> <p>Other people’s confidence the CYP can do it</p> <p>Physical skills and mobility</p> <p>Habit of accepting help</p> <p>Potential interventions:</p> <p>Relaxation techniques (e.g. music)</p> <p>Adaptations, setting up the space in the right way</p> <p>Carers’ behaviours: listening to the CYP, respecting the CYP, accepting CYP choices, having a laugh</p> <p>Equipment</p> <p>Training (for carers, parents)</p>
<p>Friends and social activities</p> <p><i>Priority #2</i></p>	<p>Important dimensions:</p> <p>Relaxing and enjoying spending time together</p> <p>Relating to people appropriately</p> <p>Doing things together</p> <p>Potential facilitators/barriers:</p> <p>“Self-reliance”: independence, ability to take care of oneself so</p>	<p>Important dimensions:</p> <p>Have fun, feel included, go where other people go</p> <p>Potential facilitators/barriers:</p> <p>Being confident, but not too confident, to make friends</p>

	<p>as not to put 'caring expectations' or 'be a burden/hindrance' on friends – also self-care (independence in it, see below) Knowledge of current topics, 'having the right chat'</p> <p>Potential interventions:</p> <p>Transport (to get to places) Funding Equipment Wheelchairs, powered mobility (to get to places, to keep up) Peer support and relationships (built over time) Providing information (especially learning from other disabled people) Social media</p>	<p>If you're nervous, you may need to hang back Goals, motivation of the person</p> <p>Potential interventions:</p> <p>Opportunities (it might not work the first time making friends, but they may open up to you after several meetings; most of my friends I met through sport) Access to like-minded people (It's easier to become friends if the other person has similar physical abilities, because they understand what you can and can't do. It's easier because you can both access the same environments, do similar activities; important to have people you get along with well) Advice, guidance to the person (You might need help avoiding getting in with the wrong group, the naughty kids; help to use social media) Social support and role modelling (e.g. by parents) Information to other people (Society needs more information on how to make friends with someone who is a wheelchair user, and needs to know how to treat wheelchair users) Transport/practical help (to get to where you need to go to see your friends, to plan activities)</p>
<p>Physical activity (PA), sports & active lifestyle</p> <p><i>Priority #3</i></p>	<p>Important dimensions:</p> <p>Doing what one enjoys – focus on here-and-now not just long-term health benefits Positive outcomes/goals related to sports/PA: socialisation, belong to a group, fun and enjoyment, staying healthy, being challenges and achieving</p> <p>Potential facilitators/barriers:</p> <p>Motivation</p> <p>Potential interventions:</p> <p>Adapt activities Set goals around the positive outcomes (here-and-now, not just long term health benefits)</p>	<p>Important dimensions:</p> <p>Going to clubs, playing sports Confidence in sports Positive outcomes/goals related to sports/PA: feeling physically good, achieving things, having something to do, happiness, fun, making friends</p> <p>Potential interventions:</p> <p>Transport Finance Social support and encouragement (e.g. from family, friends)</p>

		<p>Information about what's available (e.g. from others with disability, from sports organisations)</p> <p>Opportunities for right kinds of sports</p>
<p>Community leisure</p> <p><i>Priority #4</i></p>	<p>Important dimensions:</p> <p>Accessing and using shared environments, e.g. libraries, swimming pools, cinemas, parks, beaches, etc</p> <p>Going to community-based clubs, hobbies, church, etc</p> <p>Knowing people in the community, outside immediate family and friends, and interacting and communicating with them</p> <p>Moving around in the community</p> <p>Making choices</p> <p>Complying with wider community rules and regulations</p> <p>Potential facilitators/barriers:</p> <p>Staff/community confidence</p> <p>Knowledge of available options</p> <p>Having confidence to go out and join in</p> <p>Feeling welcome and accepted</p> <p>Potential interventions:</p> <p>Adapting activities</p> <p>Providing information about what leisure activities, events, places are available</p> <p>Providing information about what support is available (transportation, funding, facilities, social support)</p> <p>Support for YP and parents (peer groups, professional support)</p> <p>Transport</p> <p>Community makes the person feel welcome</p>	<p>Important dimensions:</p> <p>Range of activities: sports, music, cinema, seeing friends, going to parties, going shopping, going to restaurants, etc</p> <p>- important to follow your own goals and priorities</p> <p>Safety, so that young people and their parents are confident</p> <p>Being spontaneous, and doing stuff when I want, not just when there is a special session for people with disabilities</p> <p>Potential interventions:</p> <p>Transportation, parking</p> <p>Social support – knowing the people/staff, the people there can help and advice if needed, support from carers</p> <p>Facilities, e.g. for self-care</p> <p>Information about opportunities, funding, access</p> <p>Wheelchair to be able to self-propel</p>
<p>Sleep</p> <p><i>Priority #5</i></p>	<p>Important dimensions:</p> <p>Falling asleep, staying asleep, healthy sleep patterns</p> <p>Quality of sleep, being comfortable</p>	<p>Important dimensions:</p> <p>Being comfortable</p> <p>Falling asleep</p> <p>Potential facilitators/barriers:</p> <p>Being stiff</p> <p>Having enough space in the bed</p>

	<p>Potential interventions:</p> <p>Sleep routines Medical management Equipment (sleep systems, sensory, alarms, postural management) Health visiting/school health interventions to support good sleep-related parenting Preventative/early sleep interventions</p>	<p>Equipment (e.g. special mattresses, pillows, wider bed)</p> <p>Potential interventions:</p> <p>Massage Medication Bed time routines (e.g. going to bed at a reasonable hour)</p>
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Figure 1. The draft Menu of Interventions, with potential factors to target (in blue-white boxes) and intervention techniques to target them with (green boxes)

MENU OF INTERVENTIONS

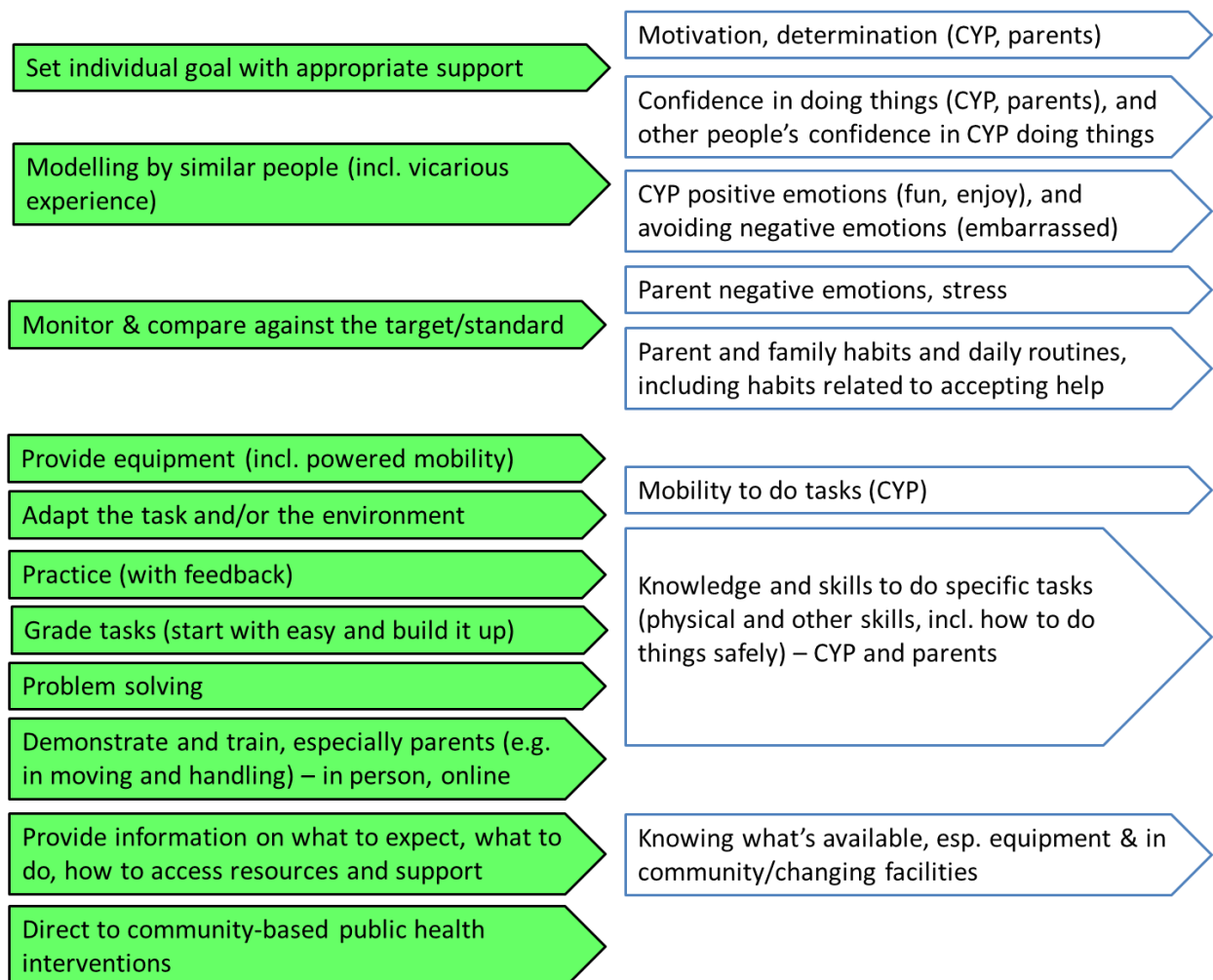


Table 2. Summary of the agreed intervention design

Title: Personalised self-care support for children and young people with neurodisability and their parents: a mixed methods feasibility study of the Menu of Interventions

RQ: How will a personalised Menu of Interventions for self-care support be taken up and experienced by children and young people (CYP) with neurodisability, their parents, and healthcare professionals; and how will these compare to current self-care support?

Aims:

To investigate the feasibility, acceptability, and potential costs and benefits of the Menu of Interventions.

To investigate if CYP, parent or therapist characteristics relate to uptake or acceptability.

To describe current self-care support in terms of feasibility, acceptability, and potential costs and benefits – and descriptively compare it to the Menu of Interventions.

Design: A mixed methods (QUALITATIVE+quantitative) feasibility study with six NHS Trusts and their service users. The main health outcome will be self-care, defined as a combination of (i) “making decisions about things that affect me, including having my values and opinions respected, and being listened to” and (ii) “developing and learning skills for looking after myself, ranging from basic tasks (e.g. dressing, personal hygiene, eating) to complex sets of tasks (e.g. managing money and time, snack preparation and cooking, laundry and clothes care, cleaning, accessing resources, and managing and directing care providers)”.

Feasibility, acceptability, and potential costs and benefits to be evaluated as: uptake and adherence to the Menu (e.g. numbers of therapists, parents and CYP; their characteristics; and aspects of the Menu used), satisfaction and perceived impact – especially in relation to expectations, challenges to uptake, coverage and what needs added to the Menu, frequency of contact following the use of the Menu, and time and emotional costs to stakeholders.

The study population will be occupational therapists, physiotherapists and/or speech and language therapists in community/outpatient healthcare settings providing care for CYP with neurodisability from birth until after the transition to secondary school (estimated age 0-13 years); the CYP seen by these therapists; and the CYPs’ parents. Neurodisability will be understood in line with the UK consensus definition.(Morris *et al.* 2013)

The intervention: The Menu of Interventions is a prototype for an interactive interface to support CYP, parents and therapists to work together to identify: (i) what factors to target for change for that particular CYP and family, and (ii) what interventions they wish to use for this. The Menu consists of eight intervention categories developed together with service users and from a review of existing literature: practice and feedback, adapt tasks, provide equipment, adapt environment, demonstrate, provide information, set individual goals with support, direct to community-based public health interventions. These intervention categories target ten factors: parent knowledge, CYP confidence to undertake self-care activities, parent confidence to support CYP in learning skills, family habits and routines, CYP motivation and determination, and parent motivation and determination, CYP physical skills and mobility, CYP task-specific skills, parent task-specific skills, and parent emotions. The Menu is used with CYP and parents who indicate they have goals related to self-care.

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