CHESS: Investigating the clinical and cost-effectiveness of CHildren's Early Self-care Support in children with neurodisability: the cluster randomised controlled trial

PROTOCOL

A UK Collaborative Trial funded by the National Institute for Health and Care Research (NIHR) Health Technology Assessment (HTA) Programme (project number NIHR156487)

This Protocol has regard for the HRA guidance and order of content

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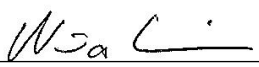
Signatures

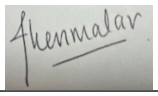
The undersigned confirm that the following protocol has been agreed and accepted and that the Chief Investigator agrees to conduct the trial in compliance with the approved protocol, GCP guidelines, the Sponsor's (and any other relevant) SOPs, and other regulatory requirements as amended.

I agree to ensure that the confidential information contained in this document will not be used for any other purpose other than the evaluation or conduct of the clinical investigation without the prior written consent of the Sponsor.

I also confirm that I will make the findings of the trial publicly available through publication or other dissemination tools without any unnecessary delay and that an honest accurate and transparent account of the trial will be given; and that any discrepancies and serious breaches of GCP from the trial as planned in this protocol will be explained.

Niina Kolehmainen (Chief Investigator): 

Date: 10 February 2025

Thenmalar Vadiveloo(Statistician): 

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VERSION HISTORY

Amendment no.	Protocol version no.	Description of changes (*incl. author(s) of changes*)	Date of document
	Version 1	New Document	31 Jul 2024
	Version 2	*Changes following comments from REC.* *Section 10.3.2 - Clarification of reminder process for letters or emails* *Section 16 - Guidance for clinicians regarding concern for the well-being of participants*	15 Oct 2024
	Version 3	Section 12.1,15.1, 16 – Update to safety reporting guidelines 15.1, 15.3, 22.3 – Update to test-retest SWAT to move testing to 12 months 21.1 – Addition of sub-analysis of CHU9D in children over 2 years 24.4 – clarification on the storage of participant questionnaire data using the Pearson Clinical Q-Global server 9.3 and 22.2 – Additional information on the targeted sampling SWAT	10 Feb 2025

Contents

1. Trial summary

TRIAL TITLE	Investigating the clinical and cost-effectiveness of CHildren's Early Self-care Support in children with neurodisability: the CHESS cluster randomised controlled trial
Short title	CHESS: CHildren's Early Self-care Support
Rationale	Caring for oneself ('self-care') is essential to survival. It encompasses independence (e.g. learning to feed oneself) and involvement in self-care situations (e.g. making choices and coping with mealtimes with others). It is estimated 3-4% of children in the UK have neurodisability, with self-care problems that are both significant and common. While many can achieve self-care levels close to their typically developing peers this requires significant parent and therapy support. There is currently little evidence on effective self-care interventions, no national guidelines, and no cost-effectiveness evidence for commissioners. Parent, young person, and multidisciplinary professional consensus is that additional evidence is urgently needed. The aim is to determine the clinical and cost-effectiveness of CHESS in young children with neurodisability compared to usual care. The objectives are to: 1) Determine the clinical and cost-effectiveness of CHESS compared with usual care, on self-care skills and involvement for young children with neurodisability, measured at 6 and 12 months after cluster randomisation. 2) Estimate the relative efficiency of CHESS compared with usual care, in terms of self-care skills and involvement in young children with neurodisability. 3) Conduct an embedded evaluation of recruitment, intervention acceptability, and implementation to support trial delivery.
Trial design	A two-arm pragmatic cluster randomised controlled trial across 40 clusters with embedded economic and process evaluations.
Eligibility criteria	**Cluster inclusion criteria:** NHS children's therapy services/teams, providing i. paediatric community and/or outpatient occupational therapy, physiotherapy, and/or speech and language therapy; ii. for eligible children (age from 12 months to 4 years 6 months, with neurodisability); iii. in England, Wales, Scotland or Northern Ireland. **Cluster exclusion criteria:** services and therapists providing hospital-based care only. Tertiary services offering diagnostic-only or consultation-only care, and not therapy to child's life at home, will be excluded. Education-based services with no, or minimal, input to the child's family and the child at home, will be excluded. **Patient inclusion criteria:** Any child with neurodisability; age at least 12 months and not older than 4 years 6 months on the date of sampling the child; seen by at least one paediatric community/outpatient therapy service: where self-care support is indicated; where the family and therapist agree on a need for self-care intervention; who has been randomly sampled for data collection from the participating therapists' caseloads; AND whose parent is willing to consent to data collection and comply with study procedures. No restriction is

	placed on child mobility, cognitive or communication capacities and due to the nature of caseloads and random sampling of children we anticipate the same to include a mix of children across capacities. **Patient exclusion criteria:** Children with: no neurodisability; with only sensory impairment (e.g. visual, hearing); degenerative condition(s); no clear self-care problems or goals; receiving one-off advice and discharge only; hospitalised or in end-of-life care; and/or a sibling already participating in the CHESS trial.	
Sampling	Clusters are purposively sampled for representation of populations with higher levels of socioeconomic disadvantage and from minority ethnic backgrounds. Children are randomly sampled from eligible caseload lists.	
Interventions	CHESS (CHildren's Early Self-care Support), a manualised, multicomponent behaviour change intervention compared to usual children's community NHS therapy (occupational therapy, physiotherapy, speech and language therapy) for self-care limitations.	
Randomisation and blinding	CHESS is a cluster trial, at a level of a whole NHS organisation, with participants being young children and their parents. There is no individual child or parent randomisation, and no parent consent will be sought for randomisation. All families within an NHS organisation, whether or not they participate in the trial data collection, will receive the same treatment based on the randomisation of that cluster. Following sampling and case identification, clusters will be randomised to treatment or control group using covariate constrained randomisation. This approach minimises imbalance on cluster level covariates, which is a potential risk in cluster randomised trials with fewer clusters. We will use Carter and Hood's algorithm to optimise balance on the following: geography (urban/rural); cluster size (number of therapists); and child population sociodemographics (namely significantly higher/lower levels of poverty or minoritised ethnic communities compared to UK average). Covariate constrained randomisation is the recommended allocation procedure when all clusters can be recruited before allocation and measurable prognostic cluster-level covariates are known. Participants will be blinded at the therapist level, to reduce risk of trial participants being given enhanced treatment. Local cluster PIs will not be blinded and will assist in participant sampling.	
Planned sample size	The unit of randomisation is at the cluster level, with a target sample size of 40 clusters, each cluster recruiting 24 children (a total sample size of 960 children).	
Duration of trial	48 months	
	Objectives	Outcome measures
Primary	Determine the clinical effectiveness of CHESS compared with usual care, on self-care skills and involvement for young children with neurodisability, measured at 12 months after cluster randomisation.	Primary outcome: parent-reported Pediatric Evaluation of Disability Computer Adaptive Testing (PEDI-CAT) Activities Of Daily Living module at 12 months after start of intervention.

Secondary	Determine the clinical and cost-effectiveness of CHESS compared with usual care, on self-care skills and involvement for young children with neurodisability, measured at 6 and 12 months after cluster randomisation.	Secondary outcomes: Self-care involvement within dynamic child-caregiver interactions at baseline, 6 and 12 months, with focus on the child's agency, comfort, stress, psychological safety, measured using the Vineland-3 coping skills subdomain; Child health related quality of life (CHU9D) at baseline, 6 and 12 months (a versions of this has been developed suitable for pre-school children; Caregiver health (SF-36v2) at baseline, 6 and 12 months. Chosen to capture health impacts on carers and allow calculation of QALYs; Carer care-related quality of life at baseline, 6 and 12 months: ICECAP Carer Experience Scale (CES). Chosen to capture aspects of carer quality of life beyond health likely to be affected by the intervention; Potential risks and harms, including physical injury for the child and parent and other potential harms identified via the harms reporting process and process evaluation; Satisfaction with treatment, identified via the 12-month questionnaire and process evaluation.
	Estimate the relative efficiency of CHESS compared with usual care, in terms of self-care skills and involvement in young children with neurodisability.	Health and social care service use, collected via a service use questionnaire (SUQ) at baseline, 6 and 12 months Cost for carers/families for accessing services and purchased care: assessed through a time and travel questionnaire (TTQ) at 9 months.
	Conduct an embedded evaluation of recruitment, intervention acceptability, and implementation to support trial delivery.	Process evaluation interviews with parents and site staff of study process and acceptability (throughout the study).
Statistical methods	Baseline and outcome data will be described using summary statistics, broken down by group. All analyses will be based on the intention-to-treat principle. Primary outcome will be analysed using a repeated measures mixed effects	

	linear model extended for cluster randomised trials to include a random effect for cluster as well as participant. Models will include a fixed effect for treatment, nominal time, and the baseline outcome measures. Treatment effects will be estimated at each time point using a treatment-by-time interaction: the primary measurement time point is 12 months after cluster randomisation. The primary analysis will use an unstructured time and covariance structure, which gives unbiased treatment effects when outcome data are missing at random (MAR). A MAR mechanism is unlikely to be the case in this population, and we will explore the impact of missing data using pattern mixture models under missing not random assumptions using models for repeated measures data in cluster randomised trials outlined by Fiero et al. Secondary outcomes will be analysed in a similar way, with generalised linear models appropriate for the distribution of the outcome. All treatment effects will be presented using 95% confidence intervals. We will report tables disaggregated by sex and do subgroup analysis by sex, mobility (using adapted GMFCS), and socioeconomic status by adding a treatment-by-subgroup interaction to models. We plan no interim efficacy analysis, only one final analysis after the last participant has finished follow-up.
Co-ordination	Local: by local research teams Central: by Trial Office in Aberdeen (Telephone 01224 438405). Overall: by the Project Management Group and overseen by the Trial Steering Committee and the Data Monitoring Committee.

2. Plain summary

WHAT IS SELF-CARE?
Self-care means developing skills and independence in everyday activities, like using the toilet, having a bath, and getting dressed. It also means getting involved in the activities by making choices, joining in with routines, and coping with problems by finding individual solutions.

WHAT IS NEURODISABILITY?
Neurodisability describes a range of long-term conditions that affect movement, learning, hearing, vision, communication, emotion, and/or behaviour. It includes cerebral palsy, autism, and learning disabilities, as well as other diagnoses. Some children do not have a defined diagnosis, and some children's diagnosis has not yet been confirmed. Neurodisability affects over 500,000 UK children.

WHY IS THIS TOPIC IMPORTANT?
For children with neurodisability and their parents, achieving self-care is a major priority for health, wellbeing, and living an ordinary and fulfilled life. Most children with neurodisability need self-care support from therapists in the NHS and social care.

WHAT IS "CHildren's Early Self-care Support" (CHESS)?
CHESS is a new way to support self-care of young children with neurodisability between age 12 months and starting school. CHESS includes materials that therapists use to help families identify and communicate their self-care priorities, nurture helpful and enjoyable self-care routines, overcome barriers, and develop children's independence through movement skills. CHESS includes training for therapists and online resources for therapists and families.

WHAT IS THE RESEARCH AIM?
We will investigate whether CHESS is better than the usual self-care support provided by therapists and can save the NHS money.

WHAT IS THE RESEARCH DESIGN?
We will ask around 40 NHS services and 960 parents to join our research study. Social care and voluntary sector therapy services that usually provide self-care support will also be involved. Half the services will provide CHESS and half will provide their usual self-care support. We will measure children's self-care, health, and quality of life, and parents' health and wellbeing. We will also measure how much parents and services spend on accessing and providing support. We will compare the services several months after the start of CHESS to see whether CHESS improves self-care and is a sensible use of public money. To help the study run smoothly and explore people's experiences of CHESS, we will do interviews and focus groups with parents and therapists in some services. We will closely monitor whether CHESS has any potential risks or harms for children or parents.

WHO WILL CARRY OUT THE RESEARCH?
Our team includes researchers with expertise in large studies in the NHS and social care, therapists with expertise in self-care support, and a parent of a young person with neurodisability.

HOW WILL THE RESULTS BE SHARED?
We will share the results through the NHS and social care, academic journals, conferences, the British Academy of Childhood Disability and other professional bodies, national charities, parent carer forums, social media, and press releases. We will also develop creative ways to share the results with children.

HOW WILL PARENTS AND CHILDREN BE INVOLVED?
We will work with PenCRU (Peninsula Childhood Disability Research Unit), who are experts in involving families with disabled children in research through their Family Faculty group. We will also work with local and national charities and create links with community groups specialising in supporting families with children under five years of age.

3. Glossary of abbreviations

AE	Adverse Event
CHaRT	Centre for Healthcare Randomised Trials
CI	Chief Investigator
CONSORT	Consolidated Standards of Reporting Trials
CRF	Case Report Form
CTU	Clinical Trial Unit
DCE	Discrete Choice Experiment
DMC	Data Monitoring Committee
EQ-5D	EuroQol Group's 5-dimension health status questionnaire
GCP	Good Clinical Practice
GP	General Practitioner
HRQoL	Health Related Quality of Life
HTA	Health Technology Assessment
ISD	Information Statistics Division
ISF	Investigator Site File
ISRCTN	International Standard Randomised Controlled Trial Number
NHS	National Health Service
NIHR	National Institute Health Research
NRES	National Research Ethics Service
PI	Principal Investigator
PIL	Patient Information Leaflet
PMG	Project Management Group
PPI/PPIE	Patient and Public Involvement/and Engagement
PQ	Participant Questionnaire
QA	Quality Assurance
QALY	Quality Adjusted Life Year
QP	Qualified Person
RCT	Randomised Controlled Trial
R&D	Research and Development
REC	Research Ethics Committee
SAE	Serious Adverse Event
SAP	Statistical Analysis Plan
SD	Standard Deviation
SOP	Standard Operating Procedure
TMF	Trial Master File
TSC	Trial Steering Committee
UK	United Kingdom
UKCRC	United Kingdom Clinical Research Collaboration
UoA	University of Aberdeen

4. Trial Personnel

Chief Investigator

1	Niina Kolehmainen Newcastle University

Grant Holders

1	Samantha Armitage Sheffield Children's NHS Foundation Trust	8	Christopher Morris University of Exeter
2	Graeme MacLennan University of Aberdeen	9	Joanne Marshall The Newcastle Upon Tyne Hospitals NHS Foundation Trust
3	Jennifer McAnuff Pennine Care NHS Foundation Trust	10	Ceri Allen Sheffield Children's NHS Foundation Trust
4	Tim Rapley Northumbria University	11	Lindsay Pennington Newcastle University
5	Luke Vale London School of Hygiene and Tropical Medicines	12	Jeremy Parr Newcastle University
6	Nathan Bray Bangor University	13	Tomos Robinson Newcastle University
7	Shaun Treweek University of Aberdeen	14	Richard Sutton PPI Representative

Trial Office Team

1	Chief Investigator	7	Trial statistician
2	CHaRT Director	8	
3	Trial Manager	9	
4	Data Co-ordinator	10	
5	Senior Trial Manager	11	
6	Senior IT Manager	12	

Project Management Group (PMG)
This group comprises the grant holders along with representatives from the Trial Office team.

Oversight Group Members

Trial Steering Committee (TSC) Members
The membership of this committee comprises independent members along with the Chief Investigator (CI) (Niina Kolehmainen) or a nominated delegate. The other CHESS grant-holders, a Sponsor and Funder representative, and key members of the central office (e.g. the trial manager) may attend TSC meetings as observers. The terms of reference of the Trial Steering Committee, the template for reporting and the names and contact details of members of the TSC will be filed in the Trial Master File.

Data Monitoring Committee (DMC) Members

This committee is comprised of independent members and the trial statistician contributes as appropriate. The CI and / or a delegate may contribute to the open session of the meetings as appropriate. The terms of reference of the Data Monitoring Committee, the template for reporting and the names and contact details of members of the DMC will be filed in the Trial Master File.

Role of the Trial Sponsor and Funder

The Sponsor has responsibility for the initiation and management of the trial as defined by the UK Policy Framework for Health and Social Care Research v3.3 07/11/17. This is further defined within a sponsorship agreement outlining the roles and responsibilities of the parties involved in the research. Specific responsibilities delegated to another party are formally agreed and documented by the Sponsor.

The funder has oversight of the study through regular reports from the trial office. The funder appoints the independent members of the Data Monitoring and Trial Steering Committees and receives minutes from these. The funder is made aware of all outputs from the study but does not have a role in the decision to publish results from the study. In any publications, the funder is acknowledged, and appropriate disclaimer used to indicate that the views expressed are those of the author(s) and not necessarily those of the NHS, the NIHR or the Department of Health.

5. Introduction

5.1. Background and rationale

Caring for oneself ('self-care') is essential to survival and a key health outcome for children with neurodisability.[1, 2] Self-care encompasses two dimensions: self-care independence/skills (e.g. learning to feed oneself); and involvement in self-care situations (e.g. coping with mealtimes with others).[2]

Children with neurodisability, i.e. long-term conditions attributed to impairment of the brain and/or neuromuscular system,[3] represent the largest group of disabled children in the UK, an estimated prevalence of 3-4%.[4] They are at increased lifelong risk of multiple health and functioning problems, and their self-care problems are both significant[1, 5] and common.[6, 7] While many can achieve self-care levels close to their typically developing peers[8] this usually requires significant parent and therapy support (occupational therapy, physiotherapy, speech and language therapy), and more than half receive support for self-care.

There is currently little evidence on effective self-care interventions, no national guidelines, and no cost-effectiveness evidence for commissioners. As a result, therapy provision is variable and families receive differing advice.[9, 10] Clinical teams we spoke to emphasised the substantial difficulties, staff stress, and inefficient resource use that they face in trying to support self-care in absence of evidence about effective interventions. They described these as key reasons for their interest in participating in the trial.

There is parent, young person, and multidisciplinary professional consensus that additional evidence is needed,[2, 11] including from the British Academy of Childhood Disability-led James Lind Alliance (JLA) and Priority Setting Partnership (PSP).[12] In our 2016 PPIE work,[2] children with neurodisability, their parents, and professionals prioritised self-care for generating evidence about effective interventions. Children with neurodisability require significantly greater support for self-care from parents and caregivers, compared to their typically developing peers. Parents describe family life as immensely stressful. Parents and children are often supported by therapists and medical, education and social care professionals, with therapists leading self-care support. Published estimates of the costs of childhood-onset disabilities are limited but are likely very high[13, 14] and persistent.[15] Timely therapy, tailored to the child and family, may have substantial benefits for parent as well as child health by reducing caregiver burden, and may offer service efficiencies.

Our systematic evidence review and synthesis of self-care determinants and interventions[16] included 5 RCTs, 2 qualitative and 51 observational studies involving 7785 participants (largest study n=818). Meta-analysis was not possible due to heterogeneity of participants, outcomes and interventions. As consistent results across studies, we found that child's movement function (mobility and upper limb) was positively associated with self-care in 37/38 studies; cognitive function in 8/9 studies; and physical environment in 3/4 studies. Evidence of personal and social environmental factors was more limited. While promising intervention techniques were identified (e.g. adaptive equipment, goal setting, environmental modifications), these focused primarily on children with cerebral palsy rather than a wider range of neurodisability conditions.

Working together with our stakeholders (children, parents, professionals, designers, researchers) we have identified interventions across fields (e.g. behaviour science, family-centred care, cerebral palsy) with elements that show a consistent signal of efficacy; high desirability for children, parents, and professionals; and feasibility to use in NHS self-care support. These intervention elements include:

- Person-centred goal-setting and action planning (including related brief interventions), which can improve patient health across outcomes,[17-20] and facilitate more efficient therapy provision.[21, 22]

- Positive, daily self-care habits, which are key for long-term behaviour change and health,[23, 24] and require in-situ identification and use of contextualised cues, plans and rewards.[25]
- Enabling early movement, which can increase children's self-initiation, independence and control, and reduced caregiver assistance.[26]
- Equipment to enable early movement and self-care, where the equipment is feasible and acceptable but for which NHS provision is limited.[26, 27]

Our pre-trial work with stakeholders further strongly concluded that the delivery of self-care support needs to follow three key principles: (i) self-care support needs to be highly personalised and contextualised to the family's everyday routines; (ii) children with disabilities, like all children, develop through everyday play and self-care support needs to take play seriously; (ii) intervention delivery needs to be developmentally appropriate.[28]

While these elements and principles are generally viewed positively, and are known to relate to improved outcomes, they are rarely or inconsistently used in NHS practice.[11, 27-31] This trial will provide much-needed evidence of effectiveness and cost-effectiveness for commissioning and practice by answering the question: **Does Children's Early Self-care Support (CHESS) improve self-care in young children with neurodisability and is it cost-effective compared to usual care?**

5.2. Assessment and management of risks to the participants

An overview of the key risks to children and parents, and mitigations for these, are as summarised in Table 1.

Table 1: Risks and Mitigations

Risk	Mitigation	Protocol section
Recruitment: although cluster engagement in the pre-trial work has been good, there is still a risk of not being able to involve enough clusters, and/or there not being enough eligible participants within clusters	To maximise cluster and family participation, we have undertaken, and continue to undertake, extensive preparations with >40 potentially interested, definitely eligible clusters to plan the trial in a way that makes cluster participation, and sampling of family participants within clusters, as easy and as minimally disruptive as possible. The design and methods choices in this protocol represent the outputs from this co-design work.	**10**
Participant burden: providing data to the trial will have some burdens on parents who are already scarce on time and energy.	The parents of children sampled to contribute data to the trial, will be provided realistic information about the possible benefits, risks and burdens of contributing data to the trial. This will be provided in a Participant Information Leaflet (PIL) to parents, as well as discussion(s) with some combination of: the research team; a local cluster PI; and/or a local PI's designated research team member. The participant burden has been reviewed by PPI. The data collection tools and PILs have undergone extensive PPI feasibility and testing. This will continue to be reviewed throughout the study.	**10**
Consent: a risk that the consent taken does not follow the principles of GCP	Parents who agree to participate in the trial (i.e. to provide data) will sign a consent form approved by an NHS Ethics Committee. They	**10.3**

informed consent; or the consent is not documented properly.	will consent to the data collection with follow up within the trial. They can also choose to consent, or not, to being contacted in the future for further follow up, including electronic tracing using NHS data, and data linkage with computerised NHS data sources. Participants who are not able or not willing to provide data for the trial will not be recruited. Participants will be asked to consent for further contact for other research but may decline this and still participate in the CHESS trial. All clusters will receive appropriate training, and delegation logs will be kept; and the CTU will monitor consent forms and offer additional training as required.	
Intervention: parents and children who are not participating in trial data collection will also receive the allocated intervention and be exposed to information about the trial.	Parents of all children who are on the participating clusters' caseloads during the trial will be informed about the trial through a brief flier that includes a link to the study website. The flier will provide summary information of possible benefits and known risks of both the intervention and current care; and will provide contact details for parents to discuss any concerns.	**10**
Outcome measurement: some of the measures may be distressing to parents, due to the questions asked.	The outcome measures selected are among the most widely used in research with this population, with the most robust psychometrics and the least measurement burden for parents. We have used most of the measures in previous studies, and have also had extensive PPIE input to understand and mitigate any potential issues related to the short-comings in the measures – and this has resulted in a framework and materials for the trial staff to deliver the measures in a way that helps parents understand the trial and the context of the questionnaires. In addition, flexibility in method used to complete the questionnaires (hard-copy/online/telephone/in-person) has also been included.	**14**

The following are not considered risks in CHESS: the trial does not involve any intervention components that would not have been previously or are not currently used within NHS routine (i.e. non-research) context; we do not anticipate that children or parents will run additional risks by providing data to the CHESS trial. The CI will ensure, through the TSC, that adequate systems are in place for monitoring the quality of the study (e.g., compliance with GCP) and appropriate expedited and routine reports of adverse events, to a level appropriate to the risk assessment of the study.

In addition to risks to the participants, risks to trial delivery will also be monitored and assessed throughout as described in the Trial Monitoring plan. Reports of such monitoring will be presented with any mitigation requirements discussed during regular PMG, TSC and DMC meeting, where appropriate.

6. Trial aims and objectives

Aim: To determine the clinical and cost-effectiveness of CHESS in young children with neurodisability compared to usual care.

Objectives:

Primary Objective:

- Determine the clinical effectiveness of CHESS compared with usual care, on self-care skills and involvement for young children with neurodisability, measured at 12 months after cluster randomisation.

Secondary Objectives;

- Determine the clinical and cost-effectiveness of CHESS compared with usual care, on self-care skills and involvement for young children with neurodisability, measured at 6 and 12 months after cluster randomisation.
- Estimate the relative efficiency of CHESS compared with usual care, in terms of self-care skills and involvement in young children with neurodisability.
- Conduct an embedded evaluation of recruitment, intervention acceptability, and implementation to support trial delivery.

7. Trial design

CHESS is a 2-arm pragmatic cluster RCT across 40 clusters (paediatric community and outpatient NHS therapy services) with embedded economic and process evaluations (Figure 1). The trial will evaluate CHESS (CHildren's Early Self-care Support), a multicomponent behaviour change intervention, compared to usual children's community NHS therapy (occupational therapy, physiotherapy, speech and language therapy) for self-care. In CHESS, the therapists and families will receive materials and training about communicating self-care priorities, nurturing helpful and enjoyable self-care routines, overcoming barriers, and developing children's movement. In usual care, the therapists and families will do what they would have done if no trial was taking place. As CHESS is a cluster randomised trial, all eligible children within a cluster will receive the allocated treatment; i.e. there is no random allocation of individual children. For details of the intervention, see section 13.1.

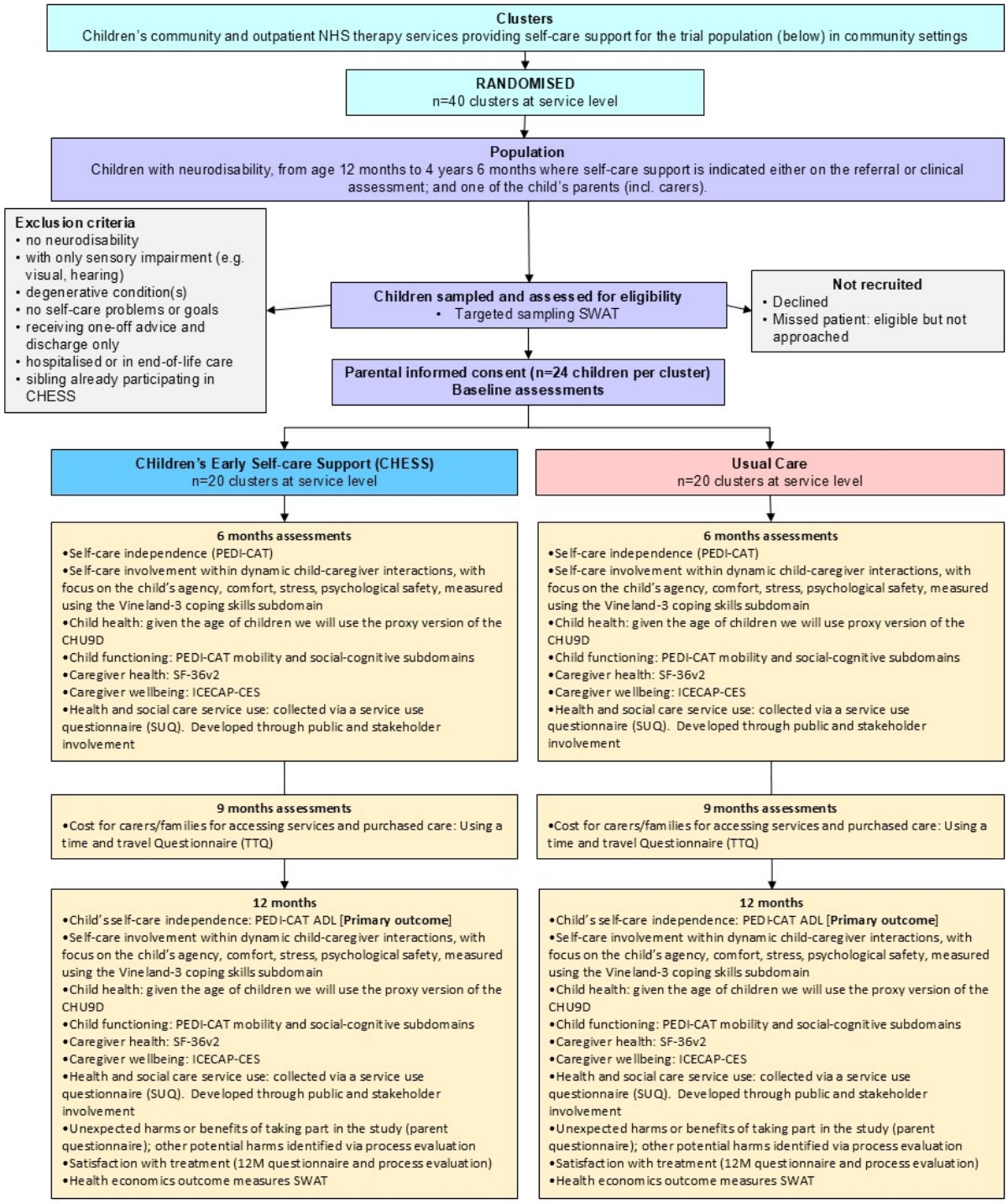


Figure 1: CHESS flow diagram

8. Trial population

8.1. NHS clusters: description, inclusion/exclusion, sampling and setting

In CHESS, a cluster is an NHS organisation. A cluster may work with other NHS, social care, and/or early education providers. This forms the context in which the clusters operate.

Clusters that are eligible meet all of the following:

(i) provide paediatric community and/or outpatient occupational therapy, physiotherapy, and/or speech and language therapy;

(ii) for eligible children (age from 12 months to 4 years 6 months, with neurodisability);

(iii) in England, Wales, Scotland or Northern Ireland.

Within a cluster there will be therapists of different grades, and therapy assistants working under their supervision, who deliver care across settings, including clinics, homes, childcare, and early education. Within a cluster there may also be health visiting, community paediatrics, and acute care services, among others. Other services (e.g. social care, Third Sector, local authority services, childcare) provide context but are not part of the cluster itself. The therapists may provide input to a range of services, i.e. to different populations of children across settings and clinics – some that will be eligible for inclusion in the trial, and others that will not be.

Exclusion at cluster level: as the intervention focuses on home and community settings (as opposed to children in hospital), services providing hospital-based care only are excluded. Tertiary services offering diagnostic-only or consultation-only care, and not therapy to child's life at home, will be excluded. Similarly, education-based services with no, or minimal, input to the child's family and the child at home, will be excluded.

As part of the pre-trial work, we established a sampling frame of n=144 paediatric therapy services in the UK,[32] and have used this, alongside professional networks, to identify eligible clusters. We will proactively seek to include therapy teams who have not traditionally been research active, opening opportunities to research participation to regions, and populations currently underrepresented in research. This will improve the diversity of both participants and professionals, widening the applicability of our results and in line with NIHR EDI policy. The CI has a track record in research capacity building, including within NIHR Academy (e.g. NIHR Academic Training Advocates), and will take the lead on supporting the cluster ('site') PIs. We will also work closely with cluster R&D teams and other professional groups (e.g. medical consultants) to put in place local mentoring and support.

8.2. Target patient population: description, inclusion, exclusion

The target patient population is children with neurodisability with or without a defined clinical diagnosis, from age 12 months to 4 years 6 months (at time of sampling the child), where self-care support is indicated either on the referral or clinical assessment, together with one of the child's parents or carers. Neurodisability refers to the established consensus definition,[3] and covers a range of children typically seen in paediatric community/outpatient care. For the child's age, the upper limit cut off is based on estimated age of school entry, where school entry is the significant developmental transition recommended by our PPIE.[2]

Inclusion criteria:

- Any child with neurodisability;[3]
- age at least 12 months and not older than 4 years 6 months on the date of sampling the child;
- seen by at least one paediatric community/outpatient therapy service; where self-care support is indicated;

- where the family and therapist agree on a need for self-care intervention;
- who has been randomly sampled for data collection from the participating therapists' caseloads;
- AND whose parent is willing to consent to data collection and comply with study procedures.

No restriction is placed on child mobility, cognitive or communication capacities and due to the nature of caseloads and random sampling of children we anticipate the same to include a mix of children across capacities.

Exclusion criteria:

- Children with no neurodisability;
- with only sensory impairment (e.g. visual, hearing);
- with degerative condition(s);
- no clear self-care problems or goals;
- receiving one-off advice and discharge only;
- hospitalised or in end-of-life care at the point of sampling;
- and/or a sibling already taking part in (i.e. providing data to) the CHESS trial.

8.3. Co-enrolment

Participants are permitted to take part in other non-interventional studies (e.g. questionnaire studies). Participants may be enrolled into other interventional studies providing that the Chief Investigators of both studies agree there is not impact on ability to deliver the intervention or on the primary outcome. A co-enrolment agreement may be required.

9. Sampling

Sampling in CHESS will involve recruiting the clusters, identifying the relevant caseload lists within the cluster, and sampling a sub-set of eligible children from within those caseloads.

9.1. Identifying and approaching clusters

We will recruit around 40 NHS organisations (clusters) providing community or outpatient occupational therapy, physiotherapy, or speech and language therapy for children (from 12 months to 4 years 6 months) with neurodisability. Within a cluster organisation there may also be health visiting, community paediatrics, and acute care. Other services (e.g. social care, third sector, local authority services, child care) are part of context but not the cluster itself.

We will include clusters from an existing list of 144 eligible NHS organisations, as well as through our wider professional networks. We will proactively seek to include clusters that cover populations living in higher areas of income deprivation and/or of non-white ethnicity. The identification of the clusters will be informed by existing routine data, e.g. data provided by the Office of National Statistics.

Contact with, and the engagement of, clusters has already taken place, over a period of months, as part of informing the funding application and the development of this protocol. This has allowed time for questions and discussions about trial acceptability with the clinical team. Generic non-identifiable information about the clusters will be collected, e.g. relevant staff FTE, population size, local area distribution and service boundaries, cross-working to / with other NHS organisations that could risk contamination, and previous experience in research participation.

9.2. Identifying eligible caseloads and sampling children

Before site/cluster randomisation, the local NHS organisation cluster PI (or a person designated by them), who already has access to the relevant child data, will identify their local caseload (sampling frame) of eligible children. The cluster PI, or their delegate will then collate a non-identifiable list of relevant caseloads within that cluster. That is, lists that consist of children from 12 months to 4 years 6 months old, with neurodisability, with likely self-care problems.

9.3. Simple random sampling

Simple random sampling will be the standard method used to randomly select which children and their parent to invite to participate in the data collection for the trial. We will also evaluate targeted sampling based on socioeconomic status in a Study Within A Trial (SWAT); see section 22.2.
The cluster PI, or their delegate, will send the full, non-identifiable caseload list of potential participants to CHaRT at the central trial office in an excel file format (or similar) using a secure file transfer system (such as a password protected encrypted file/ZendTo). This list will consist of a non-identifiable ID and a postcode for the child. The trial statistician will import the tables to STATA v18 for simple random sampling. Appropriate tools will be used to obtain deprivation data for the postcodes provided by the sites. Each eligible child will then be allocated a random number (using *runiform()* in STATA). A table which includes unique code, postcode, index of multiple deprivation(IMD) and assigned random numbers which has been generated will be exported into excel file and a process verification check will be conducted to ensure no duplicates or errors. The trial statistician will send the excel file back to the PI/delegate using a secure file transfer system (such as ZendTo). The PI/delegate will invite the parents of the sampled children to participate in data collection in CHESS, beginning with the randomly assigned first child on the list and continue through the list until the cluster target sample of 24 parents have agreed to participate in the data collection and follow up in CHESS, or the full list exhausted. The cluster PI will receive guidance from the research team as required.

Caseload and clinical lists are excluded from the sampling frame if they: involve very little contact with parents (e.g. services provided primarily to education settings); provide service to an organisation external to the approved trial organisation (e.g. a neighbouring NHS organisation); offer tertiary or other similar highly specialist care to a narrowly concentrated group (e.g. a specialist Fragile X service). Any discussions and judgements about excluding parts of caseloads or lists will be recorded by the research team, and in the site file.

10. Informing parents and children within clusters

The CHESS trial has a two-layer approach to informing parents about the trial. This approach takes into account the specific requirements of the cluster design – information sharing at cluster and individual levels. The approach has been designed in response to PPIE, where families – especially from backgrounds with minority ethnic, socioeconomic and religious backgrounds – emphasised the importance of informal as well as the usual, more formal, information sharing.

Prior to sampling patients, general information about the trial will be made available to all service users and the wider community connected to the participating clusters. This includes brief fliers and posters, as well as informal oral sharing, including sharing between families and communities. The trial research team will provide standard, preprepared materials for this to the clusters, but has little involvement in its wider dissemination (see below, Section 10.1).

Following sampling, the second layer is specific recruitment materials, provided only to the sampled subset of parents at each cluster who will be invited to contribute to data collection. This consists of the typical cover letter, participant information leaflet, etc. (see below, section 10.2).In addition to these two layers, we also anticipate that some parents may become aware of the trial through wider, general public information, e.g. press releases, funder's website, social media, generic trial news, wider engagement events.

Given the age group of the children (from 12 months to under 4 years and 6 months old), child specific information leaflets/posters will not be used as we are not asking the child to agree to participate in CHESS, but instead asking the parents to contribute to data collection. All children attending a cluster site will receive the same care (according to the site randomisation allocation) whether or not the parent agrees to participate in data collection for CHESS for their child.

10.1. General information about the trial for all service users

CHESS is a cluster randomised trial, and all eligible children within a cluster will receive the treatment allocated to that cluster. We will therefore make generic information about the trial freely available to all families with a potentially eligible child, who is receiving care within a participating cluster.

The parents will receive, from the relevant clinical manager / team lead, a generic, brief, clear information about the trial. The final version will be co-produced with PPI parent partners, but the content will consist of information about:

- What the trial aim is and why it is taking place,
- Who the intended target populations are,
- What the trial means for self-care support offered within the participating NHS organisations – including that all children within the service will receive the same care, and there is no allocation to different treatments within the service,
- How parents can get more information, including a link to the trial website, if they so wish.

This initial information leaflet will make it clear that: there is no need for the parents to take any action at this time; the researchers will not be involved in the child's care; and that the research team will not access any of the child's or family's details without the local clinical team first contacting the family to ask for a permission.

10.2. Recruitment of specific children and parents

The approach to recruit individual families will come from the local clinical team – either by post, by phone or in person.

The local PI, or their delegate, will send or give the parents of sampled children a standard recruitment pack (containing an invitation letter, the participant information leaflet (PIL) and expression of interest form) inviting them to participate in data collection for the trial. The recruitment packs for the trial have been designed through extensive PPIE (including with families with diverse backgrounds and characteristics). The packs will be pre-printed and compiled by the trial team and sent out or given to the subgroup of sampled children and parents by the local NHS team, from within the clusters. The trial researchers/trial office team will not access any identifiable patient or family details at this point.

The recruitment materials will specifically invite the parent to participate in the trial data collection – making it clear that their decision to participate in data collection will have no impact, either way, on their child's treatment allocation or options. It will invite them to contact the agreed point of recruitment to discuss participation in the trial, and if willing, to sign the consent form and complete the baseline questionnaire. It will also provide information that if the parent is willing, the research team will obtain clinical case note data on the child's diagnoses, development and treatment(s) provided.

Parents will be invited to indicate their willingness to discuss participation by telephoning, texting or emailing the trial team, by returning a pre-prepared expression of interest slip in a prepaid envelope, or by verbally indicating their willingness to a member of the local cluster team / service. This indication of willingness to discuss participation will start the process for informed consenting, below (section 10.3).

Sampled parents who have not made contact to discuss their potential participation, will be followed up by a first and second reminder (telephone, postal or in person where applicable) by the local clinical team, a week and two weeks later, respectively. If the parent/carer volunteers a reason for not participating, this will be recorded. After two reminder attempts, no further contact about participation will be made.

10.3. Informed consent

10.3.1. Consent and the professionals

At cluster level, local relevant approval from clinical lead/service manager/R&D will be considered confirmation of consent for randomisation of the cluster.

For professionals within the clusters, attending training in the trial process and intervention will imply consent to be involved in CHESS. For process evaluation, a separate process for consent to audio recording and participation in interviews will be sought, see section 17.

10.3.2. Consenting parents

Informed consent, for participation in the trial data collection, will be sought from parents of the sampled children, according to Good Clinical Practice (GCP) guidelines. For each child, a parent (primary adult caregiver) will be asked to consent to: data collection at baseline and follow-up; and contact in the future about this and about other relevant research.

CHESS is a cluster trial, at a level of a whole NHS organisation, with the participants being young children and their parents. There is no individual child or parent randomisation to treatments, and no parent consent will be sought for randomisation of the cluster. All families within an NHS organisation, whether or not they participate in the trial data collection, will receive the same treatment based on the randomisation of that cluster.

The following three steps will be taken:

Step 1: Sampled parents will receive **pre-prepared trial information packs,** before randomisation of the cluster, provided to them by the local clinical team (see 10.2, above), and at least one week left before following up (telephone, post or in person if applicable). Follow up will be by the child's NHS provider, usually in writing (by letter, email or text message – depending on the parent's stated contact preference with the provider); but may also be in person or by telephone, especially where there are established requirements for adapted communication (e.g. interpretation or parent's adapted communication needs) or where the provider is already in regular contact with the parent (e.g. a discussion during a routine appointment).

Step 2: For parents who indicate their willingness to discuss participation in data collection **a mutually convenient time to discuss the trial participation** will be arranged, e.g. by returning the expression of interest slip, emailing, or otherwise contacting the designated staff (see section 10.2, above). The discussions will be at a time, location and via method(s) preferred by the parent (in person, over the phone, Teams/Zoom), as mutually agreed. This discussion will be with an appropriately trained individual who is listed on the delegation log – this may be either a dedicated person within the local cluster team or a member of the central trial team, as agreed as part of cluster set up.

It will be explained to parents that participating in the trial data collection is entirely voluntary, that they are free to decline their child's data to be collected, that they are free to withdraw from the study at any time, and that their decisions will not influence their child's treatment and care in any

way. It will be explained that, in the event of withdrawal, any data collected to date cannot be erased and will be used in the final analyses.

As part of the informed consent process, the parents will be made aware of all aspects of the data collection, including the potential risks and burdens, as well as their responsibilities. Potentially participating parents will be given time to accept or decline involvement and will be given opportunity to ask questions and to have these answered before giving consent.

Step 3: If the parent/carer is willing, **informed consent to participate in CHESS will be sought and obtained** from one parent, according to Good Clinical Practice (GCP) guidelines, by an appropriately trained individual who is listed on the delegation log. This will be either a person at the cluster, or one of the members of the CHESS research team, as has been agreed with the cluster. If the parent/carer volunteers a reason for not participating, this will be recorded.

The informed consent will be received in person, by post (with the informed consent discussion happening over the phone/video call), electronically (eConsent) or verbally (audio recorded), in line with local and the parent preference.

a. *If consenting in person*: the consent form is checked, signed and dated by the person *receiving* the informed consent, listed on the delegation log with appropriate delegated responsibilities. A copy of the completed consent form will also be sent to the trial office where it will be further checked.

b. *If consenting by post*: the parent gives verbal agreement over the phone and signs, dates and returns the form by post to the local cluster or trial office, where the form is checked, signed and dated with the date of receipt by a member of the team confirming fully informed consent, who is listed on the delegation log with appropriate delegated responsibilities. The countersignature should only take place after discussion has taken place with the parent and any questions have been answered. A fully signed copy of the consent form will be sent back to the parent for their records.

c. *If using eConsent*: the parent will use the secure web-based trial management system provided by CHaRT to record their consent. If this option is preferred, the parent/carer will be asked to provide their email address which will be entered into the secure web-based trial management system. They will be sent a verification email with a link to verify their email. Once the email address is verified, the parent will be automatically emailed the PIL and a link to the e-consent form for their unique study number. The e-consent form will be identical to the approved paper version of consent form, with the approved PIL version number and date automatically populated. The parent will be asked to provide their signature online via a signature box using a finger tracing via a touch screen or using a mouse. Completed e-consent forms will be checked, and electronically counter-signed by someone listed on the delegation log with appropriate delegated responsibilities. The countersignature will only be recorded after discussion has taken place with the parent participant about the study and any questions have been answered. Only once both the parent participant and person receiving consent signatures are present will informed consent be considered to have been obtained. The trial management system used to record e-consent has a clear audit trail with tracking of all inserts or updates made. Database interactions logged against a user and date/time and the audit trail can be downloaded and analysed at any time by authorised users. A fully completed copy of the consent form will be emailed to the parent for their records.

d. *If using verbal consent*: the parent gives verbal agreement either over the phone/video call or in person following the informed consent discussion. The delegated member of the team receiving fully informed consent will ask for consent to audio record the verbal consent.

Once recording, consent to audio recording will be verified again. The delegated team member receiving consent will read through the approved consent form and confirm verbal consent (or not, for the optional questions) to each section and confirm consent to participate. The recording will be saved and uploaded to the trial website as evidence of verbal consent. The delegated team member receiving consent will complete a paper copy of the consent form indicating verbal consent was received. A completed copy of the consent form will be given/sent to the parent for their records.

Informed consent will be received by appropriately trained staff at cluster, or a member of the CHESS research team, delegated to do so. In this, we will apply a well-developed and tested approach to consenting, that we have used highly successfully in several NIHR and MRC-funded multisite (n>16 NHS organisations across England and Scotland) studies, in this population. This involves the research team working closely, and flexibly, with the local cluster PIs, and their delegates, as described, below. All parents will have the option to complete the consent form electronically rather than a hard copy. Details of the consent discussion, including discussion date, will be recorded on the trial inclusion CRF.

Flexible arrangements for consenting, where the cluster PI, their delegate, and/or the CHESS research team work closely together, will be essential due to the varying research capacity and capability within the workforce of the trial. CHESS will take place in community allied health care, where the treating professionals see children and parents across a variety of settings (homes, nurseries, clinics, community centres) as well as a substantial part of care being delivered remotely. Everyone who will be delegated to discuss the trial with parents for the purpose of consenting will have an understanding and appreciation of the context of early childhood neurodisability, and the multitude of challenges that parents of the children often face. This includes the understanding of cultural, socioeconomic, genetic, and other factors that mean often the parent as well as the child are affected.

If the person receiving consent feels that the parent has not understood the premise of the trial and/or the key information about what their participation in the trial involves, they will not take consent to recruit that parent to the trial. We anticipate this will be a very rare occurrence as parents need to have a level of capacity to safely care for young children, and we will monitor the number of consents declined by staff on these grounds – to identify any potential risk of un/conscious bias resulting in exclusion of eligible participants.

Following informed consent, parents will be sent/given a copy of the consent/e-consent form for their own records and a copy will be retained in the investigator site file and Trial Master File (TMF). For the clusters, only the cluster PI, or their delegate, will have access to the list of consented parents; participation is concealed from all other cluster clinical staff. This is to reduce potential bias in intervention delivery.

If a parent loses the ability to provide data for their child (e.g. due to an illness or accident, or a substantial change in care arrangements), a decision needs to be made, in conjunction with any other carers, about any further data collection for the child in relation to the trial.

Should potential parent participants, who have contacted the trial team and provided their names or contact details, subsequently choose not to take part in the trial their details will be deleted from the trial management system after 3 months.

10.3.3. Consent and the child participants

All participating children will be too young to provide informed consent, and no data will be directly collected from the children, and therefore no assent related to the trial will be sought.

All children will continue to have a say for all clinical procedures in the context of the therapeutic relationship. This will be managed by the child's care provider.

10.3.4. Non-recruited participants

The following anonymised information will be monitored and collected for all eligible children, and used to monitor representatives of the trial sample in relation to the overall, eligible caseload:

- Year of birth
- Postcode
- If the child was sampled for recruitment to data collection, the date when approached about participation

11. Promoting diversity and inclusion

We are committed to advancing inclusion and diversity in research. The focus of the CHESS trial is already, in and of itself, on a population with several intersecting characteristics found in populations often underserved in research: children, under 5's, with neurodisability, community-based healthcare, allied health professions.

In the CHESS trial design and methods, we further build on this by:

i. Ensuring the trial design, methods and materials are well-informed, through evidence and strong PPIE, about the particular characteristics related to neurodisability.
ii. Building on previous methods and materials that have been shown to promote and enable inclusion – to set data-informed targets for CHESS.
iii. Using an experimental intervention that is specifically designed to support and enable families to be heard, and authentically seen, by healthcare professionals.
iv. Deploying advanced sampling and recruitment strategies that build on our previous research with a track record of inclusivity and diversity.

Our approach is informed by the ongoing STRIDE project (https://www.abdn.ac.uk/hsru/what-we-do/research/projects/stride-supporting-recruitment-and-retention-improvements-for-diverse-ethnicities-283), which is developing guidance for trialists when selecting ethnicity targets. We will monitor ongoing recruitment against the stated targets (see 11.2) by creating and updating summary descriptions of the clusters and participating children, in line with the ongoing PRO EDI project to improve the way EDI issues are described in trials (https://www.abdn.ac.uk/hsru/what-we-do/research/projects/tools-to-help-reviewers-make-equality-diversity-and-inclusion-assessments-339).

11.1. Child and family characteristics, and neurodisability

We have completed NIHR INCLUDE Ethnicity and NIHR INCLUDE Socioeconomic Disadvantage Frameworks (https://www.trialforge.org/trial-forge-centre/diversity/). Information about the non-clinical characteristics of children with neurodisability and their families is generally limited, and suggest a complex relationship between neurodisability, socioeconomic factors, and ethnicity.[4]

From existing data, the consistent key points that will inform our recruitment and sampling strategy are that:

- There is a strong, well-established relationship between sociodemographic disadvantage and increased prevalence and impact of neurodisability. E.g. children with neurodisability are more

likely to live in lower income, single parent households and are more likely to live with a parent who also has a disability, compared to their non-disabled peers.[4]

- Evidence about ethnicity and neurodisability is limited. While the prevalence of neurodisability broadly increases in line with population density, there is some evidence of children from non-white backgrounds being at increased risk[4] and a suggestion that in some specific geographical locations, e.g. in London and Bradford, certain ethnic groups are experiencing higher-than-expected rates of congenital disorders associated with childhood neurodisability.
- The incidence of neurodisability varies by sex (males more commonly diagnosed);[4] however, this may relate, at least in part, to diagnostic criteria emphasising characteristics more common in males.

11.2. Building on previous methods: EDI targets for the CHESS trial

The CHESS trial builds on a previous longitudinal study (ActiveCHILD) as a benchmark for inclusion and diversity. That study, of n=301 children across 13 regions in the England, with a mean age of 21 months (SD=8), collected continuous data for an average of 10 hours per day for 3-7 consecutive days, repeatedly over up to six waves at every six months (average waves completed n=4).

ActiveCHILD achieved a sample that:

- Was balanced across sexes (56% female).{Thornton, Under review #11622
- Included families across all IMD categories (55.2% in the 4 most deprived IMD deciles).{Thornton, Under review #11622}
- Included children speaking languages other than English as their home language (10% of the n=118 families recruited through specialist services, including allied health therapies – with the main languages Arabic, Bengali, Bulgarian, Danish, Polish, Punjabi, Spanish, and Sign Language).[34]
- Included children with cognitive impairments (9%, n=19/223)[35]
- Included children using mobility aids (10%, n=22/219)[35]
- Including children with substantial communication limitations (13.8% of the n=118 families recruited through specialist services).[34]

The main limitations of the ActiveChild study, in terms of inclusivity and diversity, were an overrepresentation of more highly educated parents (71% were educated to at least degree level), and non-collection of specific data on ethnicity.[35]

As a target for inclusivity and diversity for the CHESS trial, we aim to match or exceed ActiveCHILD, shifting the dial closer towards population representation.

Characteristic	Operationalised definition for CHESS	Relevant ActiveCHILD sample estimate	UK population estimate	CHESS target
Sex	Biological sex assigned at birth	56% female	107 males born for every 100 females	45-55% female
Deprivation / Socioeconomic disadvantage	% living in the 4 most deprived IMD areas	55% in the most deprived 4 IMD deciles	-	≥55% in the most deprived 4 IMD deciles
Language	home language other than English	10%	8% of over 3 year-olds first language other than English	10% children home language other than English

Ethnicity	Parent-reported ethnicity for the child	Not collected	18% from other than white backgrounds	18% non-white
Disability - child	Significant cognitive and/or mobility impairments	10%	1 in 10 children have complex disabilities	10% significant cognitive and/or mobility impairments
Disability - parent	Self-reported long-term health conditions or disability	Not collected	24% working age adults report disabled	24% report disability or long-term conditions
Education	Parent self-reported educational level	71% educated to degree or higher	33% adults with level 4 (diploma, degree, PGR); 18% no qualifications	<40% with degree; >10% no qualifications

11.3. Self-care support tailored to different family cultures

Our pre-trial and intervention development research have highlighted that supporting self-care is particularly personalised, and sensitive to the child and wider family's values, preferences, and sociocultural beliefs. We anticipate intervention delivery to require to be mindful of family religious and wider cultural beliefs.

11.4. Diversity and inclusion in sampling and randomising clusters

Informed by the existing data, we will proactively seek to include clusters that serve populations living in higher areas of deprivation and above population average ethnic diversity. To aid this, we have used open-access Census data, and have developed profiles of potentially interested clusters, in terms of the population deprivation and ethnicity characteristics, to enable focus on those clusters with most deprived and/or most ethnically diverse populations.[36]

11.5. Diversity and inclusion in sampling children

We will also implement a Study Within a Trial (SWAT) to evaluate if a targeted sampling approach increases the number of children living in socioeconomically disadvantaged areas who take part in a trial. The SWAT will evaluate alternative ways of trying to ensure that children living in socioeconomically disadvantaged areas are well-represented with the CHESS population. Further details on the SWAT are given in section 22.1.

11.6. Diversity and inclusion in recruiting families

Evidence to support effective recruitment and retention strategies in paediatric trials (e.g. from Cochrane reviews) is poor; addressing this gap was highlighted as a priority in our 2018 Cochrane recruitment review;[37] and work on updating these reviews does not so far suggest new high-certainty evidence. Based on our pre-trial feasibility work, we expect inclusive recruitment of parents to rely on finding ways that allow them to engage with the trial in the context where: the parent energy and time is scarce due to the demands of their child's needs and family socioeconomic deprivation; and parent access to materials is hindered by parent disability and lower parent health literacy.

Given the lack of existing evidence and the need to find new ways for meaningful involvement, we built in a 6-month pre-recruitment period of set-up and tailoring work, involving PPIE with children, parents and expert advisors, to develop effective strategies to address sampling, recruitment and retention. This work (see section 5.1), including purposeful engagement of representatives from ethnic minority communities, has been instrumental in designing the present protocol for the CHESS trial. We have also had extensive discussions with the potentially interested trial clusters, to understand what data they do and do not hold, and what data can and cannot be accessed before sampling and recruitment.

Through the set-up and tailoring work, we have developed an explicit and conscious approach to participant relationship management, including recruitment and retention. The key principles, developed through the PPIE with parents, children and expert advisors, are:

- Commitment to taking active steps that allow children and parents to feel seen and appreciated, and feel that their (children and parents') participation is doing good.
- Encouraged to feel good about themselves and about being part of the research, and
- The trial team clearly demonstrating how much we value the participants at every opportunity.

We have brought these principles together with evidence-informed components (e.g. on best practice for questionnaire returns), learning from previous PPIE and the resulting successfully-applied strategies – and co-produced 'most feasible' approach to inclusive recruitment. This is described in Section 11.7 and operationalised in the trial promotion and recruitment materials.

11.7. Diversity and inclusion in consenting parents

To make the trial accessible and inclusive for people from diverse backgrounds, with different ways of communicating and thinking about information, and with varying resources, we have developed a suite of adaptations that can be deployed, as required, to tailor steps 1-3, above, for individual parents. These adaptations have been carefully designed with PPIE partners from across communities, ethnic and cultural backgrounds, and sexes – and with different ways of communicating and thinking. The adaptations relate to how we describe, present and share information about the trial; the ways in which we enable parents to ask questions and learn more about the trial; and how we receive consent. The adaptations will be available to all clusters and be deployed at an agreement of the cluster PI, their delegates, the parent(s), and/or the trial team, as appropriate. The suite of adaptations include the following, and can be combined as appropriate:

- Translated materials and information, including picture communication.
- An interpreter.
- Audio format of the key materials.
- In-person support to access and process information, explain and answering questions.
- Sharing materials with another person, trusted by the parent, and leaving them a copy that they can share and communicate about with others.
- Making clear, workable plans for support that the parent can access for completing the trial questionnaires.
- An option for verbal consent, audio recorded.

12. Randomisation and allocation of clusters

We will allocate clusters to treatment and control group using covariate constrained randomisation[38]. This approach minimises imbalance on cluster level covariates, which is a potential risk in cluster randomised controlled trials with fewer clusters. We will use Carter and Hood's[39] algorithm to optimise balance on the following: geography (urban/rural); cluster size

(number of therapists); and child population sociodemographics (namely significantly higher/lower levels of poverty or minoritised ethnic communities compared to UK average).

12.1. Blinding

There is no blinding of allocation. However, we will aim to keep therapists (apart from the cluster PI) blind to information about which families are providing data to the study. Cluster PIs will not be blinded to which families are providing data to the study because they need to assist in participant sampling and reporting harms (section 16).

12.2. Code break/Emergency unblinding procedures

There is no requirement for emergency unblinding procedures because randomisation is at cluster level and therefore knowledge of whether a family is providing data to the trial will not directly impact management decisions if an adverse event occurs.

12.3. Administration arrangements post recruitment (if applicable)

Following a family's entry in the trial the local cluster research team will:
- File a copy of the consent form in a local site file, held by the cluster PI.
- Enter trial data regarding the participant into the bespoke trial website.
- Maintain trial documentation at cluster.
- If completed by hard copy, return a copy of the signed consent form to the Trial Office in Aberdeen.

We will not inform the children's GPs that the family has been selected for data collection. The intervention is unlikely to have an impact on the child's medical care.

13. Trial intervention

13.1. CHESS

CHildren's Early Self-care Support (CHESS) is a manualised, multicomponent behaviour change intervention specified using the CONSORT TIDiER 12-point checklist. It is packaged as materials for therapists to use with families, and as training for therapists. The materials consist of:

- a CHESS intervention overview manual;
- a brief intervention technique for therapists to enable the child and/or parent to set personalised self-care goals and plan related actions within a routine appointment (at clinic/home);
- a home visit pack, with tailored pre-visit materials for the parent and child, and guidance for the therapist in how to facilitate the family to identify and habituate feasible opportunities for daily, positive, nurturing self-care actions and overcome barriers;
- a movement play pack, with invitation materials for the child and family to attend one movement play drop-in session, and guidance for the therapy team in how to facilitate the session, including access to early powered mobility equipment, to help explore options for longer-term access to movement play;
- an online self-care knowledge hub for therapists and families with illustrations, videos, case studies, and evidence related to self-care.

The training for therapists consists of:

- one in-person session for therapy teams (4hrs), using videos and team coaching prompts, to become familiar with the intervention principles and materials, and to tailor these to

local context; followed by remote implementation advice (e.g. phone, Zoom) for up to 6 weeks.

- A minimum intervention cycle with a family includes:
  - brief goal-setting (5-15min);
  - at least one home visit (~60min) using the related materials; and
  - an invitation to at least one movement play session (up to 90min).

The intervention can be tailored to children of varied development and abilities, builds on good therapy practice in the UK as well as research evidence, and has been co-created with public and stakeholder involvement.

Overall, the intervention is designed to activate four main change pathways:

(i) achieve a consistently high and longer-lasting dose of self-care skills practice for the child by making daily, developmentally advantageous self-care more automatic, habituated, and routine;

(ii) significantly reduce child and parent stress related to self-care activities by focusing on identifying opportunities for and overcoming barriers to emotionally nurturing self-care interactions within family routines;

(iii) increase children's independence and control and reduce caregiver assistance by enabling child self-initiation through early movement; and

(iv) ensure therapists' practice change and maintenance through cues, prompts and peer processes.

13.2. Comparator/usual care

Our research,[10, 27] has shown current therapy support for self-care consistently targets parent and child knowledge and skills (e.g. by instruction on how to perform the self-care behaviour, practice and rehearsal, graded tasks), and environment (e.g. by restructuring social environment, social support).

14. Outcome measures

14.1. Primary outcome measure

Child's self-care independence across daily life, measured using parent-reported Pediatric Evaluation of Disability Computer Adaptive Testing (PEDI-CAT) Activities Of Daily Living module, at 12 months after the start of the intervention. The choice of primary outcome was driven by the specification set by young people and parents in the initial PPIE.[2] The PEDI-CAT is one of few measures that: measures self-care in a way that relates to the child's everyday life; can be used across clinical diagnoses, settings and ages; minimises the response burden without compromising accuracy by using the Item Response Theory (IRT) statistical models to estimate a child's abilities from a minimal number of items that are most relevant to the child; has been developed and tested to assess change following an intervention; with strong psychometric properties in this target population; and has age and mobility filters that prevent irrelevant and potentially upsetting items from being presented. We have used the PEDI-CAT in a previous multisite study with 300 children (1-5yrs) across conditions, health states and abilities. It takes 15-20min to administer and can be completed online, in person, or over the phone at a time most convenient to the parent.

14.2. Secondary outcome measures

The choice of secondary outcomes has been informed by the requirements of the commissioning brief, and PPIE and wider stakeholder advice.

- Self-care involvement within dynamic child-caregiver interactions at baseline, 6 months and 12 months, with focus on the child's agency, comfort, stress, psychological safety, measured using the Vineland-3[40] coping skills subdomain.
- Child health related quality of life: given the age of children we will use the proxy version of the CHU9D, at baseline, 6 and 12 months.[41] A version of this has been developed suitable for pre-school children
- Caregiver health: SF-36v2[42], at baseline, 6 and 12 months. Chosen to capture health impacts on carers and allow the calculation of QALYs
- Carer care-related quality of life at baseline, 6 months and 12 months: ICECAP Carer Experience Scale (CES).[43] Chosen to capture aspects of carer quality of life beyond health likely to be affected by the intervention
- Potential risks and harms, including physical injury for the child and parent and other potential harms,[44] identified via the harms reporting process (section 16) and process evaluation
- Satisfaction with treatment, identified via the 12 month questionnaire and process evaluation
- Health and social care service use: collected via a service use questionnaire (SUQ). Developed through public and stakeholder involvement
- Cost for carers/families for accessing services and purchased care: Using an SUQ and a time and travel Questionnaire (TTQ) (TTQ developed through public and stakeholder involvement)

The subsections of these tools cover the most common important child health indicators across existing paediatric core outcome sets,[45] including pain, communication, social interaction, and mobility. Data on the children's diagnoses will be extracted from clinical notes.

15. Data collection and processing

15.1. Assessing outcomes

Assessment will be at baseline and at 6, 9 and/or 12 months after the therapy team has been trained in the intervention (Table 2, below). Further details about collection of outcome data are provided elsewhere in this section.

Table 2: Timing of data collection

Outcome	Baseline	6 months	9 months	12 months
Baseline CRF[1]	X			
Harms		X		X
Accidents during the powered mobility sessions[2]		Throughout		
Unplanned attendance at hospital	X	X		X
PEDI-CAT	X	X		X
Vineland-3 coping skills subdomain	X	X		X
CHU9D (proxy version)	X	X		X
SF-36v2	X	X		X
ICECAP CES	X	X		X
CHU9D, SF-36, and ICECAP CES 2week test/retest				X
Satisfaction with treatment				X
Health Service Use Questionnaire (SUQ)	X	X		X
DCE (sub-set of participants)			X	
Time and travel questionnaire			X	
Case note review/follow up CRF				X

[1]Completed by local cluster team at baseline, [2]Local cluster team will provide DATIX (or similar) reports of any accidents during the powered mobility sessions

Based on our previous longitudinal study of 300 children (1-5yrs) across 13 NHS organisations, with data collection every 6 months, we anticipate >90% of parents will prefer to complete the questionnaires over the phone or online, with high levels of flexibility required to accommodate around family routines (e.g. evenings, weekends, lunch breaks). PPIE has further emphasised that: helping parents to understand the trial and the context in which questionnaires are used matters; and having a trusted relationship with the trial team and a sense of being heard is important. We have previous experience of successful longitudinal study protocols of multiple questionnaires with this population; and will further build on this in the 6-month pre-trial set-up and tailoring period.

15.2. Baseline

Participating parents will complete the baseline questionnaire after consent and prior to cluster randomisation, including child and parent ethnicity and sociodemographics. The local cluster team will complete the baseline CRF including information on child age and diagnoses.

At baseline, we will also collect contact details of participating parents (including postal address, email, home and mobile number) and their contact preferences (email, phone call, post - and where required as a method of adapted participation to support inclusion, in-person visits).

15.3. Follow-up

The vast majority of the parent reported outcomes will be collected at baseline, 6, and 12 months post-randomisation, using questionnaires completed at home using their preferred method of completion (phone, post, email or in-person). The primary outcome measure (PEDI-CAT) uses computer adapted testing, which means it cannot be completed on paper. The questionnaires will include PEDI-CAT, SF-36v2, CHU9D, Vineland 3 Coping Skills Subdomain, and questions about current and recent treatment, satisfaction with treatment and harms. First reminders will be issued to participants (according to their stated contact preference). If there is no response, up to two follow-up reminders (according to their stated contact preference) will be attempted – with at least one of these via text message to allow for a change of postal / email address. Test-retest evaluation will require re-administration of the 12-month measurements 2 weeks after initial data collection, and will be collected from a 20% subset (Target Sample Size = 192, see section 22.5 for selection methods) of total participants. The Discrete Choice Experiment (DCE) and Time and Travel questionnaire (TTQ) will be collected at approximately 9 months post-randomisation only, with the DCE being completed by a sub-set of the respondents only (Target Sample Size = 240, see section 21.3 for selection methods).

Questionnaires will be administered to all parents consented for data collection, regardless of adherence with treatment, unless they have opted out of questionnaire follow-up. This means that consented parents and children who have not received their allocated treatment, have received the non-allocated treatment, or where therapy has been discontinued, will continue to be followed up in the study.

If questionnaires are returned as non-deliverable, attempts will be made by the local cluster team or staff at the Trial Office to trace the participant. In case of non-return of questionnaires, attempts are made by local cluster staff or staff at the Trial Office to trace the participant directly, and/or indirectly by contacting the GP.

15.4. Change of status, and withdrawal procedures

Participants remain in the trial unless they choose to withdraw consent. Participants are free to withdraw from the trial at any timepoint. All changes in status, except for complete withdrawal of consent, means the participant is still followed up for all trial outcomes wherever possible. All data collected up to the point of complete withdrawal is retained and used in the analysis.

Following informed consent, if a parent loses capacity, the consent given when capable remains legally valid. In such circumstances, a decision needs to be made, in conjunction with any other parent(s), family or carers, in relation to the child's ongoing participation in the study.

Participants who do not receive the cluster randomised treatment, or receive the other (non-allocated) intervention, or discontinue therapy are not considered withdrawals and will be followed-up for all trial outcomes unless they request otherwise. This is a pragmatic trial and we will monitor accruing data on treatment initiated and continued during the study.

Participants who request that no further questionnaires are issued (i.e. completing questionnaires) will be followed up for other trial outcomes unless they are complete withdrawals.

Participants for whom any outcome data are available will be included in an intention to treat analysis.

15.5. Data processing

Local cluster staff who are delegated to do so will enter locally collected data in the password protected, secure study website. Staff in the Trial office will work closely with the local cluster PI, their delegate(s) and teams, to ensure the data are as complete and accurate as possible. Postal questionnaires will be entered into the study website by trial office staff.

15.6. Long term follow-up

In the future, we will seek further support from NIHR to study longer-term outcomes. This may involve linkage with routine data from, for example the Office of National Statistics (ONS), NHS Digital, and other relevant government bodies or future contact with participating families. We will seek consent for this follow up at the point of recruitment and apply for funding for this as part of an additional application. Specifically, such funding would be used to collect more detailed long-term outcome data, such as the child's independence at primary school and parent longer-term health, wellbeing and care-related burden.

16. Harms

Our collection of data relating to harms within the study is underpinned by the following:

- We do not anticipate that the CHESS intervention per se will pose safety concerns to children or their families.
- All families in the cluster will receive the intervention, but only a sample will have provided consent for data collection.

There are potential for accidents during the powered mobility play sessions (for example a child falling or bumping the powered mobility equipment into someone else in the room). Such incidents would routinely be reported through the hospital DATIX system (or equivalent). To capture these incidents into the study data we will request copies of the DATIX reports as submitted. These will be reviewed by the trial team and forwarded to the Sponsor. Incidents will be collated and reported to the PMG, DMC and TSC at their regular meetings.

For children in the families contributing to data collection in both arms, parents will be asked to report any unplanned attendances at hospital at baseline, 6 months and 12 months. Reason for attendance will be recorded. There will be no assessment of expectedness or relatedness.

In the event that the researcher has concerns for the well-being of a participant or others, action would be taken to disclose concerns to a named contact (e.g. the participant's treating NHS clinician) though the researcher would speak to the participant about this first.

Events will be tabulated by arm, forwarded to the Sponsor. They will also be reported to the PMG, DMC and TSC at their regular meetings.

At 12-month follow-up, parents will be asked to report any unexpected harms or benefits of taking part in the study.

Our collection of data relating to harms will be risk adapted should anything flag up any signals of harm. If these are observed we will review the intervention, our processes and collection of harm data in collaboration with the Sponsor, trial oversight committees and funder.

17. Embedded process evaluation

We will conduct a mixed-methods process evaluation, involving a range of stakeholders, theoretically informed by Normalization Process Theory (NPT)(44, 45) and Theoretical Framework of Acceptability (TFA),(46) in order to: further develop recruitment and trial processes, with particular attention to inclusion and diversity; further specify the description of current care; and understand the determinants of successful provision and outcomes of CHESS, including potential subgroup differences.

17.1. Process evaluation design

This process evaluation will draw on interviews with parents who accept and who decline participation in data collection, interviews and focus groups with local cluster staff, written feedback from local cluster staff, as well as non-participant observations of the management and delivery of CHESS by local cluster staff.

17.2. Recruitment to the process evaluation

Our sampling strategy is informed by what we know about the study context,(14, 28, 31, 50) our current and prior experience with other trials,(51) our theoretical frameworks (as above), and wider key literature in the area. In keeping with the principles of rigorous qualitative research, sampling will be responsive to the study context. In some cases fewer interviews, focus groups and non-participant observations may be conducted with some groups, and in others, additional data will be collected in response to our emerging analysis and/or study events.

17.3. Identifying and recruiting process evaluation clusters

Clusters will be purposefully sampled for variation across a range of factors including geography, deprivation and ethnicity from those clusters taking part in the trial. During the internal pilot phase, we propose to recruit n=6-8 clusters. During the main trial, we propose to recruit n=3-4 additional clusters.

Clusters chosen to take part in the process evaluation will be informally approached by the process evaluation team. If they would like the cluster to take part in the process evaluation a meeting will then be held with the cluster PI and other relevant staff to explain the research aims, answer any questions and agree potential timetables of research activity.

17.4. Identifying and recruiting process evaluation participants: parents and staff

Participants for the process evaluation will be purposefully sampled for variation across a range of factors including geography, deprivation and ethnicity from those parents/carer invited to take part in the trial as well as staff at recruited clusters.

- Parents: We propose to recruit approximately n=30-60 parents accepting trial participation and approximatelyn=3-5 parents declining trial participation.

- Staff: We propose to recruit approximately n=60-120 staff involved in the management and delivery of CHESS.

Parents and Carers: For parents, we will have two distinct recruitment routes:

- *Core process evaluation route*: This will seek to recruit parents to take part in interviews and/or observations only. The majority of parent participants will follow this route.

- *Audio-recording recruitment discussions process evaluation route*: This will seek to audio-record the recruitment discussion with parents as well as seek to recruit parents to take part in interviews and/or observations. Only a small number of participants (n=1-3 per cluster) will follow this route and this route will only be used during the internal pilot phase.

Parents – Core process evaluation route
Initial information about the process evaluation will be included in the pre-prepared trial information packs that parents receive (see section 10.3.2, Step 1) and the consent discussion (section 10.3.2, Step 2). This will include a brief outline of the purpose and design of:

- Interviews to understand experiences of the CHESS study
- Observation of clinical appointments
- Audio-recording of recruitment discussions (internal pilot only)

The consent form for the main trial will include the options for parents to consent to be contacted to discuss participation in the process evaluation. Not every person that consents will be invited to take part in an interview or observation, and this will be clear to them in the pre-prepared trial information packs and main trial consent discussion.

The process evaluation team will be notified when a consent form has been received from a trial participant or trial decliner who has specifically consented to be invited to take part in the process evaluation. The process evaluation team will have delegated access to the names and contact details of the person, whether they have declined or accepted trial participation, and for those that have accepted, which cluster – control or intervention - they are situated in. The process evaluation team requires this information so that they can make appropriate sampling decisions and arrangements to conduct the interview and/or observation. If the process evaluation team samples them to invite them to participate, they will contact them to further discuss their participation.

Parents - Audio-recording recruitment discussions route (internal pilot only, 1-3 per cluster)
We want to audio-record a small number of recruitment discussions per cluster (approximately n=1-3 per cluster). We know clusters can be key to improving recruitment processes in trials. We also know it poses challenges because consent to audio-record needs to be obtained in advance of consent to participate in the main CHESS trial. In designing our consent process for this small process evaluation sub-study we have been mindful of the need to avoid overburdening participants with information, and of the need to consider consent for others (e.g. family member, friends, advocate) who may attend the recruitment discussion.

We have designed a three-stage consent process for the audio-recording - balancing the need for informed consent with the need to not disrupt the process of consent for the main CHESS trial. As in the core route, the initial information about the process evaluation will be included in the pre-prepared trial information packs that parents receive (see section 10.3.2, Step 1). This will include a brief outline of the purpose and design of:

- Interviews to understand experiences of the CHESS study
- Observation of clinical appointments
- Audio-recording of recruitment discussions

Stage One: At the start of the recruitment discussion (see section 10.3.2, Step 2), verbal consent to record the conversation will be obtained; it will be explained that more information about this will be given during the discussion and that there will be an opportunity at the end of the discussion to rescind consent. All present must give verbal consent; if anyone declines to give verbal consent then audio-recording will not take place.

Stage Two: Formal consent for recording will be taken as part of the formal consent process for CHESS. There are separate consent forms for those for others (e.g. family member, friends, advocate) present. All present must give formal consent for audio recording; if anyone present declines consent then the recording must be deleted immediately.

Stage Three: Those parents (and any others, e.g. family member, friends, advocate) giving formal consent to keep the audio recording are given a follow-up information sheet. Prominently on the front page of this information sheet is information that parents and others involved have a further opportunity to change their minds on the recording and ask the study team to destroy the recording.

As in the core route, the consent form for the main trial will also include the options for parents to consent to be contacted to discuss participation in a process evaluation interview or observation. Not every person that consents will be invited to take part in an interview or observation, and this is made clear to them in the pre-prepared trial information packs and main trial consent discussion.

Again, as in the core route, CHaRT will inform the process evaluation team when they receive a consent form from a trial participant or trial decliner who has specifically consented to be invited to take part in a process evaluation interview or observation. They will provide the names and contact details of the person, whether they have declined or accepted trial participation, and for those that have accepted, which cluster – control or intervention - they are situated in. The process evaluation team requires this information so that they can make appropriate sampling decisions and arrangements to conduct the interview and/or observation. If the process evaluation team decide to invite them to participate, they will contact them to further discuss their participation.

Staff: The local PI, or their delegate, will give or send staff involved in the management and delivery of the CHESS study a staff information pack about the process evaluation work and an expression of interest form. If they want to participate, they will be asked to complete the expression of interest form and return it to the process evaluation team. The expression of interest form will enable staff to be contacted to discuss participation in the process evaluation. Every person that expresses an interest will be invited to offer brief written feedback about their experiences of the management and delivery of the CHESS study to help identify further training needs, areas for learning, or additional support. However, not every person that expresses an interest will be invited to take part in a focus group, interview or observation and this is made clear to them in the information pack.

The local PI, or their delegate, will inform the process evaluation team when they receive an expression of interest form from a staff member. They will provide the names, role and contact details of the person. The process evaluation team requires this information so that they can

make appropriate sampling decisions and arrangements to conduct the focus group, interview, observation and/or written feedback. If the process evaluation team decide to invite them to participate, they will contact them to further discuss their participation.

17.5. Process evaluation informed consent: parents and staff

Informed consent procedures will ensure that participants understand that participation is entirely voluntary and that they can withdraw from the interviews, focus groups or non-participant observation at any time without this affecting their child's trial participation or other medical treatment, and in the case of local cluster staff, without this affecting their trial delivery role. Parents can agree to participate in the trial without participating in the process evaluation; cluster staff can be involved in trial management and delivery without participating in the process evaluation.

All individuals will be asked to provide informed consent in person, over the phone, or electronically (eConsent) to take part in interviews, focus groups and observation prior to the start of any interview, focus group or observation. Those participants taking part in more than one interview or focus group or observation will be asked for verbal reaffirmation of consent prior to the start of the next data collection; the researcher will keep a record of verbal consent for these additional events. For the brief written feedback, staff sending anonymised written feedback to the process evaluation team is considered consent to take part in providing this written feedback.

If a participant does not wish to be included in a specific non-participant fieldwork event the researcher would not make any anonymised notes about their participation in that observation.

In order to respond to unexpected people attending an event being observed, the researcher will wish to be able to consider a staged approach to consent. In relation to parents this could include people such as additional family member, friend or advocate and in relation to staff this could include people such as new member of staff or placement student. In the situation where an unexpected person is attending an event, the researcher will be introduced to them at the start of the session. They will briefly introduce the study to them, explain what they are doing and the purpose of it, as well as providing the full written information. The researcher will seek their verbal consent to observe their part in the event. If they do not wish to take part the researcher would not make any notes about their participation. If they are happy for the observation to take place however, verbal consent will be sought, consenting to the observation. The researcher will contact that person in the next few days (at least 24 hours later) to check they still wish to participate, and to gain consent for the observation. If they did not wish to participate the researcher will destroy any notes taken regarding that specific participant in the previous non-participant observation.

17.6. Process evaluation: data collection

Internal Pilot Phase – Parents: We will undertake interviews with parents accepting (approximately n=10-15) and where possible those declining trial participation (approximately n=3-5). We will explore trial processes (e.g. recruitment processes and materials; ideas and/or concerns about randomisation and consent) and the intervention (e.g. acceptability, ideas and/or concerns about self-care focus). Interviews will last approximately 20-60 minutes. They will be offered a choice of location and method (e.g. face to face; telephone; video conferencing) of participation. We know from our experience that parents of children with neurodisability routinely prefer online/telephone data collection as they can more easily accommodate them into their (often rapidly changing) schedules. We will also offer the option of someone else accompanying them in the interview (e.g. family member, friend, advocate) to offer additional information, support

or reassurance. Participants will be given the option to review their anonymised transcript of the interview and remove any data they do not wish to be used in analysis.

Internal Pilot Phase – Staff: We will undertake a focus group (n=1 per cluster) with cluster staff at participating clusters (n=6-8 clusters) during the internal pilot phase. Each focus group will include staff (approximately n=6-10 staff) working on any aspect of the local management and delivery of CHESS at that cluster. This could include people in a wide range of roles, including clinical, support, and management roles. We will focus on trial processes (e.g. information provision across languages and literacy levels; recruitment processes and materials; ideas and/or concerns about randomisation and consent) and the intervention (e.g. acceptability, ideas and/or concerns about self-care focus; interest, engagement and training needs). Focus groups will last approximately 45-60 minutes. Clusters will be offered a choice of format (e.g. face to face; video conferencing; hybrid). We know from our experience that aligning the focus groups to run before or after existing events where cluster staff come together makes coordination easier for staff. Participants will be given the option to review their anonymised transcript of the focus group and remove any data they do not wish to be used in analysis.

Internal Pilot Phase – Observation of CHESS delivery: We will also undertake non-participant observations (approximately n=1-5 per cluster) of the management and delivery of the intervention at participating clusters (n=6-8 clusters) during internal pilot stage. This could include a wide range of events, including cluster initiation visits, team meetings, goals setting in clinic appointments, a movement play session, as well as shadowing particular members of staff undertaking a home visit. We will focus on trial processes and intervention delivery. Non-participant observations will generally last 30 mins-4 hours. We expect most observation will take place face-to-face, with some via video-conferencing (e.g. if cluster arranges online team meeting to discuss CHESS related training needs). Staff involved in CHESS will also be prompted to write very brief written feedback – every two to four weeks - to document their experience and identify further training needs, areas for learning, or additional support.

Internal Pilot Phase – Observation of Recruitment Discussions: In addition, study recruitment discussions will, where feasible and with consent of all parents and staff involved, be audio-taped or observed (approximately n=1-3 per cluster). We will focus on information provision, ideas and/or concerns about randomisation, consent and data collection. Recruitment discussions will last approximately 15-60 minutes. We know from our prior experience that audio-recording and/or observing the recruitment discussions can enable us to obtain an objective record of the recruitment interaction with minimal disruption.

Main Trial – parents: We will undertake interviews with parents accepting trial participation. Those parents (approximately n=8-10) that already took part in interviews during the internal pilot phase (about initial experiences), will be approached at approximately 2 to 6 months after recruitment, in order to understand their experiences of the CHESS intervention, or usual care, over time. Additional parents accepting trial participation (approximately n=10) will also be approached to understand their experiences of their allocated intervention. The new parents will be sampled to explore, refine or refute emergent issues and findings; interviews will last approximately 20-60 minutes. Furthermore an additional subset of parents (n=10) will take part in interviews (approx. 30mins) to inform the development of the DCE; these interviews will focus on identifying the various attributes of the intervention(s) which impact patient/carer satisfaction, uptake, compliance, and adherence (see section 22.3 for further details). All interviewees will be offered a choice of location and method (e.g. face to face; telephone; video conferencing) of participation. We will also offer them the option of someone else accompanying them in the interview.

Participants will be given the option to review their anonymised transcript of the interview and remove any data they do not wish to be used in analysis.

Main Trial – local cluster staff: We will also undertake focus groups and interviews with cluster staff. Those clusters (n=3-4) that already took part in focus groups during the internal pilot phase (about initial experiences), will be approached later in process in order to understand their experiences of the CHESS intervention, or usual care, over time. Additional new clusters (n=3-4) will also be approached to take part in focus groups to understand their experiences of the allocated intervention. Finally, cluster staff (approximately n=15-25) will also be invited to take part in one-to-one interviews to further understand experiences of intervention. These clusters and participants within them will be sampled to explore, refine or refute emergent issues and findings. Focus groups will last approximately 45-60 minutes. Interviews will last approximately 20-60 minutes. Participants will be offered a choice of location and method (e.g. face to face; telephone; video conferencing) of participation. Participants will be given the option to review their anonymised transcript of the focus group and/or interview and remove any data they do not wish to be used in analysis. Staff involved in CHESS will be also be prompted to write very brief written feedback – every two to four weeks - to document their experience and identify further training needs, areas for learning, or additional support.

Main Trial Phase – Observation of CHESS delivery: In addition, the delivery of patient/parent-facing aspect of CHESS will, where feasible and with consent of all parents and staff involved, be observed (approximately n=1-5 per cluster, within n=6-8 clusters). This could include a wide range of events, including goals setting in clinic appointments, a movement play session, as well as shadowing members of staff undertaking a home visit. We will focus on intervention delivery to identify, characterise, and explain factors that shape CHESS intervention use, receipt and self-care. Non-participant observations will generally last 30 minutes - 4 hours. Data will be recorded via written field notes. No personal identifiable information will be recorded about clusters, parents or staff in written field notes. We expect most observation will take place face-to-face, with some via video-conferencing.

17.7. Process evaluation: data analysis

Data analysis will be on-going and iterative throughout the study. Interviews, focus groups and recruitment discussions will, with consent, be audio-recorded, transcribed verbatim and edited to ensure anonymity of respondent. Contemporaneous field notes from non-participant observation will be anonymised as they are produced to ensure anonymity of participants. The analysis will be theoretically-informed by our theoretical frameworks (44-46) and conducted according to the standard procedures of rigorous qualitative analysis(52) including open and focused coding, constant comparison, memoing,(53) deviant case analysis,(54) and mapping.(55) We will undertake independent coding and cross checking. Data will be analysed collectively in weekly 'data workshops' where the qualitative research team share and exchange interpretations of key issues emerging from the data, as well as workshops with the broader research team and PPI group.

17.8. Process evaluation data handling

The process evaluation will fully comply with the terms of the General Data Protection Regulation (GDPR) and Data Protection Act 2018.

To support anonymity of the participants, all clusters will be anonymised and given a unique reference number that will be used throughout all notes, transcriptions and publications. Identifiable data (e.g. contact details) will be held on a separate database (i.e. will not be linked to any data) and will only be used to contact the participant about the study.

For interviews, focus groups and audio-discussions when participants have been recruited into the study and given informed consent, they will be assigned a non-identifiable reference number and all data (paper and electronic) will use this number. Interviews, focus groups and recruitment discussions will be digitally recorded and transcribed verbatim in order to ensure fidelity to the views and actions of participants is retained in the analysis. Throughout transcription any identifiable information shall be anonymised. In the event that any participant was to withdraw their consent, any of their existing data would be destroyed and would not be used in further analysis.

Transcription of interviews and focus groups will be done by a professional company external to the research team.

Brief written feedback will be assigned a non-identifiable reference number and all data (paper and electronic) will use this number. We do not expect any special category or identifiable data will be collected or recorded regarding staff (or parents) in this written feedback. Any identifiable information shall be anonymised.

In relation to non-participant observation research no special category or identifiable data will be collected or recorded regarding parents or staff. Staff job roles will be recorded and included in the fieldnotes as this information is central to the core aims of the study. During non-participant observations no personal identifiable information will be recorded or any participant details, ensuring participant anonymity.

All data will be held on secure, password-protected Northumbria University servers. The analysis will be undertaken by the process evaluation researchers and they will be the only members of the team who will have access to field notes, audio-recordings and anonymised interview transcripts. The analysis will take place on Northumbria University computers. The digital voice recordings will be destroyed at the end of the study. All other records will be retained in a secure archive setting for 10 years to facilitate future analysis and publication of the study material.

17.9. Ethical issues related to the process evaluation

While every precaution will be taken to preserve patient anonymity and confidentiality there will be limits to this. In the event that the researcher has concerns for the well-being of a participant or others, action would be taken to disclose concerns to a named contact (e.g. the participant's GP) though the researcher would speak to the participant about this first. If a participant were to disclose anything indicating unsafe practices or misconduct, they would be directed to follow the complaints procedures (in PIL).

17.10. Relationship between process evaluation and main trial

The process evaluation team will present anonymised emerging findings to the trial team, PMG, TSC and PPIE stakeholders on the potential determinants of trial set-up and recruitment. These might include cluster-specific issues, issues across multiple clusters, or at the level of the organisation of the trial. We will work with the TMG, CHaRT and PPIE stakeholders, and where appropriate specific clusters, to develop a plan of action. We will focus on aspects that are

amenable to change. On-going cluster feedback will be undertaken in conjunction with CHaRT and TSC. All feedback to trialists and clusters will be supportive and constructive.

18. Trial sample size and proposed recruitment rate

18.1. Sample size

CHESS is designed to detect a target difference of 2 points on the PEDI-CAT ADL: for 90% power and 2-sided 5% α, assuming a 5-point SD from our cohort study of similar children, we require a sample size of 266 ignoring the clustered design. There is no minimally important difference established for the PEDI-CAT ADL. From our pilot data children improve about 8 points over a 24-month period, adding 2 points to that trajectory improves PEDI-CAT ADL scores by about 6 months crudely speaking -- with the caveat that improvement is not linear. A 2-point difference is equivalent to a moderate standardised mean different of 0.4, which is a worthwhile and achievable difference given the lack of an effective intervention for these children.

The unit of randomisation is at the service level. Inflating for the clustered design assuming an ICC of 0.1 (from our cohort study) and 40 clusters of size 20 has 90% power. Based on our previous studies in this population, we have assumed a drop-out of about 15%, each cluster will recruit 24 children, a total sample size of 960.

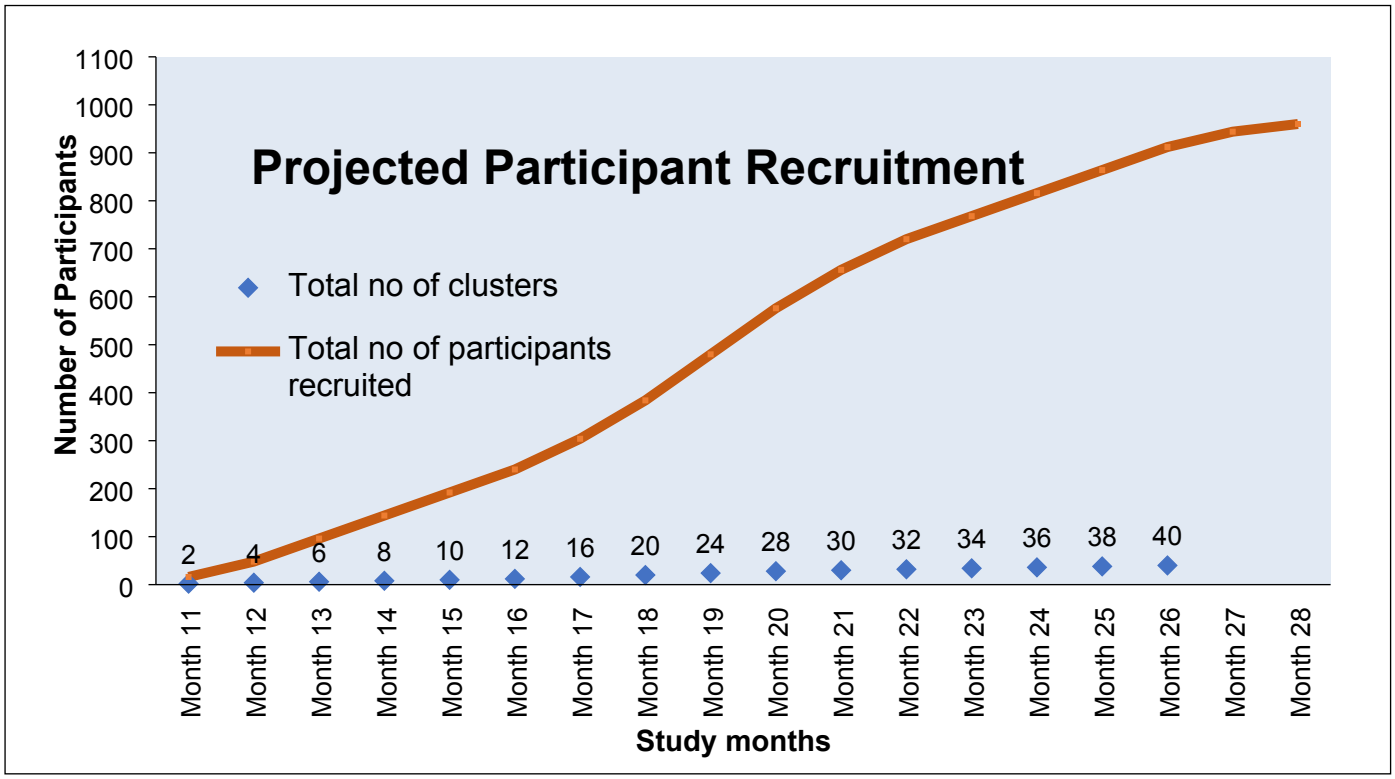
18.2. Recruitment rates

To optimise the timing and process of recruitment, we will sample and recruit all 24 children per cluster in a single sweep, by sampling across the cluster caseload (current cases and waiting list), inviting children and parents to participate, and collecting baseline data from those consented.

Our pre-trial work[46] showed that a cluster RCT is acceptable to therapists, and preference for treatment allocation at cluster-level has continues to be emphasised by potential clusters. Cluster sizes are likely to vary from small (~5 therapists) to large (>70 therapists), with an average of around 16 therapists. The overall caseload sizes per cluster also vary, from <100 to >7000 children (0-21yrs), averaging around 1088. There is no published, conclusive data on the breakdown of these caseloads; based on our scoping and previous work, we estimate around a third to be preschool age, of whom at least half with difficulties in self-care.

Our projection is based upon 40 clusters contributing 24 participants each over 3 months. Incorporating a staggered cluster set-up, we would expect to recruit the first 240 participants by month 16 (i.e., after 6 months' recruitment), 720 children and parents by month 22 and the remaining 240 by month 28, making a total of 960 participants after 18 months of recruitment.

Figure 2: Recruitment projection



18.3. Internal pilot study

We plan an internal pilot phase to test recruitment and randomisation over the first 9 months of recruitment (month 11 – month 19). Recruitment of 60% of the anticipated number of randomised clusters and participants at month 19 will be used to inform progression from pilot phase to full trial.

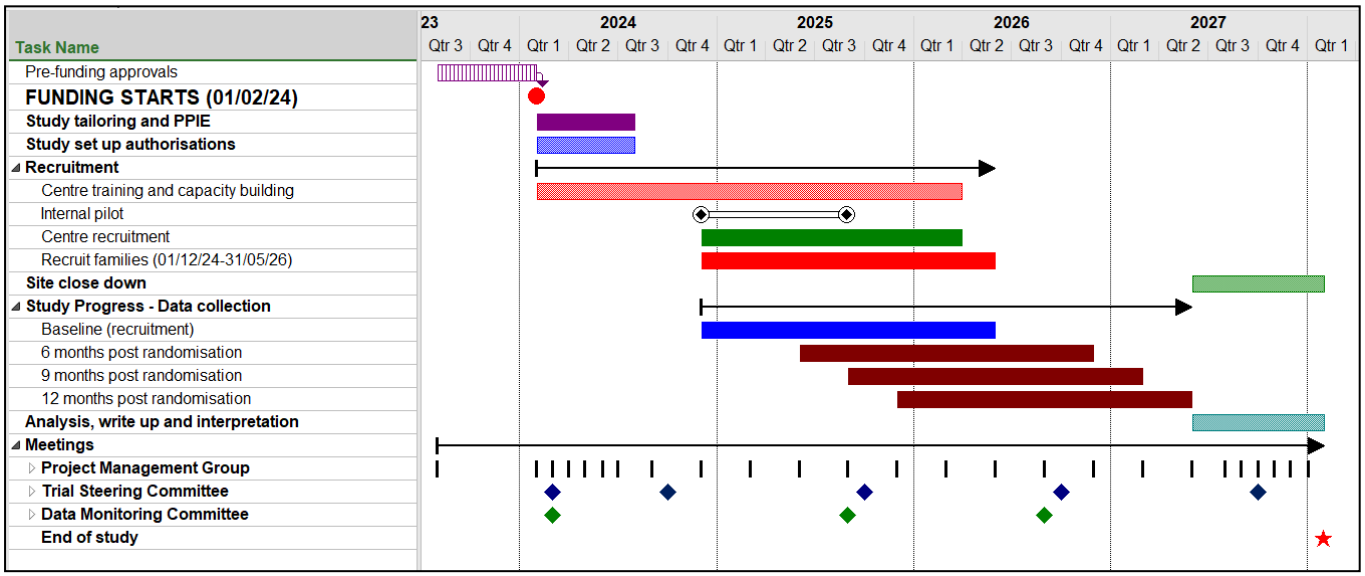
Table 4: Stop/go criteria at 9 months

	Red	Amber	Green
Cluster recruitment	<60% (<14 clusters)	60-100% (≥14 clusters)	100% (24 clusters)
Participant recruitment	<60% (<288 pts)	60-100% (≥288 pts)	100% (480 pts)
Intervention and trial process, including recruitment, acceptability	Qualitative work is strongly suggestive that one or both of the intervention and trial delivery are unacceptable with substantial change	Qualitative work suggests that one or both of the intervention and trial delivery require change. These changes are possible but some are non-trivial.	Qualitative work does not suggest problems with the intervention or trial delivery
Action	Discuss urgently with the TSC and potentially the funder, considering all options including discontinuation.	Consider recruitment strategies and blockages (if centres are not open), including trouble shooting, revised training and support, open additional centres	Proceed seamlessly whilst considering recruitment strategies

19. Project timetable and milestones

Start date: 1 Feb 2024. Duration: 48 months. Milestones: prefunding: regulatory approvals, cluster engagement; months 1-6: study tailoring with PPIE, set-up, authorisations; months 1-26: recruitment and training of clusters; month 11-16: internal pilot; month 11-28: child and parent recruitment; month 17 to 40: follow-up to 12 months after intervention; months 41-48: analysis, interpretation, reporting and start of dissemination (Figure 3).

Figure 3: Gantt chart



20. Statistical analysis

Baseline and outcome data will be described using summary statistics, by treatment group. All analyses will be based on the intention-to-treat principle. Primary outcome will be analysed using a repeated measures mixed effects linear model extended for cluster randomised trials[47] that includes a random effect for cluster and as well as participant. Models will include a fixed effect for treatment, nominal time, and the baseline outcome score. Treatment effects will be estimated at each time point using a treatment-by-time interaction: the primary measurement time point is 12 months after recruitment into the trial. The primary analysis will use an unstructured time and covariance structure, which gives unbiased treatment effects when outcome data are missing at random (MAR). A MAR mechanism is unlikely to be the case in this population, and we will explore the impact of missing data using pattern mixture models under missing not random assumptions using models for repeated measures data in cluster randomised trials outlined by Fiero et al.[48]

Secondary outcomes will be analysed in a similar way, with generalised linear models appropriate for the distribution of the outcome. All treatment effects will be presented using 95% confidence intervals. We will report tables disaggregated by sex. Subgroup analysis to assess potential treatment moderating effects of sex, mobility (using adapted GMFCS), and socioeconomic status will be carried out by modelling treatment-by-subgroup interactions. We plan interim efficacy analysis, only one final analysis after the last participant has finished follow-up.

Full details of the statistical analyses will be documented in the Statistical Analysis Plan.

21. Economic evaluation

A comprehensive economic evaluation will be conducted, including a within trial analysis, model-based analysis, discrete choice experiment and budget impact analysis. We will also conduct a Study Within a Trial (SWAT). The main outcome for both the within trial and the model-based analyses will be a cost-consequence analysis (CCA), in which a range of outcomes are presented to allow readers to form their opinion on the relevance and relative importance to their decision-making context. The purpose of the CCA is twofold: (i) to allow us to present impacts to the child and carer that under current methods cannot be aggregated together and (ii) identify and report outcomes that are not reflected in health related QALY estimates. Full details of the health economics analyses will be documented in the Health Economics Analysis Plan (HEAP).

21.1. Within-trial economic evaluation

The cost perspective (i.e. whose costs are considered) for all analyses will be health, social care and the family, with a narrower perspective of health and social care only being included as part of a sensitivity analysis. The perspective for effects will be child and carer, with a narrower perspective of child only or carer considered in sensitivity analyses. All costs associated with the delivery of the intervention will be collected as part of a micro-costing exercise at individual study centres. Centre level data will be supplemented with participant level data collected using study CRFs. The use of health and care services, including out of pocket expenses will be collected using a participant service use questionnaire (SUQ) administered at baseline, 6 months and 12 months post randomisation. The SUQ will be developed from our item bank of previous questionnaires and other relevant data collection tools (www.dirum.org). Time and travel costs borne by child and carers in accessing and using services will be estimated using information from the SUQ and from the responses to a one-off time and travel questionnaire (TTQ) administered at 9 months post randomisation. The TTQ will be developed from those used successfully in previous NIHR funded studies. Both the SUQ and the TTQ will be developed in partnership with the study PPIE group. The unit costs of NHS and PSS resource use will be estimated from study specific estimates and routine data sources.[49, 50] Unit costs will be combined with information on the use of services to estimate a cost for each participant/carer. For child/carer costs, the time and travel costs of accessing care will be estimated using the responses to the TTQ and data on the use of services. To this will be added the monetary cost of any private health care.

Health Related Quality of Life (HRQoL) will be calculated using a variety of measures. For the child, HRQoL will be measured using the proxy version of the CHU-9D.[41] This is currently validated for children ≥5-years-old, however a proxy version with additional guidance is available for children under 5. Recent evidence from Australia[41] concluded that the measure was found to be valid and reliable to measure HRQoL in children aged 2–4 years. As there is currently no evidence on the validity and reliability of the proxy CHU-9D for children under 2, we will conduct a sensitivity analysis which excludes those children aged under 2 at baseline from the analysis sample. For the parent carers, HRQoL will be measured using the SF-36v2. To further assess the parent carer's care-related quality of life, ICECAP Carer Experience Scale (CES) will also be collected. This tool focuses on 'care-related quality of life' rather than health-related quality of life, comprising attributes that are pertinent to unpaid carers.

In the base case analysis QALYs will be based on responses to the CHU9D (child) and the SF-6D, derived from response to the SF-36 (parent carer). QALYs for each participant will be obtained by scoring responses to the two tools at baseline, 6, 12 months post randomisation using scoring systems available at the time of analysis that are appropriate to each tool. QALYs cannot be calculated for the CES. The mean CES index at each data collection point will be estimated, with this information used to complement the QALYs calculated from the SF-6D in the CCA.

An appropriate regression model will be fitted to estimate marginal costs and QALY gains; controlling for baseline covariates. Data will be presented as point estimates and bootstrapping techniques will be used to characterise imprecision (Barber 2000). The incremental costs and QALYs based on the CHU9D and SF-6D will be reported in a CCA, presented in the form of a balance sheet. Also included in the CCA will be the mean responses to the CES, and other outcomes from the trial that cannot be directly mapped back on to the CHU9D, SF-6D, the CES or costs. The results will be expressed in the form of incremental costs, incremental QALYs and incremental cost-per-QALY. Stochastic sensitivity analysis will utilise the non-parametric bootstrapping technique with multiple bootstraps to explore the impact of statistical imprecision surrounding the point estimates of costs, QALYs (and other outcomes as appropriated in the CCA) and incremental cost per QALY. For the CCA and cost per QALY analysis, the cost-effectiveness (CE) plane will be used to illustrate the relationship between costs and measures of effect of interest. For the cost per QALY analyses cost-effectiveness acceptability curves (CEAC) will be presented. Deterministic sensitivity analysis will be conducted to explore sources of uncertainty.

21.2. Model-based economic evaluation

The 12-month follow-up of the trial will not capture all the costs and health outcomes associated with the interventions, as some events will be incurred over a longer timeframe. Therefore, an economic decision model will be developed to extrapolate costs and outcomes over the lifetime of the child and to compare hypothetical variations of the intervention that differ according to the component delivered. We anticipate the model will be a microsimulation model. We will work with our PPI and clinical team to design a model reflecting the child's journey both with and without the intervention. The model will be constructed following guidelines for best practice in economics modelling.[51] The use of services both with and without the intervention will be modelled using data obtained from the trial and longer-term follow-up. Further data will be systematically derived from the literature and from expert clinical input (e.g. the Centre for the Evaluation of Value and Risk in Health (CEVR) Cost Effectiveness Analysis (CEA) Registry).

The model will be used to produce estimates of costs and QALYs (from the CHU9D and SF-6D). Further data for the economic model (for instance predicted uptake rates for alternative configurations of the intervention) will come for the DCE (please see Section 21.3 for more details). Relative efficiency will be reported in the same way as for the within trial analysis. The model will be probabilistic, with appropriate distributions used for the various parameters. The choice of which distributions will depend upon the data available and recommendations for good practice in modelling. The results will be presented as point estimates of costs, QALYs, incremental costs, QALYS, CCA outcomes and estimates of incremental cost per QALY gained. They will also be presented as plots of costs and effects and cost-effectiveness acceptability curves. The model will be developed in a suitable software package (e.g. R).

21.3. Discrete Choice Experiment (DCE)

Discrete choice experiment (DCE) is a quantitative method increasingly used to elicit preferences from participants (e.g., patients, the public and policymakers). In a DCE, participants are presented with a series of alternative hypothetical options (e.g., different configurations of a service or intervention) described by a set of different attributes (e.g., how often the intervention is administered, which healthcare professionals are involved), with each attribute taking one of several levels (e.g., setting the intervention frequency at weekly, monthly or annually; varying whether the intervention involves consultant and/or wider allied healthcare professionals). Participants are asked to state their preferred choice between two or three competing options, each choice consisting of a combination of these attributes/levels.

In this study, a DCE will be developed utilising the findings from the embedded process evaluation qualitative interviews, and existing and ongoing PPIE inputs. As a distinct part of the process evaluation, we will carry out separate interviews (n=10) with parents/carers to identify the various attributes of the intervention(s) which impact patient/carer satisfaction, uptake, compliance, and adherence. Participants from the main trial will be identified through the process evaluation and PPIE groups, and a patient information sheet for the DCE qualitative interviews will be sent to prospective interviewees to enable informed consent. Interviews (approx. 30 mins) will be carried out in person, online or by phone, depending on the preferences of interviewees. The interviews will, with consent, be audio recorded, transcribed verbatim and edited to ensure anonymity of responses (all data collection, data handling and ethical procedures will be in-line with the process evaluation approach in 17.5-10).

A prototype DCE will be designed and trialled with a test group of service users (n=10) and PPIE advisors. This piloting work will help ensure participants' understanding of choice contexts, generation and testing of appropriateness and understanding of attributes/levels, task complexity, length, timing, and likely response rates, and whether participants are willing to trade between the different attributes pertaining to specific policies in the subsequent DCEs. If required, translations for the DCE will also be tested at this stage, to ensure comparable understanding between different language formats. The trial will also be used to determine whether data collection is possible via a remote questionnaire or whether data collection through phone interviews will be required. Throughout this process we will engage with our PPIE representatives to seek their thoughts on developing the testing materials for use in the pilot studies. Feedback will also be sought from patient facing staff members to determine if the DCE attributes/levels are in-line with their experience of service user expressed preferences and the realistic bounds of such services.

The final DCE will be administered through 25% (n=10) of cluster settings, with the aim to recruit 50:50 from CHESS and Usual Care clusters. If possible, settings will be chosen to represent a range of geographic/ethnic/socioeconomic populations (in-line with the process evaluation approach, section 17), as well as cluster caseload sizes. Intervention clusters will be taken from a subset of those recruited for process evaluation; requirement for control clusters will depend on data collection format (with preferred collection method of remote questionnaire not requiring cluster recruitment). The trial management team will support the identification of comparable matched control clusters to the intervention clusters used. Data will be collected at around month 9 of the data collection period. A patient information sheet will be included with the main data collection at 9 months to give more information about the purpose of the DCE. Although all included cluster participants will be asked to complete the DCE, due to the time required for completion it will be made clear that it is not part of the core data collection, and it is their choice to complete it. A completion rate of greater than 50% is anticipated (n>100) in each arm, with a total target sample size of n=240.

From the full DCE responses we will produce a ranked list of attributes and their relative importance to participants' intervention preferences, as well as trade-offs between attributes. Data will be analysed in a random utility framework using appropriate logistic regression methods such as conditional logit/multinomial logit models.

The DCE will also be used to predict uptake rates for alternative configurations of the intervention that differ according to the components delivered. These alternative configurations will be incorporated as additional comparators within the economic model, with costs and impacts assigned accordingly. This will allow the estimation of the relative efficiency of alternative configurations of the intervention to inform decisions about whether the intervention could be tailored.

21.4. Budget Impact Analysis

A budget impact analysis to explore the cost impacts of implementation at scale will be conducted. This analysis will be developed from one previously developed for the previous HTA EMPoWER study (26). As costs at scale are directly linked to the uptake of the intervention, we will use predictions from the DCE to predict uptake rates of the intervention as delivered in the trial.

For the budget impact analysis, we will establish the costs of the proposed intervention, and the impact of making such provision available to all children who could potentially benefit. Provision to all by the NHS would inevitably lead to increased costs, however, this increased cost should be considered in the light of the potential benefits to the children, potential cost savings, and additional cost implications for other services in the public sector.

Cost data will be obtained from publicly available sources and publications, as well input from PPIE and clinical advisors. UK government datasets and related websites (e.g. Office for National Statistics (ONS), Department for Work and Pensions) will be consulted to inform population calculations. NHS data sets will be used to inform numbers of users and input to costings. Published reference sources, published research and freedom-of-information requests will be used to further inform aspects of the intervention costing, including staffing and equipment.

All of the relevant cost data will be synthesised into a tariff of NHS and non-NHS costs relating to the intervention, which will subsequently used to inform the budget impact analysis. A broad societal perspective will be taken; thus, cost implications for social care providers, other public sector resources and for the families of children requiring the interventions will be factored into the analysis.

Using this data we will model different scenarios of providing the intervention within the context of standard NHS care. We will develop hypothetical scenarios of service provision mapped on to the intervention elements and will aim to illustrate the cost of the intervention, and the potential budget impact of delivering it at scale.

22. SWAT

The trial will include two SWATs, one related to sampling methods and another related to health economics measurement. These are both established methodological uncertainties.

22.1. Sampling SWAT

The Cochrane recruitment and retention reviews (both led from Aberdeen) found very little evidence for strategies targeting under-served groups and the limited evidence available is methodologically poor. There are very few SWATs from paediatric trials, and scarcity of evidence on effective recruitment and retention in paediatric trials. The Trial Forge SWAT Network highlighted recruitment and retention of under-represented populations as a SWAT priority (https://www.trialforge.org/2024/02/a-list-of-11-priority-recruitment-and-retention-swats/. Greater inclusion of under-represented populations is also an NIHR EDI strategic and operational priority.

We will include at least one SWAT targeting recruitment of CHESS priority under-represented sub-populations (see Sampling, Section 9), most likely people experiencing socioeconomic disadvantage. Depending on the qualitative tailoring work coming from our 6-month set-up and tailoring phase, we may include a second SWAT evaluation. Examples interventions could be video-based or interactive layered participant information provision. By layered we mean explicitly understanding and delivering the information that is most important to participants, which may vary by participant group, allowing them to regulate the level of detail they want. Use of video is a priority coming from the SWAT Network prioritisation work because videos are already being used despite limited evidence to support or refute their use, they offer the potential to better support individuals with lower literacy and may work better for some ethnic groups that prefer

verbal over written information. Key to our SWAT design is use of the INCLUDE Socioeconomic Disadvantage and Ethnicity Frameworks and stakeholder-informed content development, both of which will come from the set-up and tailoring phase. The measured outcomes will be recruitment, retention and cost.

We will share our protocol(s) both through the Belfast SWAT Repository and with the York Trial Forge SWAT Centre to support potential replication in other host trials they work with.

22.2. Targeted sampling SWAT

The first sampling SWAT intends to determine if targeted sampling approaches increase the number of children living in socioeconomically disadvantaged areas who take part in trials.

This SWAT will evaluate alternative ways of trying to ensure that children living in socioeconomically disadvantaged areas are well-represented with the CHESS population. Living in socioeconomically disadvantaged areas means living in areas of the UK with postcodes that fall into deciles 1 to 4 inclusive of the Index of Multiple Deprivation (IMD), or its equivalent depending on which nation within the UK a person is living.

When the SWAT is implemented, clusters will be randomised either to simple random sampling (our standard approach), as detailed in section 9.3 (control) or targeted sampling (intervention). Randomisation will be stratified by allocation group. Within each allocation group, block randomisation will be carried out to ensure equal allocation.

For targeted sampling, the cluster PI, or their delegate, will identify and make available the full, non-identifiable caseload list of potential participants to CHaRT at the central trial office. This list will consist of a non-identifiable ID and a postcode for the child. This anonymised table of information will be provided in an excel file format (or similar) using a secure file transfer system (such as a password protected encrypted file/ZendTo). The trial statistician will import the tables to STATA v18 for targeted sampling. Appropriate tools will be used to obtain deprivation data for the postcodes provided by the sites. Eligible children living in IMD 1 to 4 areas will be given preference to be invited to take part in CHESS ahead of children living in IMD 5 to 10 areas. In order to do this, a dummy variable called *IMD_cat* will be generated where IMD 1 to 4 will be coded as 1 and IMD 5 to 10 will be coded as 2. The data will be split by *IMD_cat* and saved into two datasets (IMD1 and IMD2). A random number using *runiform()* in STATA will be generated and assigned to children in both the datasets separately. The list will be sorted by the assigned random numbers in both the datasets. Using the IMD1 dataset, IMD2 dataset will be appended which will result in children in IMD1 dataset at the top of the list and IMD2 at the bottom of the list. We will then generate a dummy variable called *n* to number the participants from the first child to the last child in the list. A table which includes unique code, postcode, IMD and *n* which has been generated will be exported into excel file. A verification process checking for duplicates or errors in the selection process will be carried out. The trial statistician will send the excel file back to the PI/delegate using a secure file transfer system (such as ZendTo). The PI/delegate will invite the parents of the sampled children to participate in data collection in CHESS, beginning with the randomly assigned first child on the list and continue through the list until the cluster target sample of 24 parents have agreed to participate in the data collection and follow up in CHESS, or the full list exhausted. The cluster PI will receive guidance from the research team as required.

The details of the sampling process will be recorded including the date, method used, and any issues encountered. A copy of the list will be maintained in University of Aberdeen secured network drive.

22.3. Health economics outcome measurement SWAT

While all the health measures we propose to use have been intensively tested and are widely used across populations (with some of the measures being evaluated in thousands of studies in different populations to date), evidence of their psychometric properties in the context of trials in the present population is limited. This specific SWAT will aid interpretation of the findings of this trial, but also support researchers in the design of future trials in this or similar participant groups.

As part of this SWAT, we will assess the basic measurement and psychometric properties of the health economic measures (CHU9D, SF-36, and ICECAP-CES) at 12-month follow-up; 12 month data will be used as baseline measures are taken prior to cluster randomisation. Assessment will include the percentage of completed measures with missing data; floor/ceiling effects; test-retest reliability; convergent/divergent validity; agreement and known-group validity

Test-retest evaluation will require re-administration of the 12-month measurements 2 weeks after initial data collection, and will be collected from a 20% subset (n= 192) of total participants. Similarly to the DCE (section 21.3) we will select a subset of intervention and control clusters to undertake test-retest measurements, these clusters will be separate to the DCE clusters to ensure participants are not unduly burdened with data collection.

Provisional measurements of percentage of completed measures with missing data will be evaluated following baseline and 6-month period of the pilot trial, to highlight any data collection issues, with full SWAT evaluation completed after 12-month data collection for the full trial.

Primary subgroups (control and intervention) and secondary subgroups (sex, mobility, and socioeconomic deprivation) will also be evaluated for percentage of completed measures with missing data, floor/ceiling effects and test-retest reliability.

We will pay specific attention to evaluation of the CHU9D, as work is ongoing in validating it for children in our target age group.

We will examine correlation and agreement between outcome measures using assessment of construct validity (i.e. convergent/divergent validity) and Bland-Altman plots of agreement. These assessments will be undertaken to determine the extent to which these tools are measuring the same underlying constructs and are in sufficient agreement to be used interchangeably to measure the same construct. We will undertake known-group validity assessment to examine the ability of these measures to distinguish distinct groups where we would expect to observe differences between specific groups.

23. Trial management and oversight

The trial will be led by an expert, multidisciplinary team and will be run under the auspices of the Centre for Healthcare Randomised Trials (CHaRT), a fully registered UK Clinical Research Collaboration Clinical Trials Unit based in the University of Aberdeen. CHaRT has internationally recognised expertise in the design, conduct, analysis, and reporting of multicentre trials and has significant experience in Cluster RCTs. Sheffield Children's Trust will host the clinical allied health lead (SA) and be study Sponsor; and Newcastle University will host the CI (Kolehmainen) who has worked closely with both the Aberdeen and Sheffield colleagues, and with all study Co-Is, on previous projects. A Project Management Group (PMG), an independent Trial Steering Committee (TSC) and an independent Data Monitoring Committee (DMC) will be convened.

23.1. Trial office in Aberdeen

The Trial Office is in the Centre for Healthcare Randomised Trials (CHaRT) based at the University of Aberdeen and provides day to day support for the clinical centres. The Trial Manager(s) in CHaRT will take responsibility for the day-to-day transaction of trial activities, for example approvals, cluster set-up and training, intervention material supplying, oversight of recruitment and follow-up rates etc. The data co-ordinator will provide clerical support to the trial, including organising all aspects of the postal questionnaires (mailing, tracking, and entering returned data using the secure trial web data entry portal) and supporting the research with questionnaires that need to be administered by phone.

The Trial Office Team will meet formally at least monthly during the course of the trial to ensure smooth running and troubleshooting.

23.2. Local organisation within clusters

The local cluster PI, and their designated persons, are responsible for all aspects of local organisation including identifying potential recruits, consenting, completing and maintaining appropriate documentation.

The site agreement documents the full list of responsibilities for clusters. Appropriate members of the local team are knowledgeable about the Protocol and will have appropriate Good Clinical Practice (GCP) training if applicable. A trial-specific delegation log is prepared for each cluster, detailing the responsibilities of each member of staff working on the trial. The local team is also responsible for notifying incidents recorded on DATIX to the Trial Office (see section 15).

23.3. Project Management Group (PMG)

The trial is supervised by its Project Management Group (PMG). This consists of the grant holders and representatives from the CHaRT Trial Office, the Process Evaluation and Health Economic teams. Observers may be invited to attend at the discretion of the PMG. The PMG will meet approximately monthly within the first and last six months of the trial, and quarterly in between, with additional meetings arranged if required.

The grant holders form an experienced, established multidisciplinary team of clinical and methodology experts, with strong working-relationships between most applicants from previous projects, some extending for a decade. Furthermore, several of the applicants (Kolehmainen, Armitage, McAnuff, Pennington, Rapley, Marshall) have, for the past eight years, worked together to develop evidence-base for self-care support for children with neurodisability, paving the way for the present trial. As a result, the expertise and roles, and the relationships, are well refined, with clear contributions and responsibilities.

Kolehmainen, is a senior allied health clinical researcher with broad expertise and experience across: neurodisability and population child health; complex intervention methods (qualitative and quantitative) from evidence synthesis to modelling to intervention development to evaluations; and allied health and behaviour change interventions. She has substantial experience of leading multidisciplinary collaborations, including multisite longitudinal studies; and of working on community-based research studies with NHS organisations across England and Scotland. She has a track record of over a decade in contributing to national and regional allied health, neurodisability and wider child health capacity building; and has extensive networks across the related ecosystems for promoting the trial and translating the results to change. As the CI, Kolehmainen will lead, in close partnership with the Aberdeen CTU (MacLennan), on all aspects of the trial protocol, governance, coordination, cluster recruitment and training, reporting, trial promotion, and dissemination.

MacLennan, as a highly experienced triallist, statistician and the CTU Director, will take the lead on the trial methodology as well as providing senior statistical leadership. His expertise is complemented by Rapley (qualitative and mixed methods, implementation science) and Treweek

(efficient trial design, complex intervention trials, recruitment and retention interventions) who will jointly lead on the tailoring and optimising of the trial design. Rapley also leading on the process evaluation and supporting dissemination and implementation; while Treweek will lead on the methods for involving underrepresented populations (SWAT).

This methods expertise is further complemented by Vale, a highly experienced health economist with expertise in economic evaluations within trials and in economic modelling; and by Bray with expertise specifically in measuring economic outcomes in children with neurodisability. In the study, Vale will be the overall senior health economics lead, and working with Robinson (day-to-day lead for the cost-effectiveness evaluation), will lead the within trial economic evaluation and related modelling. Bray will provide expertise on the measurement methods, and will lead the budget impact analysis.

From clinical side, Armitage, as a senior paediatric occupational therapist with substantial self-care research experience, will be the clinical lead for the trial, working closely with Kolehmainen. Together they will formally recruit and train the clusters, supporting intervention delivery. With close links to the professional networks, Armitage will also lead dissemination to clinical and professional networks, with support from Kolehmainen and other clinical co-applicants.

Sutton, as a parent of a young person with neurodisability, an experienced health and care services community organiser and an NIHR-trained PPIE reviewer, will be the PPIE Lead for the study. As a parent, he has always provided the strongest possible voice for children and families affected by neurodisability. He co-chaired a strategic partnership board to improve services in his West Yorkshire locality; founded Disability Rocks, a music and arts-based organisation that hosted successful day festivals for disabled people otherwise unable to access festivals; and is passionate about achieving the best possible outcomes for children with neurodisability. He will direct and convene all child, young person and parent involvement and engagement activities throughout the study. In this, he will work closely with McAnuff and Morris, both experienced PPIE facilitators with nationally recognised track records in paediatric research; Morris runs the PenCRU Family Faculty. Sutton, McAnuff and Morris will work closely with Rapley and Treweek on tailoring and process evaluation.

McAnuff (occupational therapist in learning disability) and Morris (former orthotist), alongside Pennington (speech and language therapist), Parr (paediatric neurologist and neurodisability consultant), Marshall (physiotherapist) and Allen (health visitor) also offer senior multidisciplinary clinical and clinical academic neurodisability expertise for the trial. McAnuff and Pennington, as co-leads in developing the intervention, will support Kolehmainen and Armitage in cluster recruitment, training and retention. Marshall and Allen will provide senior, front-line community physiotherapist and health visiting advice throughout, from tailoring of the trial processes to interpretation and dissemination. Parr, as one of few neurodisability clinical academics in the UK with experience in paediatric trials, will provide further senior support for Kolehmainen as well as advice on any medical (neurodisability, neurology) aspects of the trial, including safety.

23.4. Trial Steering Committee (TSC)

A Trial Steering Committee (TSC), with independent members, oversees the conduct and progress of the trial. It is anticipated that the TSC will meet at least annually, with the first meeting before recruitment begins. The TSC Charter documents the terms of reference of the TSC, the template for reporting and the names and contact details of members of the TSC. This Charter is filed in the Trial Master File (TMF).

23.5. Data Monitoring Committee (DMC)

The DMC will be independent of the TSC and study co-applicants. It will monitor accumulating trial data during the trial and make recommendations to the TSC as to whether there are ethical or safety issues that may necessitate protocol modification or trial closure. It is anticipated that the DMC will meet at least annually, with the first meeting before recruitment begins. The DMC

Charter documents the terms of reference of the DMC and the names and contact details of members of the DMC. This Charter is filed in the TMF.

23.6. Patient and Public Involvement (PPI)

Our PPIE lead is Richard Sutton, parent of a young person with complex neurodisability. Richard is supported by Dr Jennifer McAnuff and Professor Christopher Morris, both with extensive experience in facilitating PPIE across populations, including involvement of children and young people with complex disabilities (incl. communication, cognitive and/or physical impairments). To achieve diversity and inclusivity within our PPIE, we are working with charity partners who are experts in engaging under-represented populations; and are using the NIHR INCLUDE Ethnicity and Socioeconomic Disadvantage Frameworks to further guide us.

We will engage parents, children and young people, and wider stakeholders such as service providers with expertise in engaging under-represented populations. PPIE meetings will be timed alongside key trial phases and milestones, will be organised and facilitated by Richard Sutton, Jennifer McAnuff, and Family Faculty colleagues at the Peninsula Childhood Disability Research Unit (PenCRU), and will encompass in-person, online, and creative methods. We will support PPIE contributors by identifying individual learning needs, providing informal assistance and encouragement, and providing formal training as needed. PPIE activities will also be supported by a dedicated PPIE Coordinator at the University of Aberdeen (expertise in PPIE within trials). Our PPIE budget includes acknowledgement payments for people's time, hospitality, subsistence, childcare, travel, costs for personal assistants where needed, and costs for producing accessible materials.

24. Research governance, data protection, and sponsorship

24.1. Research Governance

CHaRT is a fully registered Clinical Trials Unit with particular expertise in running multicentre RCTs. The trial will be run under the auspices of CHaRT, University of Aberdeen. This aids compliance with Research Governance and the principles of GCP, and provides centralised trial administration, database support and statistical analyses. CHaRT SOPs will be followed.

The CI and Sponsor ensure that adequate systems are in place for monitoring the quality of the trial and that reports are prepared to a level appropriate to the risk assessment of the trial.

24.2. Data protection

Data collected during the course of the research is kept strictly confidential and accessed only by members of the trial team. Data may be looked at by individuals from the Sponsor organisation or NHS clusters where it is relevant to the participant taking part in this trial.

The CI and trial staff involved with this project will comply with the requirements of the General Data Protection Regulations (GDPR) and the Data Protection Act 2018. The HRA recommended wording to fulfil transparency requirements under the GDPR for health and care research has been included in the PIL.

Trial staff based in Scotland will also adhere to the current version of the NHS Scotland Code of Practice on Protecting Patient Confidentiality.

Access to collated participant data will be restricted to the CI and appropriate trial staff.

Computers used to collate the data will have limited access measures via usernames and passwords.

Remote access to the network will be subject to robust authentication, and VPN (Virtual Private Network) connections to the network are only permitted for authorised users, ensuring that use is authenticated, and data is encrypted during transit across the network.
No personal data will be downloaded or stored on local hard drives. All data input/access will be via the VPN and/or secure website.

Use of the PEDI-CAT questionnaire can only be completed online through a secure web-based server owned by Pearson Clinical. All collected data is securely stored on Pearson Clinical servers situated in Montreal, Canada. When transferring personal data from the United Kingdom (UK), , Pearson relies on the principles recognized under the General Data Protection Regulation (GDPR), particularly the adequacy decision by the European Commission (EC) and UK Secretary of State (SoS). Date of birth and gender will be required to complete the questionnaire. No patient names or contact details will be shared with Pearson Clinical. Once the questionnaire has been completed online, a report and analysis will immediately be transferred to the trial team. Once this report has been received, participant data will be permanently deleted from the Pearson Clinical server.

Published results will not contain any personal data that could allow identification of individual participants.

The CHaRT senior IT development manager (in collaboration with the CI) manages access rights to the data set. Participants are allocated an individual trial number which is used to identify questionnaires and case report forms.

We anticipate that anonymised trial data may be shared with other researchers to enable international prospective meta-analyses.

24.3. Sponsorship

Sheffield Children's NHS Foundation Trust is the Sponsor for the trial.

25. Ethics and regulatory approvals

Our planning of the study follows, and takes a full account of, principles set out in the UK Policy Framework for Health and Social Care Research as well as the principles set out by children and families in the guidance for research involving children published by The Nuffield Council of Bioethics.[33] The NHS Research Ethics Committee and any appropriate NHS R&D approvals will be obtained prior to the commencement of trial recruitment. The trial will be conducted according to the principles of good clinical practice provided by Research Governance Guidelines. Annual progress reports (if required), end of Trial declaration, and a final report are submitted to the Sponsor and the NHS REC within the timelines defined in the regulations.

25.1. The main ethical issues and risks, and our plans for mitigating them

- The trial seeks to recruit families of whom many may be experiencing high levels of stress and scarcity of resource (time, energy, money, social support). The benefits of the trial need to be carefully considered with the potential further burden, and the related risk needs to be explicitly mitigated against (e.g. by minimising family time spent on trial processes, by training staff to be sensitive to family needs and preferences, and by language used in materials and data

collection). We will build on strategies, protocols and materials successfully applied in our previous studies, working very closely with the PPIE lead and stakeholders to refine and further tailor the trial procedures and materials, as well as staff training. We will also establish a 24h trial telephone line through which the participants can contact one of the clinical members of the trial team for any concerns or questions they have – based on previous experience, we expect very few calls, but evidence suggests people feel better when they know they have an option to contact someone.

- The trial will very likely involve clusters that are relatively research naïve, with many therapists inexperienced in discussing research with their patients or taking informed consent, with the usual models of working with research nurses unlikely to be feasible at some clusters. To overcome these issues, and to support these under-represented teams to participate, we have included substantial support from the CI, the clinical lead (SA) and the trial team for recruitment, consent and data collection; minimising the need for these activities on the ground. We have discussed this with all the clusters enlisted so far, and they have reported this as one of the decisive factors enabling their potential participation in the trial.

With the supports and considerations in place, we do not anticipate there to be specific risks to the children, parents or therapists. The benefits of the trial are likely to be for future children and parents in terms of informing the most appropriate therapy to use for individual patient outcomes. Impact on therapy practice, service planning and costs will also be future benefits, although likely to cover most of the therapists participating in the trial assuming they will remain within the workforce.

25.2. Protocol compliance and amendment

The Investigators will conduct the trial in compliance with the Protocol given favourable opinion by the Newcastle and North Tyneside 1 Ethics Committee (ref. no. 24/NE/0162)Research Ethics Committee. Any amendment to the Protocol or other approved documents will be reviewed by Sponsor (and funder where appropriate) before submission to REC and R&D unless in the case of urgent safety measures when the Sponsor is notified as soon as possible. Sponsor will advise if an amendment is substantial / non-substantial. Any deviations from the Protocol will be fully documented.

26. Monitoring and audit

The trial is monitored to ensure that it is being conducted as per protocol, adhering to Research Governance, the principles of GCP, and all other appropriate regulations. The approach to, and extent of, monitoring is specified in the trial monitoring plan and is appropriate to the risk assessment of the trial. Investigators and their host institutions are required to permit trial related monitoring and audits to take place by the Sponsor (or on behalf of the Sponsor by the CTU) and/ or regulatory representatives, providing direct access to source data and documents as requested.

26.1. Risk assessment

An independent risk assessment has been carried out on behalf of the Sponsor.

27. Finance and insurance

The trial is funded by a grant awarded by the NIHR Health Technology Assessment (HTA) programme (project number NIHR156487). The necessary trial insurance is provided by the Sheffield Children's NHS Foundation Trust.

28. End of trial

The end of follow-up for each participant is defined as the final data capture on that individual. The end of the trial is defined as the end of funding.

The end of the trial will be reported to the Sponsor and REC within 90 days, or 15 days if the trial is terminated prematurely. If terminated prematurely, the Investigators will inform participants and ensure that the appropriate follow up is arranged for all involved, if appropriate.

A summary report of the trial will be provided to the Sponsor and REC within one year of the end of the trial. An end of trial report will also be issued to the funders at the end of funding.

29. Data handling, record keeping and archiving

Data will be collected and stored in compliance with the local standard operating procedures (i.e. participating sites SOPs, CHaRT CTU SOPs, and collaborating institutions SOPs).

Clinical data will be collected on hardcopy CRF forms. These clinical data forms will then be input into the bespoke study database by the designated team members working in each recruitment cluster using a secure, electronic, web based data capture system. If members of the study team prefer to carry out direct data entry into the study database (i.e. not complete hard copy CRFs), this will be acceptable and the electronic data capture will be the source data.

The designated team members working in each recruitment cluster will enter data from questionnaires completed at clinic, or return to the trial office to be entered there.

Questionnaires returned by post to the trial office will be entered there. Staff in the trial office will work closely with local team members to ensure that the data are as complete and accurate as possible. Extensive range and consistency checks will further enhance the quality of the data. Responsibilities for archiving are documented in the co-sponsorship / site agreement.

We intend to follow-up the whole cohort for 12 months. All essential data and documents (electronic and hard copy) will be retained for a period of at least 10 years after close of trial according to funder requirements and relevant Sponsor and CHaRT archiving SOPs. It is anticipated, and consent will be sought to allow collection of longer-term data on health resource usage. Documents will be reviewed by CI before being destroyed. Electronic data will be archived by UoA.

30. Satellite studies

It is recognised, that the value of the trial may be enhanced by smaller ancillary studies of specific aspects. Plans for these will be discussed in advanced with the PMG and, if appropriate, with the TSC, Funder and Sponsor. Depending on the nature of the satellite trial, the Sponsor may consider this to be a non-substantial or a substantial amendment to the REC approval for the CHESS trial, or to require REC approval as a project in its own right. R&D management approval may also be required. In such situations, the sponsor will be contacted for advice.

31. Dissemination

31.1. Overview

The trial will result in a range of outputs.

Academic publications and presentations: The primary academic outputs of the CHESS trial will include a report for the funder (synopsis) and high-impact open access peer-reviewed journal publications on the clinical and cost outcomes associated with a strategy of early self-care support to improve self-care in young children with neurodisability versus usual care. We will target key journals such as NEJM and Lancet. We will present the clinical findings at the main UK and international paediatric conferences (e.g. Royal College of Paediatrics and Child Health, International Alliance of Academies of Childhood Disability), and methodological learning at relevant conferences (e.g. International Clinical Trials Methodology Conference).

Clinical and service guidance: We will work closely with the relevant professional organisations to decide the best formats (e.g. NIHR Signals, Evidence Spotlights, NICE accredited Practice Guidelines) for the trial results to inform clinical practice and service delivery. We anticipate the Royal College of Occupational Therapist (RCOT) to take a joint-lead with us on this, further working closely with The Chartered Society of Physiotherapy (CSP), Royal College of Speech and Language Therapists (RCSLT) and British Academy of Childhood Disability (BACD).

Potential new health care intervention, and related implementation toolkit and workshops: We are committed to making the manualised intervention and all materials openly accessible. If the intervention is effective, these will be accompanied by an implementation toolkit (informed by the process evaluation) and a design for workshops for children's therapy services. We will work with the professional body partners (RCOT, CSP, RCSLT, BACD) to develop these, and design the most accessible channels for sharing and dissemination.

Approaches to enhance equity and inclusion: The design and implementation of our more inclusive engagement principles and practices (e.g. health literacy, accessibility, formats) to recruitment and retention of families has implications beyond trials processes. We will share our learning and work with professional body partners and services (within trial and beyond) in how to make services more meaningful and accessible to a wider range of parents.

Creative outputs for families: We will develop creative ways to share the results with children and families. We will draw on our expertise in creative co-production methods with children with neurodisability, recognised as good practice by NIHR in 2019 (Co-production in Action: Number One. Southampton, INVOLVE: https://www.invo.org.uk/wp-content/uploads/2019/07/Copro_In_Action_2019.pdf).

Paediatric trials methods: We will register the SWATs with the Belfast SWAT repository. We will publish our SWAT as a brief publication, as well as disseminate through Trial Forge, the NIHR Incubator for Applied Health Research Methods (incl. paediatric workstream), and MRC-NIHR Trial Methodology Research Network. Our second SWAT will directly address the dearth of information on methods for assessing HRQoL in young children. Using the data collected within the trial we will report basic measurement and psychometric properties of the tools (see economic evaluation, above). We will publish this SWAT in a similar way as outlined above.

We will make all outputs open access.

31.2. Authorship and dissemination

To safeguard the integrity of the main trial, reports of explanatory or satellite studies will not be submitted for publication without prior arrangement from the PMG.

Publications will in most cases be *led by* individual investigators; however, all publications (any format) resulting from the trial, including any preparatory PPIE work, the process evaluation, and the health economics, will be *co-ordinated through* the PMG to ensure effective co-ordination, cross-linkage, and cross-referencing, as well as to avoid duplication of effort. In general, the PMG is not expected to interfere with preparation of the content of individual publications.

DEFINING AUTHORSHIP

Authorship of published or presented papers is based on the International Committee of Medical Journal Editors (ICMJE) criteria:

i. Substantial contributions to the conception or design of the work; or the acquisition, analysis, or interpretation of data for the work; AND

ii. Drafting the work or revising it critically for important intellectual content; AND

iii. Final approval of the version to be published; AND

iv. Agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

In addition to being accountable for the parts of the work the author has done, they should be able to identify which co-authors are responsible for specific other parts of the work. In addition, authors should have confidence in the integrity of the contributions of their co-author.

PRINCIPLES OF AUTHORSHIP

The following principles of authorship have been derived from editorial publications from leading journals and are in accordance with the rules of the ICMJE.

All contributors must fulfil the criteria detailed in section 1: DEFINING AUTHORSHIP in order to qualify for authorship.

Contributors who meet fewer than all four of the criteria for authorship listed above should not be listed as authors, but they should be acknowledged. For example, participation solely in the acquisition of funding, collection of data or technical editing, language editing or proofreading the article is insufficient by itself to justify authorship1. Those persons may be acknowledged and their contribution described. See section 3: ACKNOWLEDGEMENTS.

The following rules may help with the ordering of authors, particularly for publications with individual authorship:

i. The person who has taken the lead in writing may be the first author.
ii. The senior author may wish to be the last named author.
iii. Those who have made a major contribution to analysis or writing (i.e. have done more than commenting in detail on successive drafts) may follow the first author immediately; where there is a clear difference in the size of these contributions, this should be reflected in the order of these authors.
iv. All others who fulfil the four authorship criteria described in Section 1: DEFINING AUTHORSHIP may complete the list in alphabetical order of their surnames.

These authorship criteria are intended to reserve the status of authorship for those who deserve credit and can take responsibility for the work. The criteria are not intended for use to disqualify colleagues from authorship who otherwise meet authorship criteria by denying them the opportunity to meet the ICMJE criterion numbers (ii) or (iii). Therefore, all individuals who meet the first criterion should have the opportunity to participate in the review, drafting, and final approval of the manuscript1.

Where possible, all the outputs from CHESS should be published using all the named contributors who qualify for authorship in the byline i.e. Jane Doe, John Doe, John Smith and Ann Other. In situations where this is not possible, for example if the journal limits the number of authors, group authorship can be applied, using bylines similar to “The XXXXX trial group” or “Jane Doe, John Doe, John Smith, Ann Other and the XXXX trial group”. Such outputs should carry a footnote of

the names of the people (and their institutions) represented by the corporate title. For some journals the journal will provide instructions on how to ensure the names of the collaborators appear on PubMed or equivalent. Group authorship may also be appropriate for publications where one or more authors take responsibility for a group, in which case the other group members are not authors but may be listed in the acknowledgement (the byline would read 'Jane Doe for the Trial Group') 2. Again, the article should carry a footnote of the names of the people (and their institutions) represented by the corporate title.

Tentative decisions on publications, dissemination, and authorship should be made as early as possible3. These should be justified to, and agreed by, the Project Management Group (PMG). Any difficulties or disagreements will be resolved by the Trial Steering Committee (TSC).

All those who contribute to a publication, but who do not fulfil the criteria for authorship, such as interviewers, data processors, staff at the recruiting clusters, secretaries and funding bodies, should be acknowledged by name, usually in an 'Acknowledgements' section specifying their contributions. Because acknowledgment may imply endorsement by acknowledged individuals of a trial's data and conclusions, authors are advised to obtain written permission to be acknowledged from all acknowledged individuals1. The acknowledgements should also reflect any agreed acknowledgements (for example with suppliers) that were documented in supply agreements (or equivalent).

Authors must ensure they include the appropriate trial funder's disclaimer: refer to the funders website for details. Be aware that other disclaimers may also be required.

QUALITY ASSURANCE

Ensuring quality assurance is essential to the good name of the CHESS co-investigator group. All outputs arising from the CHESS trial must be peer reviewed by the PMG. The PMG will be responsible for decisions about submission following internal peer review. Submission may be delayed or vetoed if there are serious concerns about the scientific quality of the report. If individual members of the group are dissatisfied by decisions, the matter may be referred to the TSC.

It is hoped that the adoption and dissemination of this policy will prevent disputes that cannot be resolved by informal discussion. However, any member of the trial team with a concern about authorship should discuss it with the Chief Investigator or TSC, as appropriate.

31.3. Informing patients, NHS and the wider population

Our sharing and engagement strategies build on our extensive experience and expertise - from projects such as NIHR ActiveCHILD, NIHR EMPoWER, NIHR FEEDS, NIHR Transition and the work by PenCRU on sharing research findings creatively with children of all ages (from the very youngest), young people, parents, NHS professionals and commissioners, policy makers, charities, and businesses.

We will flexibly use a variety of active approaches to involvement: enabling families' engagement from within their homes through creative, hands-on materials; online launches and workshops that incorporate dissemination by PPIE stakeholders (e.g. https://childresearch.co.uk/2020/11/27/empower-young-peoples-take-on-early-powered-mobility/); cross-sector workshops focusing on translation across policy, practice and business (e.g. NIHR Transition programme informed the NHSE Long Term Plan Transition strategy); and opportunities for national charities to co-lead on dissemination and consultation.

We will carefully design and tailor engagement of the study clusters, with sensitivity to their requests, needs and feedback. We have the expertise and resources to set up and support virtual and in-person meetings, either across clusters within each arm or in smaller geographical

clusters, or for each cluster individually. We anticipate the clusters' learning needs to vary across the phases and according to the starting level of the cluster experience; and our approach to support will be highly tailored to these variations and needs.

In parallel, we will employ the more traditional approaches of press-releases, participant newsletters, social media, trial website, written briefings, and professional networks and newsletters. Throughout, we will work closely with the NHS and PPIE stakeholders, and the professional bodies, to select and design the best strategies at each phase of the trial. Many of the co-applicants and collaborators sit on guideline and stakeholder committees related to this area. Where possible, we will prioritise activities that build on and extend existing work by stakeholders in order to maximise opportunities and impact; and work with the Newcastle University communications team where appropriate.

31.4. Disseminating outputs to health and care system, and society

Our strategy of introducing, embedding and sustaining the outputs to the health and care system will draw on both engaging and enrolling key people and organisations in this area.

We know that the support and engagement of families and the children's therapy services are central to implementing change on the ground. In discussing the trial with families and therapy services, they repeatedly tell us that the trial results have the potential to support them in a range of ways. For example, young people and parents really want to know what support would help them and want to see the NHS support for them improved. Therapists say the trial is: "a massive CPD opportunity"; "absolutely necessary to sort out the repeated, unhelpful tribunals"; "a way forward to moving from family centred goals to actually family centred self-care support"; and "an opportunity to get some actual evidence to inform what is our core practice". The clusters repeatedly ask for a firm commitment that they will have access to the research findings and materials post-trial, and that if CHESS is effective then training will be provided to them to adopt it.

A lack of implementation champions with a dedicated and sustained implementation infrastructure (akin to a pharmaceutical company in medicine trials) is a common barrier to further introduction and embedding of most complex intervention trials. We know that the enrolment of professional organisational and policy decision-making are central to creating contexts for change, and we are proactively doing this. Members of the core study team have worked on this topic since 2015 when the families first prioritised the topic and many of the co-applicants and collaborators sit on professional, guideline and stakeholder committees related to this area. RCOT already funded the pre-trial work and we will work closely with other professional body partners (e.g. CSP, RCSLT, BACD), advocacy organisations (e.g. Unique and LS29), to advocate change on the policy landscape.

We will actively seek to engage and enrol those with reach and influence across NHS professionals and decision makers including commissioners, service users and their advocacy organisations, the general public, and national guideline developers.

The acquisition of the equipment required to support self-care is one of the main known barriers. If the intervention is successful, for longer-term implementation, therapy teams will require support in acquiring the intervention materials and appropriate training. We are addressing this as part of the trial set-up and tailoring, including working with the equipment provides to set up loan-schemes that make the equipment more accessible for NHS organisations than what is currently possible. We are also making all possible materials open access, including specific resources to support training.

31.5. Anticipated impact

The research is expected to have an impact on all major stakeholders involved in the management of children with neurodisability. For children, families and therapists, the study will provide much needed high-quality unbiased information on the relative clinical and cost-effectiveness of CHESS compared to usual care. This will have the potential to guide evidence-based decision making. For health policy makers and funders, cost-effectiveness data will be vital in planning future NHS/ public funded strategy around care of children with neurodisability.

There are currently no published guidelines or best practice recommendations for self-care in children with neurodisability, and organisations such as NICE are currently unable to make recommendations on this topic because of the lack of trial data. This trial will provide, for the first time, randomised large-scale evidence to inform recommendation about how to support children with developmental disabilities to gain independence in self-care and activities of daily living. This can be expected to be incorporated into clinical practice guidelines and treatment recommendations from NICE and in updates of relevant Cochrane reviews; and to give care providers greater confidence in deciding where they should put resources.

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