ADAPT

Airway Drainage And Positioning at Night-Time

51827#]

A feasibility randomised controlled trial of a novel postural management night-time intervention ("Breathe-Easy") to improve respiratory health of children with complex neurodisability

PROTOCOL VERSION NUMBER AND DATE

Version 3.0 22-10-2025

SPONSOR

Sussex Community NHS Foundation Trust

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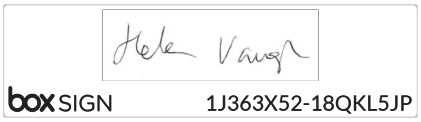
SIGNATURE PAGE

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 and will adhere to the principles outlined in the Declaration of Helsinki, GCP guidelines, the Sponsor's (and any other relevant) SOPs, and other regulatory requirements.

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.

For and on behalf of the Sponsor:

Signature: 

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Date: 16 Feb 2026

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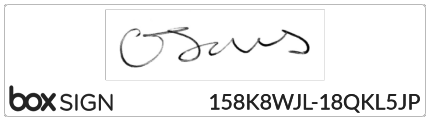
Statistician: 

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KEY TRIAL CONTACTS

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Committees	Trial Management Group (TMG) Joint Trial Steering Committee (TSC) & Data Monitoring and Ethics Committee (DMEC) Patient and Public Involvement (PPI) Co-Investigators/Co-applicants meetings

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LIST OF ABBREVIATIONS

AE	Adverse Event
ADAPT	Airway Drainage and Positioning at Night-Time
BSCTU	Brighton & Sussex Clinical Trials Unit
BRSQ	Bespoke Respiratory Symptoms Questionnaire
CI	Chief Investigator
CONSORT	Consolidated Standards for Reporting Trials
CRF	Case Report Form
CHU9D	Child Health Utility 9D
CSHQ	Child Sleep Habits Questionnaire
CTU	Clinical Trials Unit
CPAP	Continuous Positive Airway Pressure
CYPCN	Children and Young People with Complex Neurodisability
DM	Data Manager
DMEC	Data Monitoring and Ethics Committee
EDACS	Eating and Drinking Classification System
FiO_2	Fraction of Inspired Oxygen
FMS	Functional Mobility Scale
GCP	Good Clinical Practice
GJ	Gastro-Jejunal tube for feeding
GORD	Gastro-oesophageal reflux disease
GMFCS	Gross Motor Function Classification System
ICF	Informed Consent Form
ICH	International Council for Harmonisation of Technical Requirements for Pharmaceuticals for Human Use
ISF	Investigator Site File (This forms part of the TMF)
ISRCTN	International Standard Randomised Controlled Trials Number
HCP	Healthcare Professional
HRA	Health Research Authority
HTA	Health Technology Assessment
LRTI	Lower respiratory tract infection
NHS R&D	National Health Service Research & Development
NIHR	National Institute for Health and Care Research
NIMP	Non-Investigational Medicinal Product
Non-CTIMP	Studies not using investigational medicinal products
pCO_2	Partial Pressure of Carbon Dioxide
PenCRU	Peninsula Childhood Disability Research Unit
PI	Principal Investigator
PIS	Participant Information Sheet
PPI	Patient and Public Involvement
QA	Quality Assurance
QC	Quality Control
QP	Qualified Person
RE	Related Event
RCT	Randomised Controlled Trial
REC	Research Ethics Committee
rFLACC	Face, legs, activity, cry, consolability scale
RfPB	Research for Patient Benefit
RR	Respiratory Rate
RSI	Reference Safety Information
SaO_2	Oxygen Saturation
SAE	Serious Adverse Event

SCFT	Sussex Community NHS Foundation Trust
SDV	Source Data Verification
SOP	Standard Operating Procedure
SPS	Sleep Positioning System
SmPC	Summary of Product Characteristics
SRE	Serious Related Event
SSI	Site Specific Information
SUSRE	Suspected Unexpected Serious Related Event
TIDieR	Template for Intervention Description and Replication
TM	Trial Manager
TMF	Trial Master File
TMG	Trial Management Group
TSC	Trial Steering Committee
YPAG	Young Persons (Research) Advisory Group (Kent Surrey & Sussex)

TRIAL SUMMARY

Trial Title	A feasibility randomised controlled trial of a novel postural management night-time intervention (Breathe-Easy) to improve respiratory health of children with complex neurodisability	
Short Title	ADAPT: **A**irway **D**rainage **A**nd **P**ositioning night-**T**ime	
Trial Design	Feasibility randomised controlled trial	
Trial Participants	Children aged 2 to 18 years, who are: • dependent upon others to position/move their bodies, equivalent to Gross Motor Function Classification System (GMFCS) levels IV-V. • at high risk of aspiration linked to swallowing difficulties, equivalent to EDACS levels IV-V. • fed via gastrostomy/jejunostomy. • at least one lower respiratory tract infection requiring antibiotics in past 12 months.	
Planned Sample Size	50 Children 50 Parents/Guardians (Survey) 50 Health Care Professionals (Survey) 10 Children (Interview)	
Intervention duration	6 months	
Follow up duration	6 months	
Planned Trial Period	Two-year study 1 January 2025 to 31 December 2026. Regulatory approvals by month 3. Recruited participants by month 15. Data collection completed by month 21. Data analysis and report by month 24.	
	Objectives	Outcome Measures
Primary Feasibility Objectives and Outcome Measures	1. To evaluate recruitment and retention	- 1a) Recruitment: Proportion of recruited children amongst eligible participants and reasons for not participating. - 1b) Retention: Proportion of participants that completed the feasibility trial out of the number randomised.
	2. To assess feasibility of recording candidate primary outcome measures	- 2a) Proportion of data collection (from candidate primary outcomes) complete per participant.
	3. To estimate the variability of candidate primary outcome measures	- 3a) Standard deviation of the candidate primary outcomes.
	4. To determine design characteristics for a subsequent definitive study	- 4a) Proposed design, sample size and number of centres for a definitive study.

	5. To assess the acceptability of novel night-time position for participants, families, health care staff.	- 5a) For participants: Number of nights in position for minimum 4 hours. - 5b) For families and healthcare staff: Survey (acceptability measure).

FUNDING AND SUPPORT IN KIND

FUNDER(S) **(Names and contact details of ALL organisations providing funding and/or support in kind for this trial)**	**FINANCIAL AND NON FINANCIAL SUPPORT GIVEN**
NIHR RfPB	**£280,723.00**

ROLE OF TRIAL SPONSOR AND FUNDER

Trial funder (NIHR RfPB): to provide funding to enable the trial to occur, to ensure that timelines are being met and that appropriate dissemination occurs.

The sponsor, Sussex Community NHS Foundation Trust (SCFT) will assume overall responsibility for the initiation and management of the trial ensuring compliance with all appropriate regulatory and ethical governance.

ROLES AND RESPONSIBILITIES OF TRIAL MANAGEMENT COMMITEES/GROUPS & INDIVIDUALS

Trial Management Group (TMG)

The TMG will consist of the joint Chief Investigators (CIs AK and SC), CTU representative (CTU), trial manager (TM), data manager (DM), statistician, Principal Investigators and site research teams from the sites.

The TMG will be responsible for the trial set-up, the day-to-day running of the trial and the release of any trial results or publications according to the BSCTU SOPs. The TM will be involved in setting up monthly TMG meetings which will oversee the management and conduct of the study. Recruitment and data updates will be discussed to highlight any issues and to ensure they can be resolved in a timely manner.

Joint Trial Steering Committee (TSC) & Data Monitoring and Ethics Committee (DMEC)

The joint TSC and DMEC will consist of the trial co-investigators, and trial manager together with independent members (including parents of a child with neurodisability, physiotherapists, an occupational therapist with relevant expertise, paediatrician with expertise in neurodisability, and statistician).

The frequency of meetings will be every 6 months, or as agreed in a charter, and the committee will receive reports from the TMG and oversee the progress of the study. With its independent

membership it will also review the data and any safety issues, fulfilling the role for this feasibility trial of a data monitoring and ethics committee (DMEC). Financial management for the study will be overseen by the R&D Manager at SCFT, and the CIs.

The Joint TSC & DMEC will have oversight of the trial conduct. The Committee's terms of reference, roles and responsibilities will be defined in a charter in accordance with the relevant BSCTU SOP.

PPI group

Patient and Public Involvement (PPI) throughout the study will be ensured by our PPI group, which will be facilitated by SC and GB and consist of 6-8 parents, together with 2 young people if possible, all with experience of severe neurodisability.

The PPI group will meet every 4 months and will provide disease-specific input into the study. Their role and involvement is described in section 11.3 below - Patient and Public Involvement (PPI).

Co-Investigators/Co-applicants meeting

This group will meet monthly until the study has opened and is successfully recruiting and will then meet every 2 months through the duration of the study, as is considered necessary.

PROTOCOL CONTRIBUTORS

All the protocol contributors named in the table at the top of the form have contributed to the development of the protocol in a series of investigator meetings.

PPI input has been provided by the PenCRU Family Faculty and Kent Surrey and Sussex Young Persons (Research) Advisory Group (YPAG) who have had significant influence on the protocol design, especially in the area of recruitment and provision of participant information.

KEY WORDS

Chest infections, neurodisability, postural management, night-time positioning, swallowing difficulties

1. BACKGROUND AND RATIONALE

Many children and young people with complex neurodisability (CYPCN) have cerebral palsy. Recent prevalence estimates of cerebral palsy (CP) for children and young people 0-25 years of age (2004-2014) are 3.5 (95% CI 3.4-3.6) per 1000 for England and 2.8 (95% CI 2.7-2.9) per 1000 for Wales [1]. In high income countries the overall incidence of CP is 2.0-2.5/1000 live births, and severe CP (GMFCS levels IV-V) occurs following 0.4/1000 live births [2]. These figures provide a guide, but they underestimate the number of CYPCN as there are many diagnoses other than CP that result in complex neurodisability. Whilst many more children and young people with chronic neurodisability now survive into adult life due to improvements in neonatal and general paediatric care, they continue to experience serious health problems including preventable premature death [1].

CYPCN are major users of different kinds of healthcare in primary, community, acute and tertiary specialist settings; they require proactive multidisciplinary care to treat and manage disabling health conditions. CYPCN may be dependent upon different medical and assistive technologies such as artificial feeding, suction of airways and assisted ventilation, as well as mobility aids and postural management equipment. They are vulnerable to all the medical and surgical conditions that can affect anyone, but these conditions can be more difficult to diagnose and manage in the presence of complex neurodisability [1]. CYPCN need more frequent GP visits, attend numerous outpatient clinics, and hospitalisations are more frequent and prolonged. The predominant cause of hospital visits and primary care use is for respiratory issues [1, 3-7].

CYPCN frequently experience lower respiratory tract infections (LRTI), causing repeated and lengthy hospitalisations, bronchiectasis and premature death [3, 8, 9]. The impact of these hospital admissions on health service expenditure is significant and places a huge burden on children, their families and healthcare services [10, 11]. A review of US paediatric hospital admissions in 2006 found that 29% of US children's hospital expenditure was on inpatient care of children with CYPCN, with >10% of these admissions being for respiratory tract infections [12]. The most common causes of emergency admissions are respiratory illness, malnutrition, dehydration, constipation, and seizures [7].

The burden and cost of emergency hospitalisations is high [13]. Respiratory illness is the most frequent and costly cause of un-planned, emergency hospitalisations for children and young people with cerebral palsy [13].

The associated overall mortality rate is also significant: in England it was 26 times higher for children and young people with cerebral palsy than for those without (5.3 vs 0.2 per 1000 at risk) for 0-25 year olds. The mortality rate was greatest in those younger than five years of age [1]. The most commonly recorded primary causes of death for children and young people with CP were respiratory causes, accounting for 51% of cases [1].

In summary, CYPCN with respiratory health issues are extremely vulnerable, with respiratory infections being the main cause for premature death. Aspiration pneumonia is recognised as the leading risk for worsening respiratory compromise [14]; aspiration occurs when saliva, stomach contents, food, or fluid enters the airway and lungs. Twenty-two percent of identified deaths for people with CP resulted from solids or liquids in their lungs or windpipe [15].

Thus, our study is addressing a key problem with high burden of morbidity and mortality in CYPCN and intense high use of NHS resources.

Current guidelines for night-time positioning for postural management

CYPCN with limited ability to change their body position independently are at risk of developing postural deformities and secondary complications [16, 17]. CYPCN commonly have difficulty with postural alignment and if asymmetrical postures are adopted constantly, this may result in tissue damage, pain, progressive loss of function and musculoskeletal deformity including hip dislocation and spinal curvature [16, 17]. The 24-hour postural management approach aims to minimise the development of these complications by utilising a range of interventions to reduce postural asymmetry and improve function by managing postures in sitting, standing and lying [18]. A consensus statement in 2006 proposed that children in GMFCS Levels IV to V should begin 24-hour postural management programmes in lying as soon as possible after birth, in sitting from 6 months, and in standing from 12 months [19].

Postural management in lying includes positioning at night-time as a significant number of hours are spent in bed each day and a more relaxed body when asleep may facilitate postural alignment. Many healthcare professionals view night-time positioning as an essential part of 24-hour postural management; the position a CYPCN adopts overnight may also influence their ability to be positioned during the day [20]. Symmetrical supine lying is reported to be the optimum postural management position at night-time for CYPCN, to balance the functioning of muscles and distributing load-bearing [21, 22]. A Cochrane review into night-time positioning supports a proposed optimal supine position

of hips with a symmetrical 30 degrees of hip abduction and 30 degrees of hip flexion [23]. However, no studies have investigated the optimum posture for lying from the postural management perspective. In contrast, side-lying or lying prone has been found to be more beneficial for respiratory function [24]. Healthcare professionals may therefore have different priorities when advising on positioning depending upon the primary focus of concern.

Despite strong healthcare professional support for the common approach of night-time postural intervention for CYPCN, there is a lack of robust evidence for its effectiveness. A recent review cited ten papers focused on lying and night-time positioning, but the majority described either clinical practice or theory of postural management rather than providing empirical evidence of effectiveness [20]. This review concluded that much of the reviewed literature is based on specialist clinical knowledge and expertise gained from clinician's experience of working with this population rather than empirical studies. There are critics of the 24-hour postural management approach who point out the challenges of CYPCN tolerating this specialist equipment, adverse impact on sleep, and consequently poor adherence to this type of programme [25]. The focus on postural management inadvertently overlooks night-time risks to respiratory health associated with swallowing difficulties [26].

Commercial equipment is often used to facilitate the optimal night-time lying position for CYPCN – this equipment is often called a 'sleep positioning system' (SPS). Consideration of the use of this type of postural support at night is recommended by the National Institute for Health and Care Excellence for children and young people with non-progressive brain disorders [16], although no evidence of effectiveness was cited. A Cochrane review of SPS for children with cerebral palsy did not identify any randomised controlled trials that evaluated the effectiveness of sleep positioning systems on hip migration [23]. Only two low-quality randomised cross-over trials were found in relation to sleep quality and pain. The review concluded that the quality of evidence was low and highlighted the need for more research to enable families and professionals to make decisions on whether this equipment should be used to prevent hip migration.

Another recent systematic review of sleep positioning systems for children and adults with a neurodisability that included non-randomised trials, ineligible for the Cochrane review, suggested some potential benefit for hip stability, improved sleep quality and quality of life [17]. However, the quality of the evidence was poor due to small numbers, lack of methodological rigour, missing data, or studies available only as conference abstracts. Night-time positioning for respiratory function was examined as part of this review. Four studies included in the review considered respiratory function but there was no clinically or statistically significant difference between respiratory function using or not using a SPS [17]. One study recommended that children should be screened for ventilatory

function using oxyhaemoglobin saturation and carbon dioxide measures prior to prescription of a sleep positioning system over three consecutive nights [27].

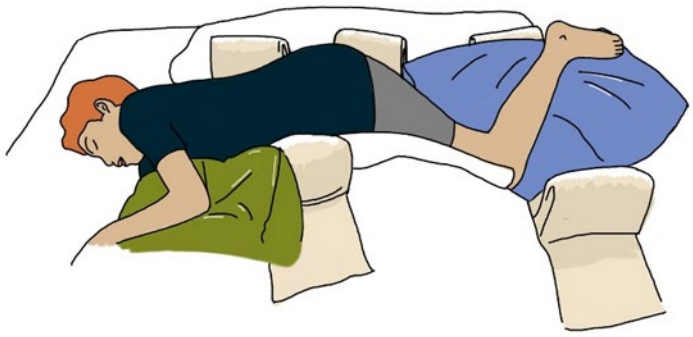
The Cochrane review emphasised the importance of considering the effects of SPS on respiratory function and the risk of aspiration [23]. However, there are potential conflicts between recommendations based on whether the focus is on respiratory and/or deformity.

The most recent guidelines for managing respiratory health in CYPCN have recently reviewed risk factors for the CYPCN population to ameliorate reversible factors, but none specifically focus on night-time positioning as our study proposes [26, 28, 29]. The consensus statement highlights that comprehensive clinical care recommendations for the management and treatment of respiratory illness does not exist, but that treatment should be proactive.

Novel night-time intervention ('Breathe-Easy' positioning)

The novel intervention (Breathe-Easy) to be used in this study is described with reference to the TIDieR checklist [30]; it is a manualised intervention whereby therapists support implementation with families and other carers. This is a night-time postural management intervention with the aim of draining upper airway secretions overnight. It involves the child lying semi-prone or on their side on a flat bed, supported by their usual night-time postural management positioning equipment or pillows. The exact position is individualised for each child to take account of safety, comfort, postural needs, and family preferences. The aim is to achieve quieter breathing and upper airway drainage of secretions out of the mouth using gravity. To reduce risk or prevent worsening of gastro-oesophageal reflux, CYPCN must be given no fluids at least one hour prior to implementation of the intervention. A drainage bag is attached to the gastrostomy to drain stomach contents overnight to minimise risk of gastro-oesophageal reflux. Fluid loss will be measured and replaced by oral rehydration salts the following day if required, to avoid electrolyte imbalance. The protocol allows for flexibility of use because of unstable health needs of CYPCN, e.g. illness, seizures or other surgical or medical issues. We anticipate that gaps in use of the novel night-time position will occur but will recommend a minimum of 4 hours per night.

Breathe-Easy novel night time intervention position example:



In a case series study, Breathe-Easy positioning initiated safely in a hospital setting, hastened discharge, reduced inpatient days and hospital readmission [31]. The limitations of this study were use of retrospective data and lack of detailed follow up (personal communication AK). We recently completed the Breathe-Easy proof-of-concept study to explore practicality of this complex intervention in an NHS community setting. Of eleven children recruited, eight (73%) completed this novel intervention for 6-months, adapted for home use. In interviews, CYPCN, parents and carers reported benefits to respiratory health, sleep and well-being. All families who completed the study have opted to continue the novel positioning post-trial. No harm or serious adverse events were noted, including musculoskeletal issues. The research was co-produced with a PPI group, who have also been heavily involved in the planning of this study. PPI was integral to the conduct of the recruitment strategy, refinement of the intervention and future plans to test the intervention in two steps; this feasibility trial, then if appropriate, for the definitive RCT.

In summary, there is evidence that overnight aspiration is a major preventable factor influencing quality of life and respiratory infections in CYPCN, and we have initial evidence suggesting that the novel intervention may be effective. Ultimately, a fully powered randomised controlled trial (RCT) is needed to evaluate the effectiveness and cost-effectiveness of the intervention, but a feasibility RCT is needed to help plan this, in order to optimise recruitment strategies, define outcomes and measures, and provide information necessary to design a definitive RCT. From the previous proof-of-concept study, we anticipate measurable improvements in respiratory health and sleep within 3-6

months. However, a future definitive RCT will likely need to run over 12 months (i.e. including one winter season) to assess definitively the impact on chest infections and hospitalisation, and to provide full reassurance about musculoskeletal concerns.

2. AIMS, OBJECTIVES AND OUTCOME MEASURES

2.1. Main Study Aims

This study has **two overarching aims**:

1. To evaluate whether the novel intervention can be delivered in the community, specifically:
    a) Assess acceptability of the intervention to CYPCN, families and clinical teams.
    b) Assess delivery of the intervention manual and training.
    c) Assess fidelity and adherence to written positioning guidance within intervention and control groups.
    d) Explore safety of ADAPT protocol for CYPCN, families and HCPs.

2. To provide information necessary to design a definitive Randomised Controlled Trial (RCT) to test the effectiveness and cost-effectiveness of the novel intervention, specifically:
    a) Assess feasibility of recruiting participants in different sites, across ethnically and socially diverse communities.
    b) Assess acceptability of randomisation to parents/guardians and healthcare professionals.
    c) Assess attrition / completion and proportion of any missing data in questionnaire measures.
    d) Test the feasibility of collection and appropriateness of candidate outcome measures from the stakeholder perspective.
    e) Appraise the performance of candidate health and wellbeing outcome measures in terms of acceptability to participants, and feasibility and interpretability for researchers.
    f) Estimate the variability (standard deviation) to inform the sample size calculation for the definitive trial.
    g) Assess feasibility of progression to a definitive trial and set out main parameters.
    h) Assess resource implications of the intervention and indications of offsetting service use savings.
    i) Explore acceptability and feasibility of using SOMNOTouch as objective measure in protocol. (limited to Sussex participants only).

Recruitment, randomisation and retention of all participants will be assessed from data recorded at

each site on number of:

- eligible CYPCN on caseload.
- families declining participation at different stages:
    - before randomisation,
    - after randomisation,
    - prior to establishing the novel intervention.
- families dropping out in intervention group, at what stage and reasons.
- families dropping out in control group, at what stage and reasons.

2.2. Primary Objective

The primary objective is to investigate the feasibility of conducting an RCT of novel night-time position intervention to improve the respiratory health of CYPCN. The underlying hypothesis is that the novel intervention reduces 'chest infections' and hospital admissions and improves quality of life for CYPCN.

2.3. Candidate Primary Outcome Measures

2.3.1. Proportion of CYPCN with chest infections

This will include number of oral or intravenous antibiotics within the trial period, including a comparison pre- and post- 6 months of recruitment. Prophylactic antibiotics will be reported as well.

2.3.2. Time to first 'chest infection'

For a diagnosis of 'chest infection' to be made, whether necessitating hospitalisation or not, CYPCN must have two or more of the following over a 48-hour period (based on clinical judgement and age-related definitions unless specified) (same definition as per the Parrot trial - ISRCTN - ISRCTN71955516: Prophylactic antibiotics to prevent chest infections in children with neurological impairment):

- Increased secretion volume/viscosity of respiratory secretions
- Change in temperature by 1°C or lethargy/fatigue
- Increased cough
- Increased work of breathing (tachypnoea or dyspnoea)
- Increased oxygen requirement
- Increased need for respiratory support (such as increased chest physio/suction/cough assist)

- Changes on chest X-ray or chest auscultation.

It is not feasible in this trial to differentiate between purely an acute viral or bacterial chest infection vs aspiration into lungs leading to breathing difficulty as there is an overlap in our cohort of CYPCN. Hence, we aim to capture both in our trial, including, antibiotic usage for both, under the term 'chest infection'.

We recognise that some participants will receive the intervention over the summer months when they are less likely to experience chest infections or have not recently experienced a chest infection. This seasonal effect should be minimised in the definitive RCT which will recruit participants over 1 year to cover all seasons.

2.3.3. Total time spent in hospital

Hospitalisation data, including reason for hospitalisation will be captured and categorised as chest infection related or other reason. Hospitalisation includes those who are admitted to hospital for only a short period with LRTI e.g. 12 hours and discharged with a course of antibiotics.

We will also be recording the number of hospital admissions. However, if participants are hospitalised again within 2 weeks of the initial admission, this will be classified as the same event.

2.3.4. Respiratory Health

Using the Bespoke Respiratory Symptoms Questionnaire (BRSQ) (adapted from [37,38] and used in Parrot trial https://parrot-trial.org.uk/) at baseline, 3-months and 6-months, we will investigate respiratory health over the course of the trial as perceived by the parents/guardians. In Sussex participants only, this will also be investigated using SOMNOtouch at baseline and at 3 months

2.4. Candidate Secondary Outcome Measures

- Sleep behaviour (CSHQ Abbreviated) [39]: validated sleep questionnaire score measuring quality of sleep as perceived by parents/guardians (this may include reasons not relating to intervention.
- Health related quality of life – children (CHU9D) [42].
- Health related quality of life – parents/guardians (EQ-5D-5L)[43].
- Pain or discomfort rFLACC [44]– required to identify potential for harm from the novel intervention. CYPCN are complex and have multiple reasons for potential pain, discomfort and agitation and parental/guardian knowledge or best guess is the most accurate option.

- Sleep disordered breathing (SOMNOtouchTM data): we are only measuring this outcome data from participants in the Sussex Community NHS Foundation Trust cohort for this feasibility study.

Based on the performance of the candidate primary and secondary outcome measures, we will determine the final set of primary and secondary outcome measures for a future definitive trial.

Table 1: Primary Feasibility Objectives and Outcomes Measures summary

Feasibility Objectives	Feasibility Outcome Measures	Timepoint of evaluation
1. To evaluate recruitment and retention	a) Recruitment: Proportion of recruited children amongst eligible patients and reasons for not participating. b) Retention: Proportion of participants that completed the feasibility trial out of the number randomised.	Recruitment: At enrolment. Retention: At 6 months.
2. To assess feasibility of recording candidate primary outcome measures	Proportion of data collection (from candidate primary outcomes) complete per participant.	Study Completion: end of data collection at 6 months
3. To estimate the variability of candidate primary outcome measures	Standard deviation of the candidate primary outcomes.	Six months post intervention.
4. To determine design characteristics for a subsequent definitive study	Proposed design, sample size and number of centres for a definitive study.	End of study data analysis and TSC final approval meeting.
5. To assess the acceptability of our novel night-time position for participants, families, health care staff.	a) Acceptability for participants: Estimated Number of nights in position for minimum of 4 hours; b) Acceptability for families and healthcare staff: Survey (acceptability measure)	End of study and survey data analysis.

3. RESEARCH PLAN

3.1. Trial Design

The study design comprises three workstreams (WS):

WS1: Implementing the ADAPT study protocol for 6 months involving 50 CYPCN from different NHS sites. This is a feasibility, interventional, multi-centred randomised controlled trial.
WS2: Process evaluation of the novel night-time intervention examining feasibility of recruitment, delivering intervention, quality of implementation, data collection tools, acceptability to all stakeholders, safety and contextual factors.
WS3: Assessment of feasibility of progression to a definitive trial and determining the design of the subsequent definitive study and health economic analysis.

4. WORKSTREAM 1 – ADAPT STUDY

4.1. Recruitment Strategy

Our primary recruitment strategy is to recruit 50 CYPCN, across multiple NHS sites chosen to provide a range of geographical, socioeconomic and ethnic characteristics. Recruitment will be aided by displaying posters advertising the study to families who attend NHS clinics. As recommended by our PPI group, study information will also be advertised using posters in non-NHS organisations including school newsletters and on school social media. Parents can either contact their site research physiotherapist for more information or email the ADAPT generic email address: sc-tr.adaptclinical@nhs.net. The ADAPT team will forward any direct query involving potential participants to the PI/research physiotherapist at the specific site.

If recruitment to the trial is slower than expected, we will anticipate opening new sites where we have had interest. The joint TSC/DMEC and Clinical Trials Unit working closely with the Chief Investigators will help in regular review of recruitment.

4.2. Trial procedures

This section describes all procedures and evaluations to be done as part of the trial to address the feasibility objectives, in relation to the established study visits. The timing of procedures at each study visit is detailed in the Schedule of Events (Appendix 1).

4.2.1. Participant eligibility criteria

Inclusion criteria:

• Dependent upon others to position/move their bodies (Gross Motor Function Classification System IV/V or equivalent on Functional Mobility Scale (FMS) [34, 35].

• High risk of aspiration linked to swallowing difficulties (Eating and Drinking Classification System IV/V) [36].

• Aged 2-18 years.

• Fed via gastrostomy/jejunostomy.

• At least one lower respiratory tract infection requiring antibiotics in past 12 months.

Exclusion criteria:

• Using a naso-gastric feeding tube.

• Overnight feeding via gastrostomy that cannot be altered to daytime only (jejunostomy feeds can continue overnight with minimal risk of stomach contents entering lungs).

• Transitioning to adult care within the trial period.

• Planned orthopaedic surgery during the trial period.

4.2.2. Identification of participants

All CYPCN in regional caseloads of neurodisability teams will be identified by their direct healthcare team, local PIs and their support teams.

Participant progress through the trial, including numbers/reasons not eligible for trial participation, will be reported according to the CONSORT Statement 2010 extension for pilot and feasibility trials (see Appendix 2)[49].

4.2.3. Invitation and Approach

This will be a two stage approach. Stage 1: if a child meets the following inclusion criteria, none of the exclusion criteria, and eligibility has been confirmed by the treating clinician/site PI, they will be approached:

- Dependent upon others to position/move their bodies (Gross Motor Function Classification System IV/V or equivalent on Functional Mobility Scale (FMS);

- High risk of aspiration linked to swallowing difficulties (Eating and Drinking Classification System IV/V);
- Aged 2-18 years;
- Fed via gastrostomy/jejunostomy.

Stage 2: the parents/guardians will be asked if the child meets the criteria for use of antibiotics in the past 12 months for a 'chest infection'.

If potential participants meet the above criteria they will be offered the opportunity to participate. They may be approached in a variety of ways:

1. Information pack - The parents/guardians of eligible participants will be sent an information pack by the direct healthcare team containing an invitation letter, participant information sheet, consent to contact form and a pre-paid addressed-envelope.
2. Healthcare/clinic team invitation – The direct healthcare team, will speak to parents/guardians of eligible participants in clinic, and will provide an invitation letter, participant information sheet and consent to contact form. Parents/guardians may also be shown a video about the trial on a tablet at their clinic visit. To ensure this is an inclusive trial, we will work with each site to ensure that participation is accessible to the diverse population of CYPCN and their families. The video will have subtitles which can be changed to different languages, as appropriate.

All parents/guardians will be invited to send a consent to contact form back to the direct healthcare team. Once the signed consent to contact form has been received, the site PI or member of the direct healthcare team at each site will contact the parent/guardian to discuss the study with them in more detail, at a suitable time and at a convenient location for them, and answer any of their questions. The discussion could be face-to-face, online via videoconferencing software or over the telephone. Following the discussion, parents/guardians can consent if they wish, for their CYPCN's and their own participation.

4.2.4. Informed Consent

1. **Parent of a child** (aged 2-15)**:** parent/guardian will consent for themselves and for their child.
2. **Young person (16-18) with capacity:** young person consents for themselves, their parent/guardian consent for their own participation. A witness will witness the young adult's verbal consent and sign the consent form.
3. **Young person (16-**18) who lacks mental capacity: we will comply with the provisions of the Mental Capacity Act 2005. A personal consultee will be approached and asked to give their

opinion on the wishes of the young person in relation to the study. If the personal consultee believes the young person would wish to participate, the consultee will sign a consultee declaration form. If no personal consultee can be found, a nominated consultee will be found and the nominated consultee will provide their opinion on the young person's wishes and sign a declaration form. The young person's parent/guardian consents for their own participation.

Consent will be received by a suitably delegated trained member of the local research/direct healthcare team. Consent will be recorded electronically, in an online password protected database (REDCap) designed by the Brighton & Sussex CTU specifically for the study. If the parent/guardian (or young person or consultee) prefers, the consent form (consultee declaration form for consultees) can be completed on paper and uploaded to REDCap. The completed and signed consent form or consultee declaration will also be stored in the CYPCN's medical record and the consent process documented in the medical notes by the local research team. Demographics (age, gender, ethnicity) of all consented participants will be collected for reporting the generalisability of the results. A letter will be sent to the participant's GP to inform them of the CYPCN's participation in the trial.

4.2.4.1. SOMNOtouch consent

Sussex Community NHS Foundation Trust participants will also have the opportunity to be involved in more detailed Somnotouch™ (respiratory polysomnography) monitoring at 0 months (pre-intervention) and at 3 months for both intervention and control participants. See the section about SOMNOtouch (Section 4.3.5.6) for more detail. Participation is voluntary and participants may consent to the study with or without the use of SOMNOtouch.

4.2.5. Participant Withdrawal

The right of a participant to refuse to give consent will be respected, and participants are free to withdraw consent at any time during the study without giving a reason or suffering negative consequences for their future treatment/medical care. If a participant is required to re-consent or new information is required to be provided to a participant, it is the responsibility of the PI to ensure this is done in a timely manner.

We will record whether the child has withdrawn from participating in the whole study, just the intervention, or just the follow-up. The reasons for withdrawal will be recorded if the parent/guardian agree to provide this.

4.2.6. Randomisation

Randomisation occurs after informed consent is documented and before collection of baseline measures. Participants will be randomised 1:1 to either intervention (Breathe-Easy sleep position) or control arm (usual sleep position). Randomisation will be stratified by age (2-10, 11-18) in randomly permuted blocks. Randomisation will be implemented using 'Sealed Envelope' online randomisation software (https://www.sealedenvelope.com/) and will be conducted by the Clinical Trials Unit. A copy of the treatment allocation will be sent by email to the local research team, who will inform the parent/guardian of the CYPCN of the outcome (by telephone call, email or in person) and arrange the first home visit.

4.2.7. Intervention arm

The baseline data will identify any potential issues to implementation of the intervention (if applicable) so they can be addressed, such as adjustments to timings of feeds or medication. Any medical concerns will be discussed with the local respiratory paediatrician, community paediatrician or the CI if no local respiratory colleague is available.

4.2.7.1. Intervention Training

Night-time positioning for postural care is usually managed by the community physiotherapy/occupational therapy teams. Adoption of the novel night-time position changes usual practice in postural care positioning, and the lead research team will work closely with the local research team, CYPCN's physiotherapists and wider healthcare team to support changes in practice, adapting existing night-time positioning and taking other needs into account. Before recruitment starts the local research team and other key Health Care Practitioners (paediatrician, community nurse, other community physiotherapists) will receive an intervention manual and in-person training. This will take place during the site initiation visit or at another mutually agreed time. This training is to support parents/guardians at home with the intervention.

4.2.7.2. Intervention implementation

The PI/site research physiotherapist and local physiotherapist will visit the CYPCN and their parents or carers in their usual home environment to set-up the novel intervention. Oxygen saturations, heart and breathing rates will be taken for each child whilst in their usual sitting and sleep position, followed by the trial bespoke novel position. This is to ensure that the new position does not cause any respiratory adverse effects.

CYPCN will then trial the novel position during the day for a minimum of 1 hour per day for at least a week. This is to determine the safety and comfort of the new position, and effectiveness of upper airway drainage, prior to night-time implementation. Individualised written positioning guidance with photographs and training of parents/guardians or nominated carers, will be provided to support consistent overnight positioning.

When CYPCN, parents/guardians and PI/site research physiotherapist are confident that the position is safe, comfortable and conducive to sleep, it will be trialled for the first part (initial 2-3 hours) of the CYPCN's night-time with the parents/guardians awake and able to monitor them.

As a safety measure, Oxygen saturations and heart rate will be monitored overnight by the parents/guardians for 2 nights pre-implementation of the trial intervention in the intervention group, and 2 full nights at the start of the intervention. Oxygen saturations of 90% or above are acceptable as long as parents/guardians feel this is usual for their child. If Oxygen saturations are predominantly below 90%, then parents/guardians should inform their clinical team. For participants in Sussex only we will also use SOMNOtouch to record these observations. We will record and download the data from SOMNOtouch at baseline and at 3 months.

Equipment and training will be provided to the parents/guardians in liaison with the community nursing team, to drain the participant's stomach contents overnight via their gastrostomy using a drainage bag and to replace fluid loss using oral rehydration salts if required. A video will be provided for training.

The intervention will continue for six months from when it is first introduced at night-time. Weekly telephone contact between the site PI, local research team and parents/guardians will be made for the first month to address any concerns or issues arising, provide further training if required, capture information about adherence to the protocol and any adaptations required. Telephone or email contact will be made every 2-4 weeks thereafter. We anticipate that there will be some nights when the intervention is not used such as when the CYPCN is in a different setting such as respite, holiday or hospital but that the night-time positioning guidance would be followed for 80% of the 6-month trial. We advise that a child should be in the novel intervention position for a minimum of four hours a night. The data collected will help us to estimate what support would be required to successfully implement the intervention. Close contact between local research teams and our expert training team (including an experienced physiotherapist, paediatric respiratory consultant, dysphagia specialist and parent representative) will be maintained throughout.

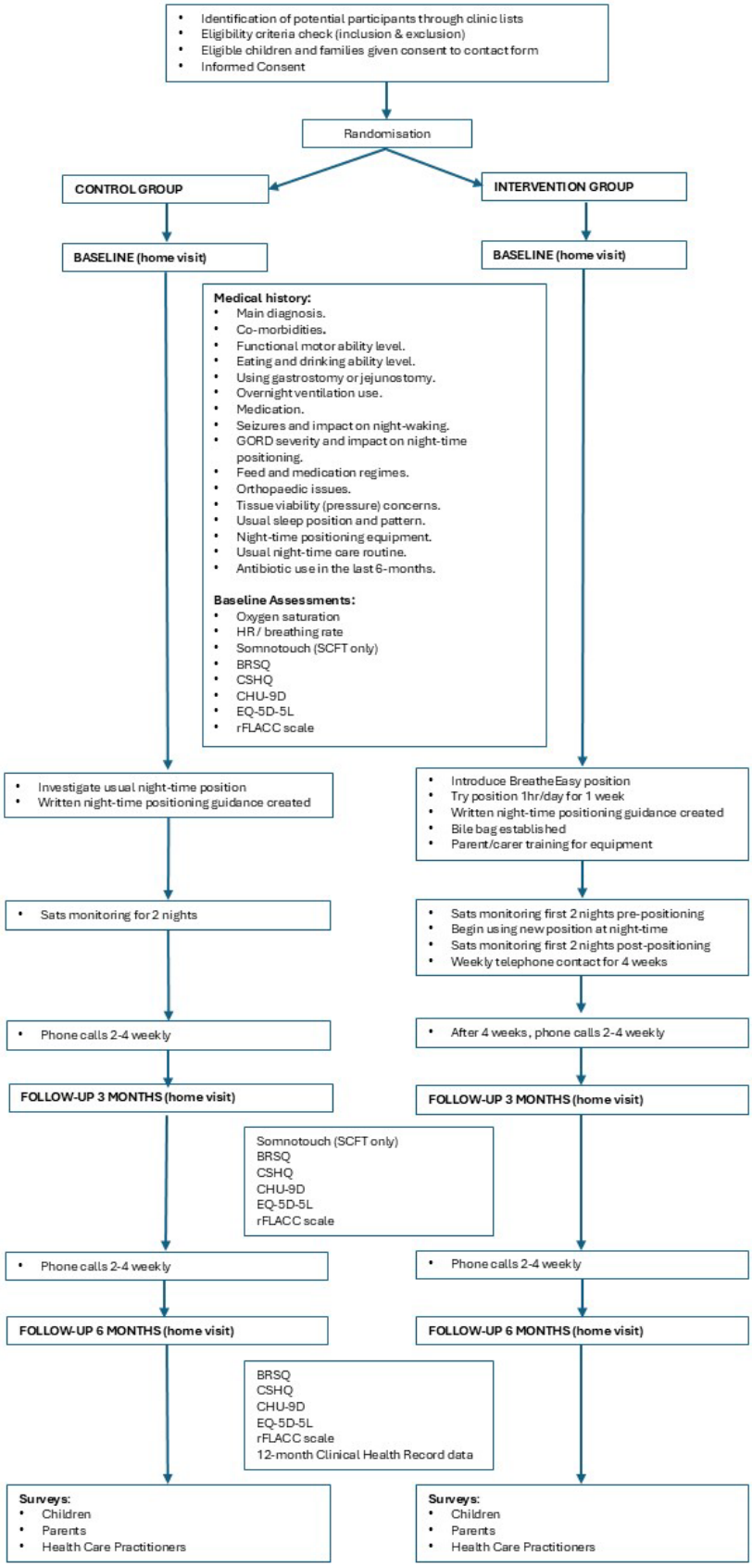
4.2.8. Control Arm

The PI/ site research physiotherapist and local physiotherapist will visit the CYPCN and their parents/guardians in their usual home environment to investigate usual night-time positioning and collect baseline data. Oxygen saturations, heart and breathing rates will be taken for each child whilst in their usual sitting and sleep position. Oxygen saturation and heart rate will be recorded by parents/guardians for 2 consecutive nights at this stage as for the intervention group. Oxygen saturations of 90% or above are acceptable as long as parents/guardians feel this is usual for their child. If Oxygen saturations are predominantly below 90%, then parents/guardians should inform their clinical team. No changes will be made to overnight positioning for CYPCN in this group for the duration of the study. The CYPCN will continue to receive their usual care with regards to night-time positioning for 6 months from date of baseline measures. To check for contamination, we will check individualised written positioning guidance to ensure that usual care is followed. Every 2-4 weeks participants in the control group will be contacted by telephone to address any concerns.

4.2.9. Both groups

Health records for all participants will be accessed through community, hospital and GP records to collect data on respiratory health: number of chest infections, time to first chest infection, chest X-rays, use of antibiotics and hospital visits for chest infections and any other hospital stay and the reason noted. Data will be collected by local research teams retrospectively at participant's entry to trial and then at trial intervention completion at 6 months, with support from the site level research delivery team. How easily this data is gathered will help design the definitive RCT.

4.3. Study Flow Diagram



4.4. Data management

4.4.1. Data collection tools and source document identification

To ensure accurate data collection, a study pack will be created which will function as the clinical case notes for all children entering the study. The pack will include Case Report Forms (CRFs) to facilitate clear recording of outcome data collected via telephone calls/emails or in person from parents/guardians. The local research team will then have the option to complete the CRFs on paper if they do not have immediate access to the online CRF, or directly into REDCap itself.

REDCap training will be provided to guide the local research team and ensure data consistency at each recruiting centre.

Once the data have been cleaned and the database locked, the data will be transferred securely to the trial statistician for analysis.

4.4.2. Data handling and record keeping

The CRF data will be kept and handled according to the BSCTU SOPs and data management plan. All individualised written positioning guidance including photographs will be stored at each site. When using the Talking Mats with the children, digital photographs will be taken of their Talking Mats responses. This descriptive data will be securely stored in the digital records for each case at each site.

4.4.3. Access to Data

Direct access will be granted to authorised representatives from the Sponsor, host institution, regulatory authorities and the CTU, to permit trial-related monitoring, audits and inspections, in line with participant consent. Anyone in the local research team who will be entering data will have study specific REDCap training and be provided with a user guide. Parents/guardians and participants who complete the questionnaire data directly onto REDCap will do so in a survey format, where the training and user guide will not be required.

4.4.4. Baseline Data collection

The following baseline data will be collected during the first home visit for all participants. Some of this data will be used to enable effective set up of the intervention whilst some will be entered into REDCap:

- CYPCN's medical history – Such as main diagnosis and co-morbidities.
- Functional motor ability level.
- Eating and drinking ability level.

- Whether using gastrostomy or jejunostomy.
- Overnight ventilation use.
- Medication including prophylactic antibiotics, reflux and saliva medication.
- Details about seizures and impact on night-waking/implementation of intervention.
- GORD severity and impact on night-time positioning.
- Feed and medication regimes prior to sleep and overnight.
- Orthopaedic issues such as scoliosis or hip dysplasia, pain
- Tissue viability (pressure) concerns.
- Usual sleep position and pattern
- Use of night-time positioning equipment
- Usual night-time care routine (if applicable).

Recruitment, randomisation and retention of all participants will be assessed from data recorded at each site on number of:

- eligible CYPCN on caseload.
- families declining participation at different stages:
    - before randomisation,
    - after randomisation,
    - in initial phase to prior to the novel intervention.
- families dropping out in the intervention group, including the timepoint and reason for dropout.
- families dropping out in the control group, including the timepoint and reason for dropout.

In consultation with our PPI groups, outcome measures will be recorded by the local research team at baseline, 3 and 6 months, in questionnaire format.

4.4.5. Questionnaires

4.4.5.1. Bespoke Respiratory Symptoms Questionnaire (BRSQ)

Parents/guardians describe their CYPCN's respiratory symptoms, frequency of symptoms and impact on CYPCN and family over previous month using five-point Likert scales [37,38]. The BRSQ questionnaire must be completed by the caregiver (see Appendix 3).

4.4.5.2. Child's Sleep Habits Questionnaire (CSHQ)

Parents/guardians describe frequency of their CYPCN's behaviours associated with common paediatric sleep difficulties over previous 3 months using three-point Likert scales [39-41]. (See Appendix 4).

4.4.5.3. Child Health Utility Instrument (CHU9D)

CHU9D is a paediatric generic preference-based measure of health-related quality of life suitable for children aged 7-17 years. It is used for the calculation of Quality Adjusted Life Years (QALYs) in economic evaluations. It consists of a short questionnaire and a set of preference weights using general population values. The questionnaire has 9 items covering emotions, sleep, schoolwork, daily routine and activities each with 5 response levels. It is for self-completion or proxy report [42]. Each item taps into a different domain (worry, sadness, pain, tiredness, annoyance, school, sleep, daily routine and activities) with a five-level response category. We have received agreement from the licensee to use this measure by proxy with parents/guardians of CYPCN in younger age groups. (See Appendix 5).

4.4.5.4. EuroQol EQ-5D-5L

Parents/guardians will be asked to complete the EQ-5D-5L instrument. This is a generic health related quality of life instrument comprising a five-dimension descriptive system (mobility, self-care, usual activities, pain/discomfort and anxiety/depression) scored using 1 of 5 levels (no problems, slight problems, moderate problems, severe problems, extreme problems). The EQ instrument also includes a vertical visual analogue scale for parents'/guardians' own rating of their health, ranging from 0 to 100 with endpoints labelled "the worst health you can imagine" and the best health you can imagine" [43]. (See Appendix 6).

4.4.5.5. Pain or discomfort score

We will use the rFLACC scale or Face, Legs, Activity, Cry, Consolability scale FLACC Scoring [44] at Baseline, 3 and 6 months for all CYPCN in the trial. It is an observational behavioural pain scale for children who are unable to self-report their level of pain due to developmental disabilities. For each of the five categories, Face, Legs, Activity, Cry and Consolability, a score of 0, 1 or 2 is given. This results in a total score of 0–10 with 0 representing no pain. (See Appendix 7).

4.4.5.6. SOMNOtouch™

We will investigate the feasibility of collecting objective respiratory polysomnography data using SOMNOtouch™ monitors at night-time. SOMNOtouch™ monitors are well validated for cardio-respiratory sleep studies and widely used to diagnose sleep-disordered breathing including use at

home [45]. We will use SOMNOtouch™ to record respiratory polysomnography data at home only with participants recruited at the Sussex site where the equipment is available as part of usual clinical practice, both in the intervention and control arms.

We will record data from 2 overnight recordings: one at baseline (pre-intervention in the intervention group) and one 3 months after the start of the trial. Data recorded will include pulse oximetry, chest and abdominal Respitrace bands, ECG and if tolerated nasal cannulae. SOMNOtouch™ data will be downloaded and analysed (by AK and PS) to provide detailed information about sleep-disordered breathing and sleep quality.

We will triangulate these objective measures with other study measures. We will use a pre-recorded video for parents/guardians on use of the SOMNOtouch™ as recommended by our PPI partners and written instructions.

4.4.6. Clinical data

The outcome measures listed below will be recorded retrospectively from community, GP and hospital records. At trial entry, the data will be accessed and recorded in REDCap. At exit of intervention period, retrospective data for 6 months over trial period will also be collected. (total of 12 months data)

- Details of all treatment courses of antibiotics for chest infections and other causes or prophylactic antibiotics e.g. Azithromycin 3 days a week.
- Number of hospital admissions and days in hospital for chest infection and total days of hospitalisation, including non-chest related days.

4.4.7. Demographic, socioeconomic and diagnostic information:

We will use parental/consultee highest level of educational attainment to determine socio-economic status. This will be collected via REDCap following randomisation.

5. WORKSTREAM 2 - PROCESS EVALUATION

The process evaluation assesses how implementation of the novel trial intervention is achieved, the influence of contextual factors including randomisation and trial design, and the quality of the implementation and acceptability of the trial [47].

The following participants will be included:

- Parents/guardians of CYPCN participating in the study (n=50)

- CYPCN aged between 5-18yrs able to understand questions and communicate at least yes/no answers. (n=10)
- HCPs involved in implementing feasibility RCT intervention and control arms n=50.

A survey (developed from thematic analysis of interviews in the Breathe-Easy proof-of-concept study) will be sent to all parents/guardians and HCPs on trial completion to obtain their views and experiences of taking part in the RCT. We will encourage parents / guardians to complete the surveys within 7 days, but have a maximum window of 4 weeks.

Survey questions will explore:

- Views on randomisation.
- Prospective acceptability (how participants felt about the trial prior to participating).
- Experiences of taking part (being in the control arm, ease of implementing intervention, perceptions of intervention effectiveness, burdens, barriers, satisfaction, ethicality).
- Influence of contextual factors including NHS settings.
- Difference made to CYPCN and parents/guardians.

Children and young people who participate in the ADAPT trial will be offered the opportunity to participate in a face-to-face interview with someone who knows them well, other than their parent/guardian. It is likely that all children and young people taking part in the trial will be unable to use speech to communicate; they will rely on alternative and augmentative forms of communication that may be challenging for unfamiliar others to understand. Children and young people who are able to indicate personal preferences such as “like”, “dislike” and “don’t know” will be supported to answer a series of questions using a Talking Mats framework [48]. The research speech and language therapist will liaise with the local research team to identify which children and young people are able to participate and who would be best to support the child to communicate their preferences. They will provide training to the person who is going to support the child to communicate their preferences; they will also join the person online when they deliver the Talking Mats session to CYP, if required. The research speech and language therapist will interview the Sussex participants. Each site will be sent a set of pictures / symbols and a mat which can be used with different children and young people. Children will be asked to arrange each picture / symbol under one of three headings for two distinct questions. The headings for the mats will be: “Like” “not sure” and “don’t like”. The two questions are:
I want to talk about different things that happen to you every day. What do you think about: 1. Waking up, 2. Getting dressed etc.

I want to talk about sleeping at night-time. What do you think about: 1. Going to bed, 2. Lying on your back, 3. Lying on your side etc.

See ADAPT Talking Mats Framework and questions document for the complete list of symbols and an illustration of a completed talking mat in Appendix 8. A photographic record will be made of children and young people's responses; these digital photographs will be sent directly to the central study team.

Families who started the study, but discontinued participation will be given the opportunity to provide feedback via an online questionnaire [47].

During the 6 month trial, the local research team will complete structured case-report forms (CRFs), directly entered into REDCap, to support process evaluation, including:

- Agreed adaptations to protocol and deviations.
- Appraisal of contextual factors.
- Appraisal of fidelity of intervention and influencing factors including number of hours/days the novel intervention is in use.
- Protocol issues and challenges arising.

For the Sussex participants who have used SOMNOtouch™ , a survey will be sent to the parents/guardians to provide feedback about the usability and comfort of the equipment.

6. Statistics and data analysis

6.1. Sample size justification

We will aim to consent 50 participants which is sufficient to estimate a proportion of around 70% with a <13% margin of error at the 95% confidence level (relevant to the follow up/data completeness outcome, see WS3). This number is consistent with recommendations for the size of feasibility studies (between 50[32] and 70[33]) to provide precise estimates of the variability of the candidate primary outcome measures and feasibility outcomes.

6.2. Statistical Analysis Plan

Participant flow through the study will be reported in a CONSORT flow chart adapted for pilot and feasibility studies [49]. Available cases will be analysed, following intention to treat principles. Demographics, feasibility outcomes and candidate definitive trial primary/secondary outcomes will be summarised descriptively overall and by arm using statistics appropriate to their distributions (count/percentage for categorical variables, mean/SD for normally distributed continuous variables,

and median/IQR for skewed continuous variables) at baseline and at each time point (where applicable). Differences between intervention arms for candidate outcomes will be reported with bootstrapped 95% confidence intervals, with estimated effect sizes. No statistical testing will be performed. Analyses will be performed in Stata version 18.0 or later. The full Statistical Analysis Plan will be reviewed and signed off by the CI and study statistician prior to any analyses taking place.

7. Qualitative data analysis

Quantitative survey data will be analysed using descriptive statistics. Thematic analysis will be conducted with the qualitative data from free text responses from the surveys and qualitative case record data. These data will be analysed through coding, combining into themes and reviewed in line with the study objectives [50]. We will identify, analyse and report patterns to evaluate the process of conducting all aspects of the trial including recruitment and retention, implementation of the intervention and outcome measures [50,51].

8. Health Economic analysis

A health economic evaluation within this feasibility trial will facilitate the planning of the definitive RCT. It will take the perspective of the health and social care system.

The resources involved in delivering the intervention to individual families will be obtained from the case records kept by therapists, including equipment costs and the time they spend supporting families. On completion of the study, data will be gathered from hospital, community and GP records on all contacts, use of antibiotics, tests (e.g. X-rays) and hospitalisations for respiratory-related problems for each child participant starting at 6 months prior to participant entry to trial. Intervention and service utilisation will be costed using national tariffs, inclusive of oncosts and overheads [52].

Health related quality of life of children, the primary outcome for the economic evaluation, will be recorded at each assessment point using CHU-9D. The health-related quality of life of parents/guardians will similarly be recorded using EQ-5D-5L. Value sets will be applied separately for children and parents/guardians-carers for the estimation of Quality Adjusted Life Years [53]. The sensitivity of CHU-9D will be explored by observing associations with other outcome measures to indicate the suitability of CHU-9D in this population.

Data completeness will be explored, and variables will be examined using descriptive statistics. Differences in service use between groups will be investigated for indications of potential savings that might offset the intervention costs. Group mean costs and QALYs of children will be estimated and compared in a preliminary cost effectiveness analysis. The full range of outcomes (children and parents/guardians) will be investigated in a cost consequences framework.

9. WORKSTREAM 3 - PROGRESSION CRITERIA AND DEFINITIVE RCT DESIGN

Findings from WS1 and WS2 will be reviewed by the central study team and PPI group.

The pre-specified progression criteria for proceeding to a definitive trial are:

Table 2: Progression criteria for definitive trial

Feasibility Measure	**Red**	**Amber**	**Green**
Recruitment	<0.3 per month per site.	0.3 to 1 per month per site.	>1 patient per month per site.
3-month and 6-month follow up completion	<50% in either arm.	50% to 70% in either arm.	>70% in both arms.
Candidate primary outcomes	No candidate primary outcomes suitable for full trial.	One or more candidate primary outcome partially suitable for full trial.	One or more candidate primary outcomes fully suitable for full trial.
Intervention use of > 4 hrs per night at 6 months	<50% in intervention arm.	50% - 70% in intervention arm.	>70% in intervention arm.
Acceptability (Process Survey)	<50% participants agree intervention is acceptable.	50% to 70% participants agree intervention is acceptable.	>70% participants agree intervention is acceptable.

If all green conditions are met, the definitive trial will be considered feasible. If any amber conditions are met, the definitive trial will be considered feasible with reasonable design adjustments to address these issues. The joint TSC/DMEC will approve these adjustments. If any red conditions are met, the definitive trial will be considered not feasible under the current design. The central study team will explore options for substantially redesigning the study, potentially with further feasibility work. The joint TSC/DMEC will make any final decisions with regards to progression to the definitive RCT.

Candidate primary outcomes will be considered suitable for the definitive trial if they are completed by >70% of followed up participants, do not show any obvious ceiling/floor effects and are likely to

be sensitive enough to show meaningful differences between arms in the definitive trial. 95% confidence intervals will be calculated for each candidate primary outcome, but we have not based the relevant feasibility criterion on coverage of these intervals suggesting an effect of the intervention, as we do not yet know if they are suitable outcomes, and the study is not powered to detect differences between arms.

If the progression criteria are met, primary and secondary outcomes for definitive trial will be selected and a sample size calculation will be performed. Scaled process survey data and free text responses will provide important contextual information about implementing the trial intervention in different sites, including refinements to recruitment or intervention strategies required for definitive trial. If identified problems cannot be managed, we will not proceed to a definitive RCT.

10. End of study

The end of the trial will be the last data capture of the last participant, following data collection and follow-up monitoring.

11. Dissemination, outputs and anticipated impact

Dissemination

Our multifaceted dissemination strategy aims to reach families of CYPCN, healthcare professionals, healthcare professional organisations, NHS commissioners and managers, researchers, and the public. We will interpret the findings and report implications for progression to a definitive RCT of ADAPT including any necessary amendments to intervention delivery and/or trial design. A complete and transparent report of the study will be produced with reference to recommendations of the CONSORT 2010 statement: extension to randomised pilot and feasibility trials. [49] We will submit the paper for publication in a high-impact, peer-reviewed, open access, academic journal targeting those with a special interest in neurodisability, chest health or child community healthcare. A plain language summary of the findings will be co-produced with members of our PPI group. The plain language summary will be sent to study participants and the NHS teams involved in the study.

Outputs

If we demonstrate that it is feasible to deliver the novel position intervention and evaluate the intervention in a trial, then we will apply to the NIHR Health Technology Assessment programme to conduct a definitive RCT of intervention effectiveness and cost-effectiveness.

Impact

If the pre-specified conditions are met within this feasibility study, we will proceed to a definitive trial with NIHR HTA or RfPB tier 1 funding. If the intervention is shown to be effective and cost-effective in the definitive trial, then our ultimate aim is for the Breathe-Easy position to be implemented as part of night-time postural management and respiratory care in the community. We anticipate that this trajectory to patient and public benefit would take approximately five to seven years.

12. Project Timetable

This study will be completed in 24 months, with additional ongoing dissemination and planning of a subsequent definitive trial after the end date.

The key milestones are indicated in the attached Gantt chart (Appendix 9). These are:

- Apply for ethical and HRA approvals by month 3.
- Site initiation visits and training of staff, planning and preparation for recruitment by month 6.
- Recruitment of participants and randomisation by month 15.
- Intervention completed and data collected by month 21.
- Analysis of qualitative and quantitative data and disseminations of findings via a full report, plain language summary, results paper for an open access journal, and conference presentations by month 24.
- Development of the protocol for the substantive trial if criteria for continuation at met by month 24 and after study completion.

13. Ethics / Regulatory Approvals / Considerations

We will ensure that all members of the Central Study Team and Local Research Teams complete NIHR Good Clinical Practice training.

Health Research Authority (HRA) approval will be in place before the start of any trial procedures. HRA approval incorporates the NHS Research Ethics Committee favourable opinion. Any substantial

amendments required throughout the duration of the study will follow the process of regulatory approval as laid down in the HRA guidelines at the time. Study sponsorship is undertaken by Sussex Community NHS Foundation Trust. All correspondence with the REC and Sponsor will be retained in the Trial Master File and investigator site files.

We will provide clear written participant information sheets during the recruitment and consent processes. Discussions about the process of randomisation in this clinical trial will take place at the time of recruitment. CYPCN and their families will be informed whether they have been randomised to the novel intervention or usual care, as it will not be possible to blind the participants or the local research team to the randomised allocation due to the nature of the intervention. Some parents may become distressed about previously unknown risks to CYPCN's respiratory health linked to their swallowing difficulties and night-time positioning. Health Care Practitioners at site level will be given the information they need to provide necessary support to CYPCN and their families. If any problems arise, the local research team will inform the core Trial Management Team who will follow up to establish whether CYPCN's participation in the study needs to stop. Families and HCPs will be signposted to the relevant services.

Contamination:

We recognise that some HCPs or families might learn about the novel intervention and want to adopt it themselves, leading to contamination of usual care conditions. Training at each site will emphasise the need for CYPCN to stay in the designated intervention arm or in the control arm for the full 6-month trial. We will check individualised written positioning guidance to ensure that the agreed night-time position has been followed. This feasibility study will consider whether the proposed individual randomisation is a suitable design. If significant contamination is found at site level, from site PI and/or site research physiotherapist feedback, cluster randomisation will be considered in a future trial design.

14. SAFETY REPORTING

The International Conference for Harmonisation/Good Clinical Practice (ICH/GCP) requires Investigators and Sponsors to follow specific procedures when notifying and reporting adverse events/reactions in research studies.

We have a risk and safeguarding protocol if concerns arise. Any adverse events will be reported to the CTU, Sponsor, Central Study Team and the joint TSC/DMEC Committee.

Pain or discomfort score:

As we are mindful of musculoskeletal concerns for these participants, we will use the rFLACC (Face, Legs, Activity, Cry, Consolability [44]) at Baseline, 3 and 6 months for all CYPCN in the trial. It is a measurement to assess pain for children between the ages of 2 months and 7 years or individuals that are unable to communicate their pain. The scale ranges from 0 to 10 with 0 representing no pain and 10 representing the most pain.

The joint TSC/DMEC will review the safety indications of any increase in rFLACC scale and possibly pause the intervention for a period of time to reevaluate if considered necessary.

14.1. Definitions

Term	Definition
Adverse Event (AE)	Any untoward medical occurrence in a participant that has received a study intervention, including occurrences which are not necessarily caused by or related to that intervention.
Related Event (RE)	An untoward and unintended response in a participant to an intervention which is related to the administration of that intervention. A causal relationship between the trial intervention and an AE is at least a reasonable possibility, i.e. the relationship cannot be ruled out. All cases judged by either the reporting medically qualified professional or the Sponsor as having a reasonable suspected causal relationship to the intervention qualify as adverse reactions.

	We anticipate there will be many adverse events due to the participant cohort. For this study, we will be reporting REs requiring medical intervention only.
Serious Adverse Event (SAE)	A serious adverse event is any untoward medical occurrence that: • results in death • is life-threatening • requires inpatient hospitalisation or prolongation of existing hospitalisation • results in new persistent or significant disability/incapacity • consists of a new anomaly or defect Other 'important medical events' may also be considered serious if they jeopardise the participant or require an intervention to prevent one of the above consequences. NOTE: The term "life-threatening" in the definition of "serious" refers to an event in which the participant was at risk of death at the time of the event; it does not refer to an event which hypothetically might have caused death if it were more severe.
Serious Related Event (SRE)	An adverse event that is both serious and, in the opinion of the reporting Investigator, believed with reasonable probability to be due to one of the trial interventions, based on the information provided.
Suspected Unexpected Serious Related Event (SUSRE)	Any **related** and **unexpected** (i.e. not listed in the protocol as expected) event that meets the definition of 'serious' that can be attributed to the trial intervention/procedure.

14.2. Reporting of Serious Adverse Events

All AEs and SAEs will be reported using the study-specific safety reporting form (held digitally on REDCap) within 24 hours of being made aware. Any SAEs will automatically notify the CTU team, or they can be contacted separately at bsctusafety@bsms.ac.uk.

1. A SAE form on REDCap must be completed by the local principal Investigator, with the causality and expectedness of the event clearly documented, within one working day of the investigator's knowledge of the event. In the absence of the responsible investigator the form should be completed by a member of the local research team (as named on the delegation of responsibilities log). The local PI should subsequently check the SAE form, make changes as appropriate, sign and then submit to BSCTU as soon as possible. The initial report can be followed with more detail as appropriate.

2. The SAE will then have the causality and expectedness assessed by the study's Chief Investigator acting as Clinical Reviewer.

3. Follow-up: Participants must be followed-up until clinical recovery is complete, or until the event has stabilised. Follow-up should continue after completion of protocol treatment if necessary. The patient must be identified by trial number and date of birth only. The patient's name should not be used on any correspondence.

4. The sponsor will notify the research ethics committee of SUSREs as per the conditions of the favourable opinion.

14.3. Assessment of Expectedness and Causality

The assessment of **expectedness** will be made by the site PI (or a delegated qualified medical professional). **Expected** adverse reactions are listed (but not limited to) below:

- Disturbance to sleep
- Local skin irritation on face
- Weight loss due to under nutrition
- other as deemed by investigator

The PI (or delegated qualified medical professional) will also assess the **causality** in relation to the trial intervention, using the definitions below:

- Unrelated: Relationship to the device and/or procedure can be excluded
- Possibly: The relationship with the use of the device and/or procedure is weak but cannot be ruled out completely. Alternative causes are also possible.
- Probably: The relationship with the use of the device and/or procedure seems relevant and/or the event cannot be reasonably explained by another cause.

- Definitely: The event is associated with the device and/or procedure beyond reasonable doubt.

14.4. Recording and reporting of SUSREs

For this study, only reports of Serious Adverse Events (SAEs) that are:

- **related** to the study (i.e. they resulted from administration of any of the research procedures) and

- **unexpected** (i.e. not listed in the protocol as an expected occurrence)

(SUSREs) should be submitted to the REC using the HRA Non-CTIMP safety report form.

14.5. Responsibilities of Safety Reporting

The principal investigator at each site will be responsible for reporting any SAE/ SRE to the CTU and CIs.

The trial manager at the CTU will be responsible for ensuring any SAE/SRE reports are complete and accurate and will follow up with the local research teams to ensure this. The trial manager will maintain and update all SAE/ SRE records required for reporting to Sponsor and REC.

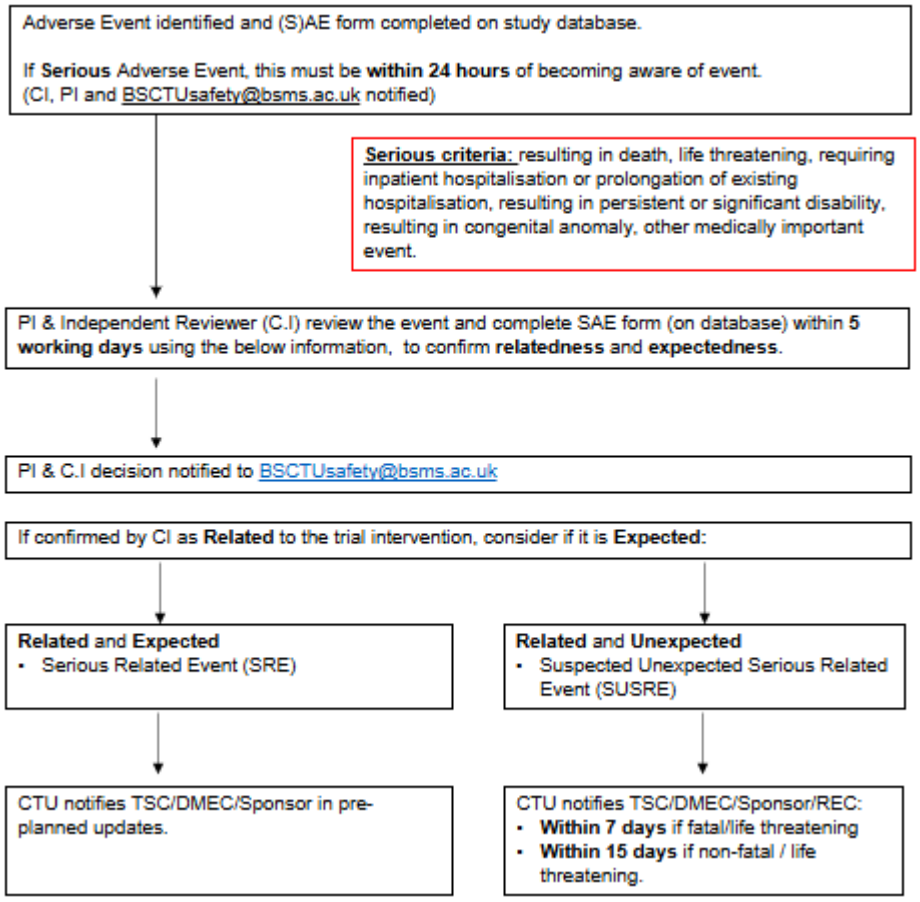


Figure 1: Safety Reporting Flow Diagram

15. Archiving

Once the end of trial report has been submitted, archiving will be authorised by the Sponsor.

All essential documents will be archived for a minimum of 5 years after completion of trial, after which destruction of essential documents will be authorised by the Sponsor.

16. Protocol compliance

All protocol non-compliances (departures from the approved protocol) will be recorded and reported to the trial manager, Chief Investigator and Sponsor, and subsequently to the joint TSC/DMEC.

Deviations from the protocol which are found to frequently recur are not acceptable, will require immediate action and could potentially be classified as a serious breach.

17. Notification of Serious Breaches to GCP and/or the protocol

A “serious breach” is a breach which is likely to effect to a significant degree –

(a) the safety or physical or mental integrity of the participants of the trial; or

(b) the scientific value of the trial

The sponsor will be notified immediately of any case where the above definition applies during the trial conduct phase.

18. Data protection and patient confidentiality

All investigators and trial site staff must comply with the requirements of the General Data Protection Regulation 2018 with regards to the collection, storage, processing and disclosure of personal information and will uphold the Act’s core principles. Details of this can be found here: https://www.hra.nhs.uk/about-us/news-updates/gdpr-guidance-researchers/

19. Financial and other competing interests for the Chief Investigator, PIs at each site and committee members for the overall trial management

No known financial or other competing interests to declare.

20. Indemnity

In the case of any harm to participants arising from the management or design of the research, the NHS indemnity scheme will apply.

21. Amendments

The CI takes responsibility for designing and ensuring correct implementation of any study amendments. Amendments which arise during the course of the study will be reviewed by the Sponsor prior to submission to the relevant authority, following HRA guidelines. The TM at the CTU will organise documentation, submission, and dissemination of any amendments to the local research teams.

22. Access to the final trial dataset

The final full dataset will be available to the CI, the local research teams, the study statistician and members of the joint TSC/DMEC. Aspects of the dataset will be available to the other members of the central study team.

23. Trial Monitoring, Audit and Inspection

The Brighton and Sussex Clinical Trials Unit (UKCRC Number 66) will take on the role of overseeing the management of the study. A trial manager (TM) will work closely with the local research teams to ensure that timelines are met, monitored, and recruitment is tracked. They will also undertake quality assurance monitoring procedures to ensure the study is being conducted in accordance with the protocol. The TM will support the setup of the sites, ensuring all documentation and processes are in line with research governance and HRA processes.

Monthly trial management group (TMG) meetings with the CI, TM, Data Manager (DM), statistician and local research teams will oversee the study progress. Recruitment and data updates will be discussed to highlight any issues and to ensure they can be resolved in a timely manner.

A joint trial steering committee (TSC) and Data Monitoring and Ethics Committee (DMEC) will consist of the TMG and the other co-applicants, together with a minimum of 3 independent members (a lay member - parent of a child with neurodisability, a paediatrician and therapists with relevant expertise, and a statistician). The joint TSC/DMEC will meet every 6 months, will receive reports from the TMG and will oversee the progress of the study. With its independent membership it will also review the data and safety issues.

24. References

1. Horridge K. Each and every need: a review of the quality of care provided to patients aged 0-25 years old with chronic neurodisability, using the cerebral palsies as examples of chronic neurodisabling conditions. London: Healthcare Quality Improvement Partnership; 2018.

2. Surman G, Hemming K, Platt MJ, Parkes J, Green A, Hutton J, et al. Children with cerebral palsy: severity and trends over time. Paediatr Perinat Epidemiol. 2009;23(6):513-21.

3. Blackmore AM, Bear N, Blair E, Langdon K, Moshovis L, Steer K, et al. Predicting respiratory hospital admissions in young people with cerebral palsy. Archives of disease in childhood. 2018;103:1119-24.

4. Carter B, Bennett CV, Jones H, Bethel J, Perra O, Wang T, et al. Healthcare use by children and young adults with cerebral palsy. Dev Med Child Neurol. 2021;63(1):75-80.

5. Meehan E, Reid SM, Williams K, Freed GL, Sewell JR, Vidmar S, et al. Hospital admissions in children with cerebral palsy: a data linkage study. Dev Med Child Neurol. 2017;59(5):512-9.

6. Chiang KL, Huang CY, Fan HC, Kuo FC. Prolonged length of stay for acute hospital admissions as the increasing of age: A nationwide population study for Taiwan's patients with cerebral palsy. Pediatr Neonatol. 2019;60(1):74-82.

7. Young NL, McCormick AM, Gilbert T, Ayling-Campos A, Burke T, Fehlings D, et al. Reasons for hospital admissions among youth and young adults with cerebral palsy. Arch Phys Med Rehabil. 2011;92(1):46-50.

8. Blackmore AM, Bear N, Blair E, Gibson N, Jalla C, Langdon K, et al. Prevalence of symptoms associated with respiratory illness in children and young people with cerebral palsy. Dev Med Child Neurol. 2016;58(7):780-1.

9. Seddon PC, Khan Y. Respiratory problems in children with neurological impairment. Arch Dis Child. 2003;88(1):75-8.

10. Westbom L, Bergstrand L, Wagner P, Nordmark E. Survival at 19 years of age in a total population of children and young people with cerebral palsy. Dev Med Child Neurol. 2011;53(9):808-14.

11. Primhak RA, Hicks B, Shaw NJ, Donaldson GC, Balfour-Lynn IM. Use of home oxygen for children in England and Wales. Arch Dis Child. 2011;96(4):389-92.

12. Berry JG, Poduri A, Bonkowsky JL, Zhou J, Graham DA, Welch C, et al. Trends in resource utilization by children with neurological impairment in the United States inpatient health care system: a repeat cross-sectional study. PLoS Med. 2012;9(1):e1001158.

13. Bear NL, Wilson A, Blackmore AM, Geelhoed E, Simpson S, Langdon K. The cost of respiratory hospitalizations in children with cerebral palsy. Dev Med Child Neurol. 2024;66(3):344-52.

14. Strydom A. Learning from Lives and Deaths - people with a learning disability and autistic people (LeDeR). In: Sheehan R, White A, editors. London, England 2021.

15. Glover and Ayub. How people with learning disabilities die. Improving Health and Lives Learning Disability Observatory; 2010.
16. National Collaborating Centre for Women's and Children's Health (2012). National Institute for Health and Clinical Excellence: Guidance. Spasticity in Children and Young People with Non-Progressive Brain Disorders: Management of Spasticity and Co-Existing Motor Disorders and Their Early Musculoskeletal Complications. London: RCOG Press.
17. Humphreys G, King T, Jex J, Rogers M, Blake S, Thompson-Coon J, et al. Sleep positioning systems for children and adults with a neurodisability: A systematic review. British Journal of Occupational Therapy. 2019;82(1):5-14.
18. Farley R, Clark J, Davidson C, Evans G, et al. What is the evidence for the effectiveness of postural management? British Journal of Therapy and Rehabilitation. 2003;10:449-55.
19. Gericke T. Postural management for children with cerebral palsy: consensus statement. Developmental Medicine & Child Neurology. 2006;48(4):244.
20. Osborne LJ, Gowran RJ, Casey J. Evidence for 24-hour posture management: A scoping review. British Journal of Occupational Therapy. 2023;86(3):176-87.
21. Hill S, Goldsmith J. Biomechanics and prevention of body shape distortion. Tizard Learning Disability Review. 2010;15(2):15-32.
22. Polak F, Clift M. The use of night time postural management equipment: a survey of UK paediatric chartered physiotherapists. Challenges for Assistive Technology. 2007;20:458-64.
23. Blake SF, Logan S, Humphreys G, Matthews J, Rogers M, Thompson-Coon J, et al. Sleep positioning systems for children with cerebral palsy. Cochrane Database Syst Rev. 2015;2015(11):Cd009257.
24. Littleton SR, Heriza CB, Mullens PA, Moerchen VA, Bjornson K. Effects of positioning on respiratory measures in individuals with cerebral palsy and severe scoliosis. Pediatr Phys Ther. 2011;23(2):159-69.
25. Gough M. Continuous postural management and the prevention of deformity in children with cerebral palsy: an appraisal. Developmental Medicine & Child Neurology. 2009;51(2):105-10.
26. Gibson N, Blackmore AM, Chang AB, Cooper MS, Jaffe A, Kong WR, et al. Prevention and management of respiratory disease in young people with cerebral palsy: consensus statement. Dev Med Child Neurol. 2021;63(2):172-82.
27. Dawson NC, Padoa KA, Bucks RS, Allen P, Evans H, Mccaughey E, et al. Ventilatory function in children with severe motor disorders using night-time postural equipment. Developmental Medicine & Child Neurology. 2013;55(8):751-7.

28. Legg J, Allen JL, Andrew M, Annesley C, Chatwin M, Crawford H, et al. BTS Clinical Statement on the prevention and management of community-acquired pneumonia in people with learning disability. Thorax. 2023;78(Suppl 1):s22-s52.
29. Simpson AJ, Allen JL, Chatwin M, Crawford H, Elverson J, Ewan V, et al. BTS clinical statement on aspiration pneumonia. Thorax. 2023;78(Suppl 1):s3-s21.
30. Hoffmann TC, Glasziou PP, Boutron I, Milne R, Perera R, Moher D, et al. Better reporting of interventions: template for intervention description and replication (TIDieR) checklist and guide. BMJ. 2014;348(mar07 3):g1687-g.
31. Perry H, Kapur A, Dewan T. Reducing repeat admissions for aspiration pneumonia in children with severe neurological dysfunction through a positioning technique – a case series report. Pediatric Intensive and Critical Care Congress: https://www.researchgate.net/figure/Upper-Airway-Drainage-positioning-instructions_fig1_339004420; 2016.
32. Sim J, Lewis M. The size of a pilot study for a clinical trial should be calculated in relation to considerations of precision and efficiency. J Clin Epidemiol. 2012;65(3):301-8.
33. Teare MD, Dimairo M, Shephard N, Hayman A, Whitehead A, Walters SJ. Sample size requirements to estimate key design parameters from external pilot randomised controlled trials: a simulation study. Trials. 2014;15:264.
34. Palisano R, Rosenbaum P, Walter S, Russell D, Wood E, Galuppi B. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Developmental Medicine & Child Neurology. 1997;39(4):214-23.
35. Graham HK, Harvey A, Rodda J, Nattrass GR, Pirpiris M. The Functional Mobility Scale (FMS). J Pediatr Orthop. 2004;24(5):514-20.
36. Sellers D, Mandy A, Pennington L, Hankins M, Morris C. Development and reliability of a system to classify the eating and drinking ability of people with cerebral palsy. Developmental Medicine & Child Neurology. 2014;56:245-51.
37. Powell CV, McNamara P, Solis A, Shaw NJ. A parent completed questionnaire to describe the patterns of wheezing and other respiratory symptoms in infants and preschool children. Arch Dis Child. 2002;87(5):376-9.
38. Trinick RE, Bunni L, Thorburn K, Hackett AP, Dalzell M, Mcnamara PS. An Observational Study Examining the Relationship between Respiratory Symptoms, Airway Inflammation and Bacteriology in Children with Severe Neurodisability. PLOS ONE. 2015;10(4):e0124627.
39. Owens JA, Spirito A, McGuinn M. The Children's Sleep Habits Questionnaire (CSHQ): psychometric properties of a survey instrument for school-aged children. Sleep. 2000;23(8):1043-51.
40. Markovich AN, Gendron MA, Corkum PV. Validating the Children's Sleep Habits Questionnaire Against Polysomnography and Actigraphy in School-Aged Children. Frontiers in Psychiatry. 2015;5.

41. Kleppang AL, Abildsnes E, Haraldstad K, Stea TH. Changes in health-related quality of life and sleep habits after a 6-month non-randomised cluster-controlled trial among children with overweight or obesity. European Child & Adolescent Psychiatry. 2024.
42. Furber G, Segal L. The validity of the Child Health Utility instrument (CHU9D) as a routine outcome measure for use in child and adolescent mental health services. Health and Quality of Life Outcomes. 2015;13(1):22.
43. Devlin N, Parkin D, Janssen B. An Introduction to EQ-5D Instruments and Their Applications. Methods for Analysing and Reporting EQ-5D Data: Springer International Publishing; 2020. p. 1-22.
44. rFLACC Pain Score – revised Version 1.1. Version date: 08/04/2020, Development lead: Jude Middleton https://media.gosh.nhs.uk/documents/Revised_FLACC_Paperwork.doc.pdf
45. Diaconu M, Bush A, Tan HL. Home sleep studies in children with neurodisabilities: success rates and parental perception. ERJ Open Res. 2024;10(2).
46. MHCLG O, NISRA, Scottish Government. Index of Multiple Deprivation (IMD) https://data.cdrc.ac.uk/dataset/index-multiple-deprivation-imd: Consumer Data Research Centre; 2019
47. Dasmahapatra P, Raja P, Gilbert J, Wicks P. Clinical trials from the patient perspective: survey in an online patient community. BMC Health Services Research. 2017;17(1).
48. Stans SEA, Dalemans RJP, De Witte LP, Beurskens AJHM. Using Talking Mats to support conversations with communication vulnerable people: A scoping review. Technology and Disability. 2019;30(4):153-76.
49. Eldridge SM, Chan CL, Campbell MJ, Bond CM, Hopewell S, Thabane L, et al. CONSORT 2010 statement: extension to randomised pilot and feasibility trials. BMJ. 2016:i5239.
50. Braun V, Clarke V. Using thematic analysis in psychology. Qualitative Research in Psychology. 2006;3(2):77-101.
51. Moore GF, Audrey S, Barker M, Bond L, Bonell C, Hardeman W, et al. Process evaluation of complex interventions: Medical Research Council guidance. BMJ. 2015;350(mar19 6):h1258-h.
52. Jones K, Weatherley H. Unit costs of health and social care 2022. Personal Social Services Research Unit (University of Kent) and Centre for Health Economics (University of York); 2023.
53. van Hout B, Janssen MF, Feng YS, Kohlmann T, Busschbach J, Golicki D, et al. Interim scoring for the EQ-5D-5L: mapping the EQ-5D-5L to EQ-5D-3L value sets. Value Health. 2012;15(5):708-15.

25. Appendices

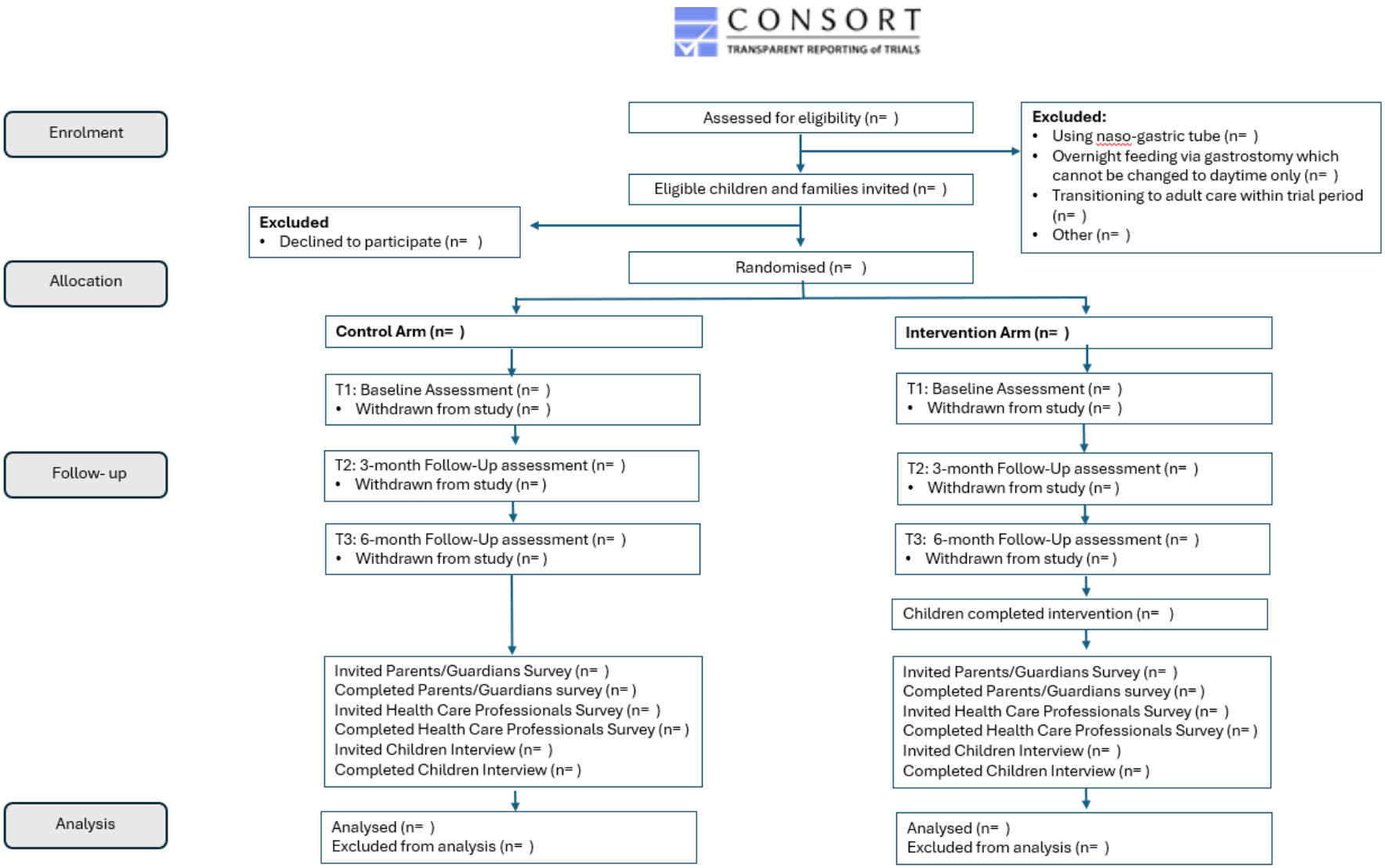
Appendix 1: Schedule of Events

	Screening	Randomisation	Baseline (0-months) Initial Home visit	Follow-Up 1 (3-months)	Follow-Up 2 (6-months)
Screening	X				
Eligibility check	X				
Informed consent	X				
Demographics	X				
Randomisation		X			
Notification of Randomisation		X			
Medical History (main diagnosis and co-morbidities)			X		
Functional motor ability level. Eating and drinking ability level. Using gastrostomy or jejunostomy. Overnight ventilation use. Medication. Seizures and impact on night-waking. GORD severity and impact on night-time positioning. Feed and medication regimes. Orthopaedic issues. Tissue viability (pressure) concerns. Usual sleep position and pattern. Night-time positioning equipment. Usual night-time care routine. Antibiotic use in last 6-months.			X		
Adverse Event Review		X	X	X	X
Withdrawal / non- participation survey (provided as soon as participant withdraws)	X	X	X	X	X

	Screening	Randomisation	Baseline (0-months) Initial Home visit	Follow-Up 1 (3-months)	Follow-Up 2 (6-months)
Assessments for intervention arm only:					
Introduce Breathe-Easy position - trial			X		
Daytime positioning trial			X		
Written protocol for nighttime			X		
Drainage bag supply established			X		
Parent/carer training for equipment use			X		
Sats monitoring during trial for first 2 full nights			X		
Weekly telephone contact for first 4 weeks			X		
After 4 weeks, 2-4 weekly calls			X	X	X
Assessments for control arm only:					
Investigate usual night-time position (usual care)			X		
Written night-time positioning guidance			X		
2-4 weekly calls			X	X	X
Assessments for both arms:					
Oxygen saturation			X		
HR / breathing rate			X		
Somnotouch (SCFT only)			X	X	
BRSQ			X	X	X
CSHQ			X	X	X
CHU-9D			X	X	X
EQ-5D-5L			X	X	X
rFLACC scale			X	X	X

	Screening	Randomisation	Baseline (0-months) Initial Home visit	Follow-Up 1 (3-months)	Follow-Up 2 (6-months)
Clinical health record data - 12month history: (no. of chest infections, time to first chest infection, x-rays, Abx use, hospital visits)			x		X
CYPCN Interviews (talking mats) (at end of study participation)					X
Process Evaluation surveys with HCPs, parents and carers (at end of study participation)					X

Appendix 2: CONSORT Flow Diagram



Appendix 3: Bespoke Respiratory Symptoms Questionnaire (BRSQ)

STUDY ID:

Respiratory Questionnaire for Children with Neurological Impairment

Alder Hey Children's NHS Foundation Trust

The following questionnaire asks about chest problems your child has had over the past **one month** and their impact on the family. It is important to try to answer all questions, even if your child has been perfectly well and the questions are not directly relevant.

Age of Child:

Mother/Father/Other completing questionnaire:

Was last month a typical month as regards your child's chest?

☐ Yes ☐ No

If no, why not?

During DAYTIMES over the last month:

1. Overall, how would you describe your child's breathing, chest or secretions?

☐ All good days ☐ Mostly good ☐ Some good, some bad ☐ Mostly bad ☐ All bad days

2. How frequently does your child have problems with their breathing (this could include noisy breathing, cough, difficulty in breathing, low oxygen saturations)?

☐ No problems at all ☐ Occasionally ☐ Sometimes ☐ A lot of the time ☐ All of the time

3. How frequently does your child have problems with respiratory secretions?

☐ No problems at all ☐ Occasionally ☐ Sometimes ☐ A lot of the time ☐ All of the time

4. What level of medical care do you give your child to help with their breathing?

☐ None ☐ Low ☐ Medium ☐ Quite High ☐ Very High

During NIGHT-TIMES over the last month:

5. Overall, how would you describe your child's breathing, chest or secretions?

☐ All good nights ☐ Mostly good ☐ Some good, some bad ☐ Mostly bad ☐ All bad nights

6. How frequently does your child have problems with their breathing (this could include noisy breathing, cough, difficulty in breathing, low oxygen saturations)?

☐ No problems at all ☐ Occasionally ☐ Sometimes ☐ A lot of the time ☐ All of the time

7. How frequently does your child have problems with respiratory secretions?

☐ No problems at all ☐ Occasionally ☐ Sometimes ☐ A lot of the time ☐ All of the time

8. What level of medical care do you give your child to help with their breathing?

☐ None ☐ Low ☐ Medium ☐ Quite High ☐ Very High

Respiratory Questionnaire for Children with Neurological Impairment Page: 1

IMPACT ON LIFE, HEALTH AND WELL BEING OF CHILD AND CARER

Over the last month:

9. How would you describe your child's energy levels?

☐ Normal /Usual ☐ Mostly normal ☐ Sometimes normal, sometimes low ☐ Mostly low ☐ Low/tired all the time

10. How has your child's chest affected other aspects of their health (e.g. seizures, bowels, weight)?

☐ No effect whatsoever ☐ Occasional effect ☐ Some effect ☐ Effect on many aspects ☐ Effect on most aspects

11. How has your child's chest affected their ability to do other things like going to school/nursery, respite, social events such as parties?

☐ No effect whatsoever ☐ Occasional effect ☐ Some effect ☐ Effect on many aspects ☐ Effect on most aspects

12. How has your child's chest affected THEIR ability to have a good night's sleep?

☐ No effect whatsoever ☐ Occasional effect ☐ Effect on some nights ☐ Effect on many nights ☐ Effect on most nights

13. How has your child's chest affected YOUR ability to have a good night's sleep?

☐ No effect whatsoever ☐ Occasional effect ☐ Effect on some nights ☐ Effect on many nights ☐ Effect on most nights

14. How has your child's chest (or the things you have to do to keep their chest well) affected things you do as a family?

☐ No effect whatsoever ☐ Occasional effect ☐ Some effect ☐ Effect on lots of things ☐ Effect on most things

15. How have your child's chest problems affected the family financially?

☐ Haven't caused a problem ☐ Caused occasional problems ☐ Caused some problems ☐ Caused lots of problems ☐ Caused significant hardship

Thank you for completing this questionnaire

Appendix 4: Child's Sleep Habits Questionnaire (CSHQ)

Child's Sleep Habits
(Preschool and School-Aged)
(Abbreviated Version)

_____
Coding

The following statements are about your child's sleep habits and possible difficulties with sleep. Think about the past week in your child's life when answering the questions. If last week was unusual for a specific reason (such as your child had an ear infection and did not sleep well or the TV set was broken), choose the most recent typical week. Answer USUALLY if something occurs **5 or more times** in a week; answer SOMETIMES if it occurs **2-4 times** in a week; answer RARELY if something occurs **never or 1 time** during a week. Also, please indicate whether or not the sleep habit is a problem by circling "Yes," "No," or "Not applicable (N/A)".

Bedtime

Write in child's bedtime: _____

	3 Usually (5-7)	2 Sometimes (2-4)	1 Rarely (0-1)	Problem?
1) Child goes to bed at the same time at night **(R)**	☐	☐	☐	Yes No N/A
2) Child falls asleep within 20 minutes after going to bed **(R)**	☐	☐	☐	Yes No N/A
3) Child falls asleep alone in own bed **(R)**	☐	☐	☐	Yes No N/A
4) Child falls asleep in parent's or sibling's bed	☐	☐	☐	Yes No N/A
5) Child needs parent in the room to fall asleep	☐	☐	☐	Yes No N/A
6) Child struggles at bedtime (cries, refuses to stay in bed, etc.)	☐	☐	☐	Yes No N/A
7) Child is afraid of sleeping in the dark	☐	☐	☐	Yes No N/A
8) Child is afraid of sleep alone	☐	☐	☐	Yes No N/A

Sleep Behavior

Child's usual amount of sleep each day: _____ hours and _____ minutes
(combining nighttime sleep and naps)

	3 Usually (5-7)	2 Sometimes (2-4)	1 Rarely (0-1)	Problem?
9) Child sleeps too little	☐	☐	☐	Yes No N/A
10) Child sleeps the right amount **(R)**	☐	☐	☐	Yes No N/A
11) Child sleeps about the same amount each day **(R)**	☐	☐	☐	Yes No N/A
12) Child wets the bed at night	☐	☐	☐	Yes No N/A
13) Child talks during sleep	☐	☐	☐	Yes No N/A
14) Child is restless and moves a lot during sleep	☐	☐	☐	Yes No N/A
15) Child sleepwalks during the night	☐	☐	☐	Yes No N/A
16) Child moves to someone else's bed during the night (parent, brother, sister, etc.)	☐	☐	☐	Yes No N/A
17) Child grinds teeth during sleep (your dentist may have told you this)	☐	☐	☐	Yes No N/A
18) Child snores loudly	☐	☐	☐	Yes No N/A

CSHQ Abbreviated

Coding

Sleep Behavior (continued)

		3 Usually (5-7)	2 Sometimes (2-4)	1 Rarely (0-1)	Problem?		
19)	Child seems to stop breathing during sleep	☐	☐	☐	Yes	No	N/A
20)	Child snorts and/or gasps during sleep	☐	☐	☐	Yes	No	N/A
21)	Child has trouble sleeping away from home (visiting relatives, vacation)	☐	☐	☐	Yes	No	N/A
22)	Child awakens during night screaming, sweating, and inconsolable	☐	☐	☐	Yes	No	N/A
23)	Child awakens alarmed by a frightening dream	☐	☐	☐	Yes	No	N/A

Waking During the Night

		3 Usually (5-7)	2 Sometimes (2-4)	1 Rarely (0-1)	Problem?		
24)	Child awakes once during the night	☐	☐	☐	Yes	No	N/A
25)	Child awakes more than once during the night	☐	☐	☐	Yes	No	N/A

Write the number of minutes a night waking usually lasts: _____

Morning Waking/Daytime Sleepiness

Write in the time of day child usually wakes in the morning: _____

		3 Usually (5-7)	2 Sometimes (2-4)	1 Rarely (0-1)	Problem?		
26)	Child wakes up by him/herself **(R)**	☐	☐	☐	Yes	No	N/A
27)	Child wakes up in negative mood	☐	☐	☐	Yes	No	N/A
28)	Adults or siblings wake up child	☐	☐	☐	Yes	No	N/A
29)	Child has difficulty getting out of bed in the morning	☐	☐	☐	Yes	No	N/A
30)	Child takes a long time to become alert in the morning	☐	☐	☐	Yes	No	N/A
31)	Child seems tired	☐	☐	☐	Yes	No	N/A

Child has appeared very sleepy or fallen asleep during the following (check all that apply):

		1 Not Sleepy	2 Very Sleepy	3 Falls Asleep
32)	Watching TV	☐	☐	☐
33)	Riding in car	☐	☐	☐

Appendix 5: Child Health Utility Instrument (CHU-9D)

Dimension	Level	Description
Worried	1	I don't feel worried today
	2	I feel a little bit worried today
	3	I feel a bit worried today
	4	I feel quite worried today
	5	I feel very worried today
Sad	1	I don't feel sad today
	2	I feel a little bit sad today
	3	I feel a bit sad today
	4	I feel quite sad today
	5	I feel very sad today
Annoyed	1	I don't feel annoyed today
	2	I feel a little bit annoyed today
	3	I feel a bit annoyed today
	4	I feel quite annoyed today
	5	I feel very annoyed today
Tired	1	I don't feel tired today
	2	I feel a little bit tired today
	3	I feel a bit tired today
	4	I feel quite tired today
	5	I feel very tired today
Pain	1	I don't have any pain today
	2	I have a little bit of pain today
	3	I have a bit of pain today
	4	I have quite a lot of pain today
	5	I have a lot of pain today
Sleep	1	Last night I had no problems sleeping
	2	Last night I had a few problems sleeping
	3	Last night I had some problems sleeping
	4	Last night I had many problems sleeping
	5	Last night I couldn't sleep at all
Daily routine	1	I have no problems with my daily routine today
	2	I have a few problems with my daily routine today
	3	I have some problems with my daily routine today
	4	I have many problems with my daily routine today
	5	I can't do my daily routine today
Work	1	I have no problems with my work today
	2	I have a few problems with my work today
	3	I have some problems with my work today
	4	I have many problems with my work today
	5	I can't do my work today
Able to join in activities	1	I can join in with any activities today
	2	I can join in with most activities today
	3	I can join in with some activities today
	4	I can join in with a few activities today
	5	I can join in with no activities today

Appendix 6: EuroQol EQ-5D-5L

Under each heading, please tick the ONE box that best describes your health **TODAY**

MOBILITY

- I have no problems in walking about ☐
- I have slight problems in walking about ☐
- I have moderate problems in walking about ☐
- I have severe problems in walking about ☐
- I am unable to walk about ☐

SELF-CARE

- I have no problems washing or dressing myself ☐
- I have slight problems washing or dressing myself ☐
- I have moderate problems washing or dressing myself ☐
- I have severe problems washing or dressing myself ☐
- I am unable to wash or dress myself ☐

USUAL ACTIVITIES (e.g. work, study, housework, family or leisure activities)

- I have no problems doing my usual activities ☐
- I have slight problems doing my usual activities ☐
- I have moderate problems doing my usual activities ☐
- I have severe problems doing my usual activities ☐
- I am unable to do my usual activities ☐

PAIN/DISCOMFORT

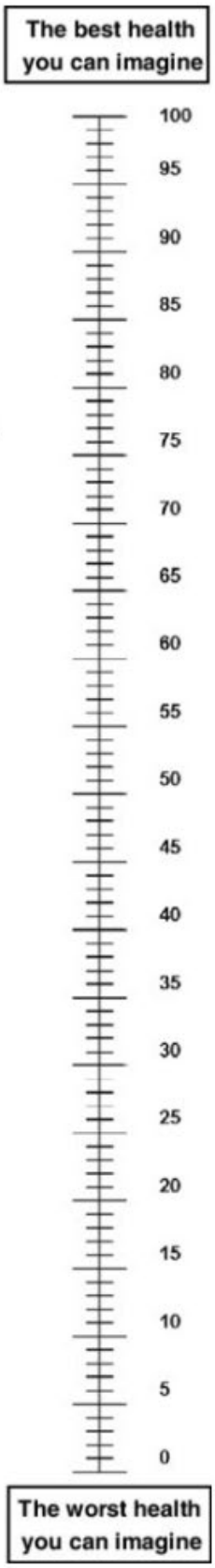
- I have no pain or discomfort ☐
- I have slight pain or discomfort ☐
- I have moderate pain or discomfort ☐
- I have severe pain or discomfort ☐
- I have extreme pain or discomfort ☐

ANXIETY/DEPRESSION

- I am not anxious or depressed ☐
- I am slightly anxious or depressed ☐
- I am moderately anxious or depressed ☐
- I am severely anxious or depressed ☐
- I am extremely anxious or depressed ☐

- We would like to know how good or bad your health is **TODAY**.
- This scale is numbered from **0** to **100**.
- 100 means the best you can imagine.
- Mark an **X** on the scale to indicate how your health is **TODAY**.
- Now, please write the number you marked on the scale in the box below.

YOUR HEALTH TODAY = ☐



Appendix 7: Revised Face Legs Activity Cry Consolability Pain Scale (rFLACC)

Name:	Hosp No:
DOB:	NHS no:

Revised FLACC Scale

Great Ormond Street NHS Hospital for Children NHS Foundation Trust

Categories	Scoring 0	1	2
Face	No particular expression or smile	Occasional grimace/frown; withdrawn or disinterested; ***appears sad or worried***	Consistent grimace or frown; frequent/constant quivering chin, clenched jaw; ***distressed-looking face; expression of fright or panic***
Individual Behaviours			
Legs	Normal position or relaxed; ***usual tone and motion to limbs***	Uneasy, restless, tense; ***occasional tremors***	Kicking, or legs drawn up; ***marked increase in spasticity, constant tremors or jerking***
Individual Behaviours			
Activity	Lying quietly, normal position, moves easily; ***Regular, rhythmic respirations***	Squirming, shifting back and forth; ***tense or guarded movements; mildly agitated (eg. head back and forth, aggression); shallow, splinting respirations, intermittent sighs***	Arched, rigid, or jerking; ***severe agitation, head banging, shivering (not rigors); breath-holding, gasping or sharp intake of breaths; severe splinting***
Individual Behaviours			
Cry	No cry/verbalisation	Moans or whimpers; occasional complaint; ***occasional verbal outburst or grunt***	Crying steadily, screams or sobs, frequent complaints; ***repeated outbursts, constant grunting***
Individual Behaviours			
Consolability	Content and relaxed	Reassured by occasional touching, hugging, or being talked to; distractible	Difficult to console or comfort; ***pushing away caregiver, resisting care or comfort measures***
Individual Behaviours			

(Adapted from Malviya et al, 2006)

Revised FLACC – Instructions for Use

- **Individualise the tool**: The nurse should review the descriptors within each category with the child's parents or carers. Ask them if there are additional behaviours that are better indicators of pain in their child. Add these behaviors to the tool in the appropriate category.
- Each of the five categories (F) Face; (L) Legs; (A) Activity; (C) Cry; (C) Consolability is scored from 0-2, which results in a total score between zero and ten.
- **Patients who are awake**: Observe for at least 1-3 minutes. Observe legs and body uncovered. Reposition patient or observe activity, assess body for tenseness and tone. Initiate consoling interventions if needed.
- **Patients who are asleep**: Observe for at least 5 minutes. Observe body and legs uncovered. If possible reposition the patient. Touch the body and assess for tenseness and tone.

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Appendix 8: Talking Mats Framework

Child will be offered each item below in a single symbol form which they then choose to arrange under one of three headings. The headings for the mats will be: “Like” “not sure” and “don’t like”. See **Figure 1** for illustration of completed mat.

1. I want to talk about different things that happen to you every day. I want to know what you think about these different things:
    - Waking up
    - Getting dressed
    - Sitting
    - Travelling in a car or bus
    - School
    - Mealtimes
    - Gastrostomy feeds
    - Music
    - Watching TV
    - Lying on your back
    - Lying on your side
    - Lying on your tummy
    - Getting ready for bed
    - Going to sleep at night

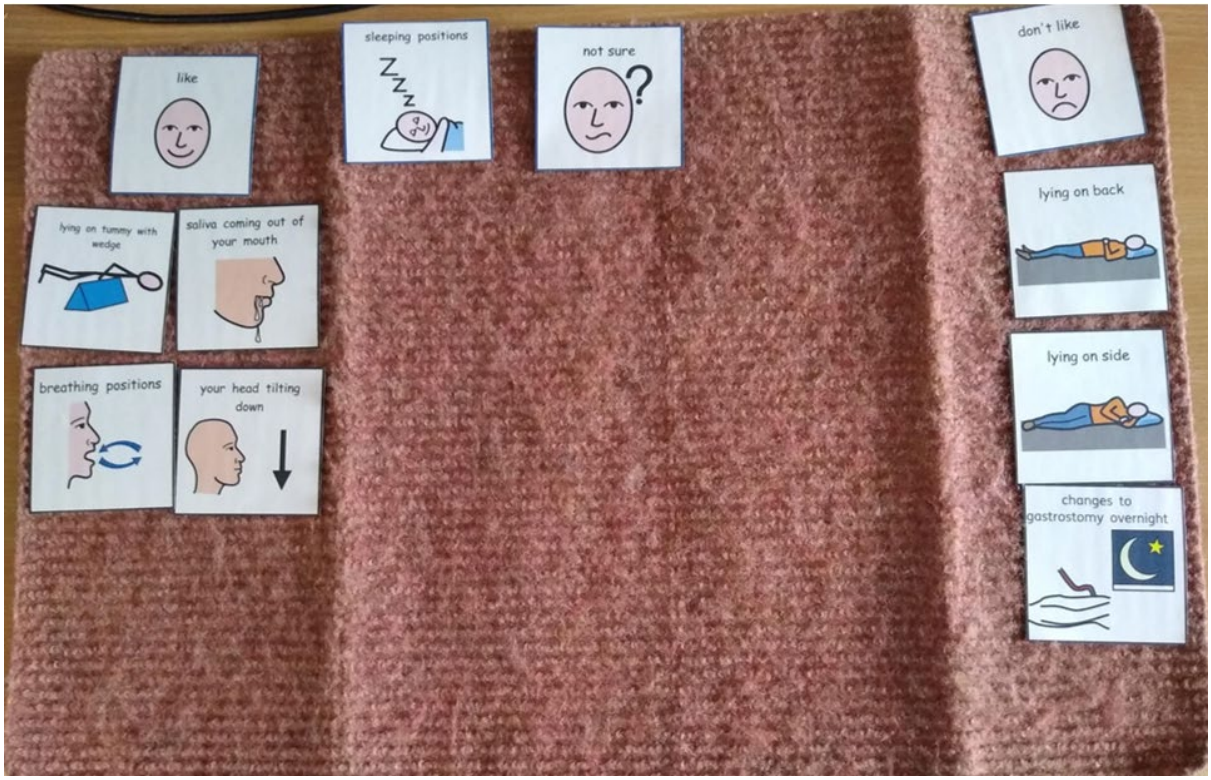
2. I want to talk about sleeping at night-time. What do you think about:
    - Going to bed
    - Lying on your back
    - Lying on your side
    - Lying on your tummy
    - Saliva coming out of your mouth
    - Saliva at the back of your throat
    - *Your head tilted downwards**
    - *Changes to your gastrostomy overnight**
    - Sleeping
    - Breathing
    - Sleeping at home
    - Sleeping away from home
    - Waking up

*Use question if child in ADAPT intervention arm.

Thank you for telling us what you think. I have a few yes and no answer questions for you.

3. *Do you want the night-time changes to carry on?**
4. Is there anything else you would like to tell us using your AAC book or device?

Figure 1:


like
sleeping positions
Zzz
not sure
?
don't like
lying on tummy with wedge
saliva coming out of your mouth
lying on back
breathing positions
your head tilting down
lying on side
changes to gastrostomy overnight

Appendix 9: GANTT Chart

RfPB reference: 39787 - ADAPT Dr Sarah Crombie, Dr Akshat Kapur et al. Feb 2025

	ADAPT – activity and deliverables	PRE-FUNDING	Months 1-3	Months 4-6	Months 7-9	Months 10-12	Months 13-15	Months 16-18	Months 19-21	Months 22-24
Set-up	Finalise protocol, PIS, study documents, survey questions; secure HRA, Ethics, R&D approvals, study data base									
	Site initiation visits: Core Trial Management Team, Core Clinical Team (PI and local team)									
	ADAPT training sessions at local sites									
	CTU management of study data									
WS1 Implementing Breathe-Easy Protocol 18 months	Recruit participants; individual participants randomised to Treatment or Control group by CTU n=50 (1st recruit – 42 days)									
	Local Clinical Teams collect / record baseline data for treatment and control group									
	Local Clinical Teams establish and oversee Breathe-Easy intervention with Core Trial Management Team support									
	Local Clinical Teams collect / record outcome measures for treatment and control group at baseline, 3 and 6 months									
	Collect 12 months' retrospective data from health records for each child, on completion of individuals' participation in trial									
	Analysis: CONSORT flow chart of study participants									
	Analysis: descriptive statistics of demographics, feasibility outcomes, candidate definitive trial outcomes									
WS2 Process Evaluation 18 months	Local Clinical Teams record case note diary for each case									
	Survey of parents / guardians who consented to participate but discontinued participation									
	Survey of parents/guardians and healthcare professionals, and interviews with children participating in completed ADAPT study									
	Thematic analysis of case notes to appraise contextual factors, process and quality of implementing protocol									
	Quantitative analysis of surveys: descriptive statistics									
	Thematic analysis of survey free text data									
WS3 Assessment of RCT feasibility	Assess feasibility of progression to definitive trial using pre-set parameters and analysis of contextual factors									
PPI Group	Advice on all aspects of the study including: set up, recruitment strategy, conduct of study, analysis and design of future RCT									
TSC / DMEC	Study oversight including: progress, adherence to protocol, patient safety and well-being, and advice to investigators.									
Management	Project Team Meetings									
Deliverables	Feasibility of definitive RCT of ADAPT; design of future RCT including selection of primary and secondary outcome measures									