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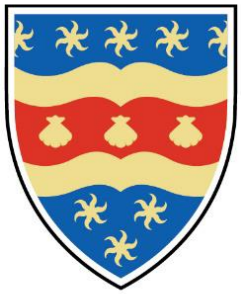
A novel interactive training device to improve walking Ability and quality of life for Children with CErebral Palsy Trial: ACCEPT study An exploration of balance, its measurement and usual physiotherapy care in children with cerebral palsy, to inform a mixed methods feasibility RCT.

Rapson, Rachel



UNIVERSITY OF PLYMOUTH

A novel interactive training device to improve walking **A**bility and quality of life for **C**hildren with **CE**rebral **P**alsy **T**rial: **ACCEPT** study

An exploration of balance, its measurement and usual physiotherapy care in children with cerebral palsy, to inform a mixed methods feasibility RCT.

By

Rachel Rapson

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Author's Declaration

At no time during the registration for the degree of Doctor of Philosophy has the author been registered for any other University award without the prior agreement of the Doctoral College Quality Sub-Committee.

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Rapson, R., Marsden, J., Latour, J., Ingram, W., Stevens, K.N., Cocking, L. and Carter, B., 2022. **Multicentre, randomised controlled feasibility study to compare a 10-week physiotherapy programme using an interactive exercise training device to improve walking and balance, to usual care of children with cerebral palsy aged 4–18 years: the ACCEPT study protocol**. *BMJ open*, *12*(5), p.e058916.

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Abstract

Rachel Rapson

A novel interactive training device to improve walking **A**bility and quality of life for **C**hildren with **CE**rebral **P**alsy **T**rial: **ACCEPT** study. An exploration of balance, its measurement and usual physiotherapy care in children with cerebral palsy, to inform a mixed methods feasibility RCT.

Cerebral palsy (CP) is a common disability with its onset in childhood, resulting from a non-progressive lesion to the brain. Motor impairments associated with CP are spasticity, weakness and reduced selective movement; these interfere with various aspects of balance. Children undertake physiotherapy to maintain or improve their mobility but find it hard to train at the correct frequency, intensity and in functional positions. A novel interactive trainer, the Happy Rehab™ (Innovaid, Denmark), was designed to address this. Its efficacy as an intervention to improve walking and balance is unknown. To evaluate this requires a Randomised Controlled Trial (RCT). The usual care to which it is being compared must first be defined. It is also essential to understand the mechanisms of balance in order to select an appropriate outcome measure. Current clinical tests for balance do not capture the selective deficits in postural control that occur during functional movements. Therefore, a new clinical test for measuring dynamic balance is required to determine the effectiveness of such interventions.

The main aim of the thesis as to determine the feasibility of a RCT investigating the clinical effectiveness of training for 10 weeks using the Happy Rehab interactive trainer and the impact on the lived experience and adherence to therapeutic exercise programmes by children with CP and their families. Four objectives were identified to meet this aim. Firstly, to determine the reliability and validity of a measure of dynamic balance to use as an outcome measure in the RCT. Secondly, to define and describe usual physiotherapy care to inform the control group of the RCT. Thirdly to establish the feasibility of an RCT using the Happy Rehab interactive trainer. Finally, to explore the experiences of children, parents and clinicians of their participation in the feasibility study to inform the effective delivery of a full RCT.

The thesis starts by exploring the mechanisms of balance used to inform the development of The Next Step test. It was developed through observations of the anticipatory postural adjustments (APAs) of children making a single step to medial and laterally placed targets. The validity and reliability of the Next Step test was established in (n=16) ambulant children with CP and (n=14) typically developing (TD) children. Children with CP had smaller medio-lateral APAs and greater stepping error. The grand average of the peak medio-lateral motion of the centre of pressure (ML COP) was identified as the Next Step summary score for use in the feasibility RCT.

Usual physiotherapy care was explored through a consensus process involving twelve physiotherapists. It used surveys, nominal group technique and a literature review. The resultant usual care checklist informed the control group of the feasibility RCT.

The feasibility RCT recruited fifteen children with CP, aged 8-18, with Gross Motor Function Classification System (GMFCS) levels I-III. They were randomised into two groups. The intervention group (n=8) undertook 10 weeks training using the Happy Rehab. The control group (n=7) carried out usual physiotherapy care. The Pediatric Balance Scale and the peak ML COP were assessed at baseline, 10 and 20 weeks. Families kept e-diaries to record adherence. Data were summarised using descriptive statistics. The PBS was more complete than the Next Step test at 10 weeks, PBS (100%) and Next Step (91%). Three children reached the ceiling of the PBS. Signals of efficacy of the intervention were detected in PBS, Next Step test as well as clinical measures of strength, spasticity and range of movement.

Qualitative interviewing was conducted with three physiotherapists, nine parent-child dyads, from the control group (n=4) and the intervention group (n=5) to understand participants' views on the feasibility of the trial and intervention. Data generation with the children was undertaken using semi-structured interviews incorporating a photo-elicitation method at the end of week 10. One decliner (n=1) interview was undertaken. Data were transcribed and analysed thematically using triangulation with the diary entries. Five main themes were identified from the interviews: 'Fitting therapy into normal life', 'Motivation to

exercise', 'The opportunity to try something new', 'Physios out of their comfort zone', and 'Altruism and the burden of participation'. Children found the gaming aspect motivating and enjoyable. Recruitment and the technical support for the intervention were impacted by the COVID pandemic.

In conclusion, the Happy Rehab intervention showed signals of efficacy for children with CP who are able to use the device at home for 10 weeks. However, some families could not accommodate the device at home. Physiotherapists required ongoing technical support to set up the device. The proposed RCT requires further work to establish a suitable primary outcome measure. A full RCT is not feasible without technical support for devices in the UK.

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Abbreviations

ANOVA	Analysis of variance
AP	Antero-posterior
APA	Anticipatory Postural Adjustment
ASD	Autistic spectrum disorder
ASIS	Anterior- superior iliac spine
BESTest	Balance Evaluation Systems Test
BOS	Base of support
COM	Centre of mass
COP	Centre of pressure
CNS	Central nervous system
CP	Cerebral palsy
GMFM	Gross Motor Function Measure
GMFCS	Gross Motor Function Classification System
HAT	Head and trunk
ICC	Intra-class correlation coefficient
IQR	Inter-quartile range
MDD	Minimal detectable difference
MDM	Mean deviation from median
ML	Medio-lateral
MRC	Medical Research Council
MTP	Metatarsal-phalangeal joint
NICE	National Institute for Clinical Excellence
NHS	National Health Service
NIHR	National Institute for Health and Care Research
PBS	Pediatric Balance Scale
QFM	Quality Function Measure
RCT	Randomised controlled trial
ROM	Range of movement
SD	Standard deviation

SEBT	Star Excursion Balance Test
SEM	Standard Error of Measurement
TD	Typically developing

1 Part 1: Introduction

In this chapter, I introduce cerebral palsy and its impact on children and their families. I will outline the aims of the thesis and describe the methodological and epistemological approaches used to achieve these aims.

1.1 Introduction to cerebral palsy and the problems with measuring and treating associated balance problems

Cerebral palsy (CP) is a relatively common disability with its onset in childhood, affecting 2.1 per 1000 children worldwide [1]. Swedish population studies show that there is a higher prevalence of CP in males compared to females at a ratio of 1.4:1, but there is no difference in levels of manual or motor function between genders [2]. Cerebral palsy is an umbrella term describing a group of permanent disorders affecting the development of posture and movement resulting from a lesion to the brain [3]. The Gross Motor Function Classification System (GMFCS) provides a common language to describe and predict motor ability. It has five levels (I-V). Children within GMFCS I have high level walking and balance skills, children within GMFCS II need to hold onto a rail to manage stairs, while children within GMFCS III require walking aids and may use a wheelchair for longer distances [4]. Children within GMFCS IV and V do not have functional ambulation. My thesis explores the rehabilitation of balance and walking and so the focus will be on children with GMFCS levels I-III [5].

Motor impairments in CP are usually described as unilateral or bilateral, and are subdivided by motor presentation into spastic, ataxic or dyskinetic CP [6, 7]. Bilateral spastic CP is the most common presentation [8]. Motor impairments associated with CP are spasticity, weakness and reduced selective movement; these interfere with various aspects of balance. Spasticity is commonly defined, after Lance, as a “motor disorder characterised by velocity dependent increase in tonic stretch reflex activity...” [9]. In addition to the primary motor impairment, secondary musculoskeletal impairments may develop. The cause of these musculo-skeletal changes are widely attributed by the clinical community to the presence of spasticity. While this view may be shared amongst clinicians, the suggested causal link is not evidenced by the research literature. Contractures most commonly occur in muscles spanning two joints, such as iliopsoas, hamstrings, gastrocnemius and quadriceps. Bony deformities, such as tibial torsion and femoral anteversion, may develop secondary to the imbalance in the imposed moments/torques in the child’s musculoskeletal system. These secondary complications often require surgery to lengthen muscles and correct bony deformity, which can be traumatic for the child as well as causing them to miss schooling and disruption to their parents’ employment. The cost of a single

episode of surgery to improve gait function in children with CP varies from £24,301-£29,679 per child [10]. Reducing financial costs and the impact of surgery on the child (e.g. pain, schooling and quality of life) [11] through effective physiotherapy is an important goal.

Children with CP experience multiple challenges to normal balance when standing and have reported that even trying to stand still can be fatiguing [12]. Young people have reported how pain, limited physical functioning and fatigue restrict their ability to participate in everyday activities [13]. Difficulties with walking and balance are common and can limit integration in schooling and independence in functional activities [14]. Children with CP, for example, only spend 3.4 hrs/week engaging in physical activity, nearly half that seen in typically developing children [15].

Clinical tests for balance and walking in children with CP mainly measure whole body movements and do not capture the selective deficits in postural control mechanisms that occur during functional movements [16, 17]. Compensatory strategies for poor balance may achieve short-term functional aims while being detrimental in the longer term. For example, lateral trunk lean and increased step width may be a functional compensation for poor stability in gait, but this becomes a problem when a child wants to move more quickly [18]. There is a need for an effective outcome measure that can measure postural control during balance in standing and walking. In order to develop effective measurement tools and treatment options, it is necessary to first observe and understand how impairment profiles effect balance [19]. Novel therapeutic interventions require evaluation using accurate and valid outcome measures that are sensitive enough to detect change. Thus, Part 2 of this thesis provides a narrative review of the pathophysiology of balance impairments in CP, and an overview of current methods of measuring balance in CP. This informs the development and evaluation of a novel measure of dynamic balance in CP- the Next Step test. As part of the evaluation of the Next Step, the reliability and construct validity will be determined and compared to the Quality Function Measure [20]. Its measurement properties will be compared through observations of the impairment profile and functional severity of children with CP and typically developing peers.

Children with CP usually undertake daily therapeutic exercise to help improve balance and walking. As walking deficits involve interactions between multiple

impairments, targeting one symptom in isolation may not produce effective results. A meta-analysis of manual passive stretching, for example, highlighted their ineffectiveness in reducing contracture and spasticity [21]. In light of this work, the National Institute for Clinical Excellence (NICE) recommends low-load active or sustained passive stretching using orthoses or equipment. Although the NICE clinical guideline [22] recommends progressive muscle strengthening programmes, studies have inconsistently demonstrated gains in function due to variability in the components of rehabilitation programmes [22]. There is no consensus on physiotherapy exercise interventions to promote balance for children with CP [17].

Adherence to home exercise can present multiple challenges to family life. Rates of adherence to home exercise differ depending on the child's motivation, parental support and the child's environment [23]. A novel intervention, called the Happy Rehab, (Innovaid™) (Figure 1) integrates a supportive standing frame with an exercise device which has a series of games controlled by leg movements. The device offers a motivating way for children with CP to train in a functional standing position, with less reliance on adult support The child operates a series of games by moving the footpads (dorsiflexion and plantar flexion) and kneepads (forwards and backwards) . Additionally, sensors under the feet allow certain games to be operated by shifting the weight side to side. The device has motors that assist or resist the movement at the knee and ankle depending on how the device is tailored to the user's needs. It has the potential for users training on the device to target strength, range of motion, selective motor control and balance. However, it currently lacks evidence to show whether training in the device can improve balance and walking and have an effect on other impairments [24].

Figure 1 Happy Rehab, (Innovaid™)

A first step to testing the efficacy of this equipment was to see if a Randomised Controlled Trial (RCT) is feasible [25, 26] and to find out if the device is acceptable to young people, their parents and physiotherapists. When undertaking a RCT the intervention of interest is compared to a control group and frequently this is usual care. However, the comparator is often poorly defined in trials. Thus, in Part 3, I report on a consensus study and a narrative review of the literature, undertaken to define the usual care. The resulting definition informs the feasibility RCT undertaken to explore an intervention using the Happy Rehab, (Innovaid™) compared to usual care. The feasibility trial used a mixed-methods approach with an embedded qualitative component. Amongst other feasibility aims, the trial assesses the potential of using the newly developed and evaluated the Next Step test of dynamic balance as a primary outcome measure.

1.2 Aims and objectives of the programme of work

1.2.1 Research Question

The overall research question is as follows.

What is the feasibility of an RCT investigating the clinical effectiveness of training for 10 weeks using the Happy Rehab interactive trainer and the impact

on the lived experience and adherence to therapeutic exercise programmes by children with CP and their families?

1.2.2 Objectives

The objectives of this programme of work are to:

1. determine the reliability and validity of a measure of dynamic balance to use as an outcome measure in the RCT

2. define and describe usual physiotherapy care to inform the control group of the RCT

3. establish whether an RCT using the Happy Rehab interactive trainer is feasible

4. explore the experiences of children, parents and clinicians of their participation in the feasibility study to inform the effective delivery of a full RCT

1.3 Design and methodology of the programme of work

The ultimate aim of this programme of work is to test the feasibility of an RCT. However, in order to do this, it is necessary to develop a suitable primary outcome measure and establish the usual physiotherapy care of the children within the study. Therefore, this programme of work uses several different research methods. Firstly, the development and evaluation of the psychometric properties of a novel outcome measure, secondly a consensus approach informed by a narrative review of the literature and finally a mixed-methods feasibility trial. This section addresses the theoretical approaches and methodological considerations used in the design of these studies.

When considering which methodological approaches would help to answer the research question, it was necessary to consider the theoretical underpinning of the methods used to gain this new knowledge. Epistemology is the branch of philosophy concerned with knowledge and explaining how ‘we know what we know’, and methodology describes the processes needed to gain that knowledge [27]. The awareness of the strengths and limitations of each approach is important as it sets the knowledge gained in the context of the chosen methodologies. The epistemological and methodological underpinnings for each study within the programme of work is outlined in Table 1 [27].

Table 1 *The epistemological and methodological underpinnings of the different studies within the programme of work presented in this thesis.*

Phase of work	Epistemology	Theoretical Perspective	Methodology	Methods
Phase 1: Developing a balance outcome measure	Objectivism	Positivism	Experimental design	Observation Measurement Data reduction Statistical analysis
Phase 2: Defining usual care	Objectivism	Post-positivist	Survey Nominal group consensus	Focus group questionnaire
Phase 3: Feasibility RCT	Objectivism	Post-positivist	Experimental design	Measurement Data reduction Statistical analysis
Phase 4: Qualitative feasibility study	Constructivism	Interpretivism	Qualitative	Interview Thematic analysis

A positivist approach is used in the first phase of the work, which uses an observational study to explore the reliability and validity of a novel measurement of dynamic balance. This scientific method adopts a positivist standpoint to establish an unambiguous, objective knowledge that can be posited, rather than arrived at speculatively [27, 28]. It is built upon three foundations: *empiricism,* which relies on scientific enquiry using observations and evidence; *determinism* which states that all things follow a common set of laws; and *scepticism* which means that all propositions are open to analysis and critique, even statements proposed by great authorities [29]. In order to measure balance, empirical data is collected in a standardised way. This reductionist standpoint allows objective reality to be tested by controlling other variables and excluding the researcher's values and biases. The units of time, velocity and distance are usually used to measure balance, and these follow accepted, deterministic, common laws of

physics. The approach used to measure balance was an observational study (phase 1) and was designed by building on the knowledge and modelling the work of other scientists [30, 31]. However, it was important to remain sceptical of the results of previous work until the results of this study supported or refuted these previous findings. One weakness of this approach is that children are very variable, even those with a similar impairment, this meant we were unable to control for their individual responses during the experiment.

Post-positivist philosophy uses critical realism to recognise the limitations of the positivist approach by recognising the fallibility of observations, for example, biases, measurement error or the variety of children's personalities or behaviours. Some philosophers question the validity of making claims based on a limited set of observations, as this limits the generalisability of the results beyond the defined population who were observed [29]. The post-positivist paradigm rejects the received wisdom that scientific methods such as randomised controlled trials are at the top of the evidence hierarchy [32]. This is particularly true when undertaking experiments that take account of human behaviour, for example when studying rehabilitation programmes [28]. A pragmatic approach to research recognises that where the positivist approach may strip the knowledge from its context and thereby reduce its meaning, combining it with qualitative research can redress this imbalance [28]. The proposed feasibility study does not try to standardise and control all variables in the child's therapeutic intervention. However, observations of potential confounding variables are recorded and considered along with the final analysis of the trial.

Physiotherapists naturally blend both qualitative and quantitative approaches when making informed decisions about patient care. Thus, it is unsurprising that a similar approach is seen in physiotherapy research [32, 33]. Shaw et al (2010) argue the best evidence to inform practice in physiotherapy uses a pragmatic paradigm, combining qualitative and quantitative data from a research perspective. The pragmatist paradigm uses a variety of different methods to derive meaningful results, applicable to the patient group studied [32, 33]. It uses a series of inquiries in a more natural and relevant setting, triangulating the data to determine new knowledge [32].

The second phase (Table 1) in this thesis is approached from a pragmatic standpoint and used several methods to gather qualitative and quantitative data to establish usual care. This used a more relational and subjective process where participants of nominal groups construct meaning together in an iterative process, to form a consensus opinion. The physiotherapists' experience in paediatrics was considered through the sampling process to capture the length of post qualification and paediatric experience of participants. The soundness of the consensus decisions was verified by conducting a systematic search of the literature for the usual care interventions, to check the new knowledge against the established evidence base. The results were triangulated with participants to gain their final consensus on whether to include interventions into a usual care checklist.

The overarching aim of the programme of work reported in this PhD thesis was to establish the feasibility of an RCT investigating the clinical effectiveness of the interactive trainer and the impact on the lived experience and adherence to therapeutic exercise programmes by children with CP and their families. This necessarily demanded a mixed-methods approach to combine quantitative and qualitative data (phases 3 and 4). The mixed-methods approach to research into a rehabilitation intervention, allowed the researcher to take account of the personal factors of the person undertaking the programme as well as more objective outcomes. This is a relatively new approach which has gained popularity in modern research [33, 34]. There is some criticism of researchers who mix methodologies that originate from diametrically opposing epistemologies [32, 34]. However, others researchers argue that there is an unnatural divide between qualitative and quantitative research and that research can only be advanced by combining methodologies [35].

The feasibility study was designed using an RCT with embedded qualitative study. The quantitative results and qualitative findings are reported separately but triangulated and synthesised in the final discussion of the thesis. The contrasting epistemologies are consciously respected by clearly separating the methods used and by seeking supervision from experts in each of the paradigms. The intervention itself can be defined as a complex intervention according to the Medical Research Council (MRC), as it has a number of interacting components [25]. These include the variety of settings in which the

device might be used, as well as the individual tailoring of the games and exercise prescription orientated to the children's individualised goals. The MRC framework describes the need to test feasibility to determine the acceptability of the intervention, to test the processes in the protocol and to examine the appropriateness of the proposed outcome measures [25]. Feasibility testing is a dynamic, iterative process that involves the views of stakeholders to design a trial that can be implemented in a real-world setting [26]. Testing the feasibility of the study's safety, and parameters such as the willingness of participants to be randomised and of clinicians to recruit, is essential before seeking funding for a full RCT [36, 37].

The next part of my thesis, Part 2, will focus on balance problems in children with CP and the development of the Next Step measure of dynamic balance.

2 Part 2: Explorations

Part 2 explores the mechanisms of postural control and balance, the development of them in childhood, and how they differ in children with cerebral palsy.

Current clinical tools for measuring balance are explored and the rationale for developing a novel measure of dynamic balance is explained. It will be proposed as a potential primary outcome measure for the feasibility study reported in Part 3 of this Thesis.

Finally, two papers are presented: an observational study of dynamic balance using the Next Step test and a study reporting the reliability of the Next Step test.

2.1 Mechanisms of Postural Control and Balance

Children with CP experience instability when walking especially when they are simultaneously carrying out a motor or cognitive task. These dual tasks lead to slower, more complex and variable gait patterns [38]. The impact of this instability means that everyday tasks are more difficult, and falls are more likely. It is necessary to understand the models of posture and balance in adults, and the development of posture and balance to address these problems in children with CP.

2.1.1 What is balance, equilibrium, and posture?

Posture refers to the organisation of body segments in relationship to each other as well as in relationship to the environment, whether the person is standing on the ground, in water, or on a moving surface [39]. Postural control mechanisms fix the orientation of the body to the environment while also orientating the body segments to each other to stabilise the centre of mass (COM) of the body and retain postural alignment. This requires integration of multi-sensory information within the central nervous system and appropriately timed and scaled postural responses. The 'postural body schema' is a person's internal representation of their body geometry and their orientation to the vertical that is used to control posture [40].

Antigravity postural control in standing relies on organising the body segments into a stable posture. A body is stable when its COM lies over its base of support (BOS), where the BOS is the area around the outside of all the points where the body contacts the surface. As long as this condition is met then the body will easily return to its starting position when it is perturbed, due to a coupling of the weight of the body and the ground reaction force [40]. When the COM lies outside of the BOS the coupling will cause the body to move to a new position. This is usually a step or a fall.

When standing still, the human body seems to be in a state of static equilibrium. However, the body is rarely still. It makes constant automatic adjustments using 'postural synergies' in order to stabilise the COM [40]. While moving, the body can be said to be in a state of dynamic equilibrium [41]. Dynamic equilibrium involves coordinating the prime task-orientated movement with complex

background postural movements from which functions such as looking, reaching and stepping can be performed [40, 42].

2.1.2 When does balance occur in humans?

In standing, the human body is inherently unstable, with a high COM and a small BOS. When standing in static equilibrium, postural adjustments are seen as a postural sway. Normally people sway mainly around the ankles like an inverted pendulum, with the motion controlled about the multiple joints of the body. People need to balance in response to an external perturbation, such as the jolting movements experienced when standing on a bus. In this situation, the person needs to be able to sense the perturbation and respond with muscles synchronised to contract with appropriate force to bring the body back into equilibrium. People also need to balance when moving a body part such as the arms or legs. In this situation, the movement of the limbs away from the COM can itself cause disequilibrium. Finally, humans must be able to recover balance during trips and falls by producing effective saving reactions.

2.1.3 Factors required for normal postural control in adults

The body requires certain tools to maintain equilibrium. Firstly, flexible joints and muscles and a certain amount of muscle activity are needed to maintain joint alignment and to produce sufficient torque around the ankle. The muscle synergies which comprise postural reactions most often occur in muscles which extend across two joints, such as the hamstrings, biceps femoris and gastrocnemius [40]. These are the same muscles where spasticity is frequently found in children with CP.

Postural strategies during quiet standing normally involve movements around the ankle, termed an “ankle strategy”. However, postural strategies involving hip flexion-extension in combination with ankle motion can be seen where the ankle mechanism is ineffective at controlling the ‘inverted pendulum’ or in highly challenging novel situations, such as when walking along a balance beam. When walking on such a narrow support, the COM is close to the edge of the BOS and fast corrections of the COM are required. Such a “hip strategy” can also be seen when there is stiffness and weakness around the ankle [42]. The ankle and hip strategies are part of a continuum responding to ever-increasing challenges to balance. The ankle strategy is usually employed first, with the addition of the hip response and finally stepping, when the limit of stability is reached [40].

In addition to flexibility and strength, the human body needs intact sensory systems including proprioceptive, visual and vestibular pathways [43]. The central nervous system (CNS) integrates visual, somatosensory and vestibular information to support postural control at multiple levels in the brain and spinal cord. In the cerebral cortex multi-sensory information is used to create a reference frame of postural verticality that can be considered to be part of our postural body schema [44]. At lower levels of the CNS, somatosensory inputs from the legs trigger and scale the level of postural reactions [40]. Proprioceptive information from extensor and flexor muscles and tendons regulate the timing of stance and swing phases while walking, while skin afferents on the feet are important for detecting and triggering automatic responses to unexpected obstacles [44]. Fast acting stretch reflex activity generated via spinal and transcortical circuits play a major role in postural control when standing and walking. In contrast, the vestibular system, acting via the vestibulospinal tract, does not influence postural adjustment during small disturbances or trigger responses, but does affect the use of the hip strategy during larger disturbances to equilibrium [44].

In adults, proprioceptive information and visual information are the main sensory sources used to maintain balance [45]. However, the sensory pathway or channel that is used depends on environmental factors and development. Young children, for example, rely more heavily on vision as a stabilising factor to help orientate the body in a standing position [39]. In adults, the system is flexible and the relative importance of sensory information in maintaining balance can vary. Vestibular information becomes more important in the dark, or when proprioceptive and cutaneous information from the feet and ankles becomes less reliable. This happens when standing on an overly compliant surface. Vision is used preferentially when standing with a narrow stance and somatosensory inputs are prioritised over vision when standing with a wide-based stance [40]. This ability to use sensation flexibly to aid balance is called sensory re-weighting [45]

Balance has been extensively investigated both clinically, by the study of disease and experimentally, by investigating the physiology of movement. In the following sections, the mechanisms of postural sway and aspects of dynamic equilibrium are explored. Additionally, how the body responds to perturbations have been

examined. This includes the use of force plates, moving platforms or by monitoring of the activity of muscles while applying forces to the arm or leg.

2.1.4 Postural sway in adults

Postural sway can be measured using a force plate to track the position of the applied resultant force, termed the centre of pressure (COP). The COP represents the applied forces that control the position of the COM. Typically, as the line of the COM travels forward within the BOS, the COP position moves further anteriorly, to bring the COM back more centrally and vice versa for posteriorly directed COM motion. This creates a pattern of postural sway, where the oscillation of the COP is always greater than that of the COM in order to maintain equilibrium[42]. The movement of the COP is bounded by the area in contact with the ground. The further away the COP is from the axis of rotation, the greater the moment that is required to restore equilibrium. If insufficient force can be generated before the boundary of the BOS is reached, the person must take a step or fall over. As previously mentioned, postural sway of the body can be explained by using the model of an inverted pendulum, controlled around the ankle. Gravity pulls the ‘pendulum’ forward, and the body is pulled posteriorly by tension of the plantar flexors, generated by active or reflex contraction plus the passive tension generated by stretching the calf muscle-tendon complex. Thus the COP is returned to a position between the heel and the toe [42]. This basic model begins to explain the mechanism of postural sway, but further work has shown that varying the stiffness of the calf muscle-tendon complex alone is not sufficient to produce the torque about the ankle required to perform this function. This is because the Achilles tendon, in series with the plantar flexor muscles, is too compliant. To accommodate the compliant Achilles tendon, a series of continuous ballistic contractions of the soleus and gastrocnemius is required to keep the inverted pendulum of the body upright [46, 47].

This mechanism is akin to keeping a balloon up in the air by tapping it. The size and timing of the tap responds to the speed and position of its descent by anticipating the force needed to keep it in the air. Thus the postural sway of the body is described as a never ending cycle of trial and error and requires the central control and multi-sensory inputs described earlier [47]. Postural sway ultimately enables people to remain upright at ‘rest’ but equally ready for action.

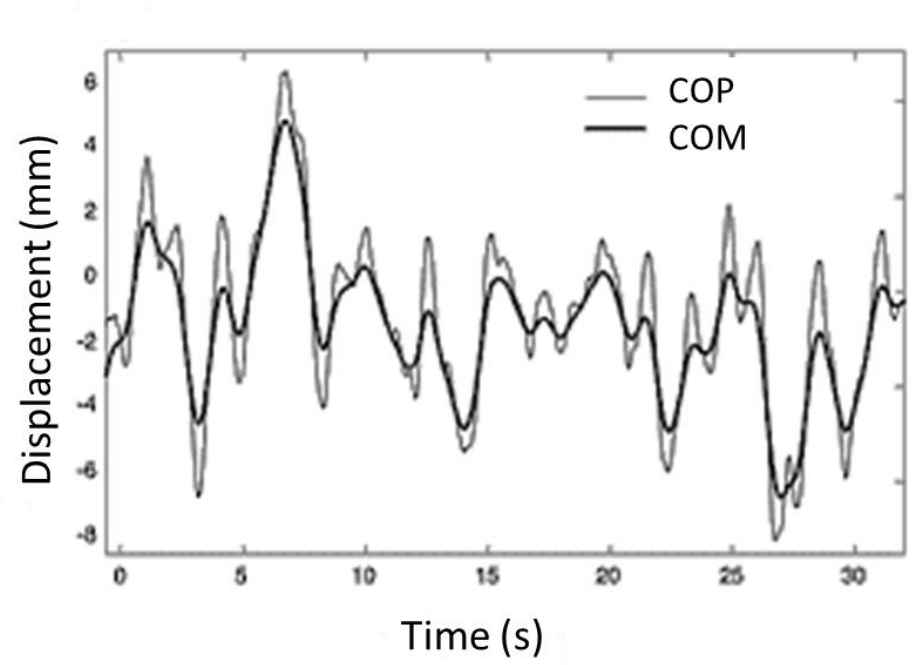

Figure 2 *Path of displacement of the centre of mass (COM) and centre of pressure (COP) during postural sway in standing [12]*

2.1.5 Postural reactions following an external perturbation

Moving platform experiments explore the body's response to sudden perturbations by measuring the electromyography signal of postural muscles. When a seated person is displaced forwards or backwards by suddenly moving the platform, direction specific postural muscles are activated, followed by fine tuning of the response modulated by sensory and supraspinal feedback [43]. Nashner's early experiments in standing balance (1977) measured activity in the leg muscles in response to displacements of the platform forwards, backwards vertically and on an incline [48, 49]. The muscles closest to the moving platform activate first in a distal to proximal coordinated muscle response. Nashner went on to explore conflict in sensory inputs by creating experimental rooms that move in unison with either the sway of the body or the incline of the platform. When the room appears to remain still but the platform tilts, the activity of the gastrocnemius reduced as the body was 'tricked' into sensing no movement, but with repetition the body adapts restoring the correct amplitude of response to maintain equilibrium [49].

2.1.6 Anticipatory postural adjustments associated with voluntary movement

The most common type of challenge to the body's equilibrium comes in the form of internal perturbations such as pulling or pushing a load while standing still or moving a limb. Here, the body produces quick anticipatory postural adjustments (APAs). Unlike other elements of postural control, these APAs precede disturbance of the body's equilibrium [40, 42]. While lifting an arm, muscle activation stabilises the legs, trunk and shoulder joints before lifting the hand up. This is followed by further instantaneous, direction-specific movements to stabilise the position of the COM as the arm reaches outside of the body's BOS [50]. The size of APAs vary in response to the predicted load or perturbation, for example, when anticipating lifting either a feather or a heavy box up from the floor.

Anticipatory postural adjustments also occur prior to taking a step. The assumption might be that this is to move the weight over the stance foot to release the stepping foot. However, studies have shown that the COM does not transfer fully over the stance leg, but remains medial to the COP within the foot, leaving the body unstable and falling medially away from the stance foot [42]. The 'throw and catch' model of dynamic balance explains this phenomenon. It proposes that the postural changes seen during pre-step activity comprise a throw of the COM initially towards the stance leg, on a ballistic trajectory towards the target step [30]. The size and direction of the throw of the COM has been shown to usually couple with the target position of the stepping foot. When the target is moved after the throw has been initiated this coupling can be disrupted [31], for example when avoiding stepping on a piece of Lego that is only seen mid-swing.

This 'throw and catch' model of dynamic balance is therefore fundamental to maintaining equilibrium while stepping and walking. It will be the focus of the development of a novel measure of dynamic balance later in this chapter.

2.1.7 Balance and postural control in the real world

The above description has broken down balance control into several components. The reality is that they are all occurring simultaneously in everyday life. Humans are usually unaware of the subtle changes in postural control during simple movements such as turning one's head to look or picking up a cup to drink. When the limits of stability are reached, the body produces automatic reactive

postural adjustments to regain stability. A common experience of loss of balance might be when turning over on one's ankle. The sensory systems send signals as the body moves out of equilibrium. The stretch on the soft tissues of the ankle, the altered pressure on the foot, the visual orientation to the horizontal and the vestibular system, all warn of falling. Once out of the state of dynamic equilibrium, reactive postural adjustments come into play. The righting responses of the head on trunk and trunk on limbs, followed by the activation of automatic saving responses of stepping and throwing out outstretched arms to break the fall. This sequence is automatic and infrequent in most healthy adults.

In the next section, I will explore the development of posture and balance in children and examine the importance of falling in order for children to learn stability.

2.2 Development of postural control and balance in childhood

Prior to understanding how postural control and balance differs in cerebral palsy (CP), it is important to understand the normal development of these phenomena. The most widely acknowledged model of motor learning is the 'neuronal group selection' theory. This combines the idea of innate factors, such as central pattern generators that generate the rhythmic activity associated with walking, with the evolution of sensorimotor pathways, where successful pathways are selected and less useful connections are pared away [51, 52]. Observations of neonates reveal that they display generalised fidgety movements, developed in utero [53]. These fidgety movements demonstrate huge variability and are not initially produced in response to stimuli. They allow the infant to learn from the sensory-motor experience of a large repertoire of movements. Later the general movements are seen to change in response to stimuli in a more goal directed way. Over time, the body selects the neuronal sets which result in successful achievement of goals [51]. For example, an infant's accidental batting of a toy, causes them to attend to the interesting sensory stimuli. Over time, and with repetition, the infant develops an intentional reach and grasp movement.

The variability of movement continues throughout motor development. When infants learn to sit, then stand and walk, they move constantly within each posture. They feel and explore movement and their ability to function within each position. The mastery of each posture in supine, prone, rolling, sitting and

standing all interrelate. For example, development in sitting relates strongly to the control of the pelvis in lying, where the infant typically plays with their toes, developing the abdominal and core muscle strength needed to control their sitting posture [54].

2.2.1 Postural sway in children

Postural sway in standing develops and matures with age. Children, like adults generally have increased postural sway with eyes closed [55]. The length of the path of postural sway in standing is larger in children aged 5-6 years and reduces with age [55]. Antero-posterior COP sway is the most stable measure across the age range of 5-18 years, suggesting that the postural action of soleus matures early on in childhood to control sway in standing [55]. As the child approaches adulthood, the speed of displacement of the COP further slows; demonstrating more advanced postural control and improved medio-lateral control.

2.2.2 Development of postural control in response to external perturbations

Moving platform experiments with infants, as young as one month, reveal innate patterns of direction specific postural responses in lying. They show the ability to adapt these innate responses when sitting at around 6 months. They initially show development of coordinated postural responses in the dorsal and then in the ventral muscles [51]. This phased development of postural muscle activity may be related to experience, as most young infants repeatedly fall forward when sitting before gaining reliable independent sitting. This repeated falling forwards stimulates the activity and development of postural control by the dorsal muscles of the spine to regain upright sitting.

The development of direction specific postural adjustments in sitting appears to be a prerequisite for the development of standing. A wide repertoire of direction specific and fine postural adjustments are observed in young children who successfully transition from supported to free standing [56]. Further platform perturbation studies in supported standing reveal that children as young as nine months can develop ‘complete’ direction specific postural responses in standing, when exposed to training using 300 repeated platform perturbations over just three days [57]. As the child learns to pull to stand, it is notable that toddlers use hip flexion to keep their body upright and stable, while their body remains over-compliant. Young toddlers initially lack the torque generation associated with the

ankle strategy to control their ‘inverse pendulum’. Moving platform experiments in children aged 5-12 years explored their dynamic and reactive balance in standing [58]. Children were able to maintain postural stability for longer without needing to take a step, the older they were. This may be due to the increased potential for plantar flexor force to produce the required torque around the ankle to stabilise their COM [58].

2.2.3 Development of anticipatory postural adjustments associated with movement

Children as young as ten months produce anticipatory postural adjustments (APAs) in the gastrocnemii muscles while standing, when they pull on a handle [59]. APAs related to gait initiation occur in children as young as 2.5 years of age and steadily increase in amplitude until eight years of age. Children from 2.5-8 years of age move the COP posteriorly before taking a step; the APA associated with forward stepping. However, it is not until the age of six that children effectively shift the COP posterior-laterally towards the standing foot. That is the medio-lateral control of stepping takes longer to develop than antero-posterior control.

Adults tend to anticipate the speed of gait by varying the posterior displacement of the COP. A greater posterior shift of the COP indicates the initiation of a faster gait speed. This feedforward control is not seen in children until the age of six years [60].

2.2.4 Development of the sensory control of balance

Children require a rich environment to move and to attempt tasks. Typically, they try, fail and repeat until successful. To achieve dynamic balance, a child must fall hundreds or thousands of times to experience, or play with, the border of stability. In a large observational study of 151 expert crawler and novice walkers, infants transitioned to walking when the fall rate was equivalent to that in crawling, but where the distance covered when walking greatly exceeded that in crawling [61]. The intensity of motor learning and practise in early walking is astonishing, with novel walkers averaging 2368 steps and 17 falls per hour of waking [61], as they explore the frontiers of stability.

2.3 Development of postural control and balance in children with cerebral palsy

The innate patterns of postural control coupled with rich motor sensory learning enable children to develop mature and responsive control of posture and balance. Children with cerebral palsy (CP) have additional challenges to developing balance. They not only have primary physical impairments, but also experience delayed or reduced exposure to sensori-motor learning experiences.

2.3.1 Factors affecting postural control in cerebral palsy

Spasticity, weakness and reduced selectivity of movement are the primary motor impairments affecting the development of balance in children with CP. Over time, further musculoskeletal changes result from the effect of gravity combined with these impairments. Muscle contracture, torsional bone deformities and hip dysplasia are common barriers to postural alignment and efficient movement in children with CP [62]. Co-contraction is commonly found in the muscles spanning two joints, which are known to be important in the development of postural stability [40]. There is a paucity of evidence to determine the effect of common procedures such as gastrocnemius and hamstring lengthening surgery on postural sway and measures of balance [63].

Postural malalignment can make standing postures less efficient and demand more energy to remain in static equilibrium, compared with typically developing (TD) children. Postural malalignment typically occurs at multiple body segments in children with CP. Children with unilateral CP typically stand with more weight on the less impaired side, while children with bilateral CP may present with crouched posture. One study found that of 15 children with independent standing balance, both those with unilateral and bilateral CP stood with a more flexed posture than their available range of movement [64]. This might suggest that alignment in standing is not only due to available ranges of movement but could be linked with difficulties sustaining postural activity or even with an energy efficiency strategy, although there is a lack of evidence in the literature to explain this. Foot posture measurement in quiet standing shows that both children with bilateral and unilateral CP experience abnormal pressure in the midfoot and forefoot, due to either knee flexion or spasticity in the calf muscles [65]. Trunk and pelvic malalignment negatively affect clinical outcomes of balance in children with CP [66]. A study using topographic imaging to measure posture in quiet

standing compared 58 ambulant children with bilateral CP with TD peers. They found three patterns of postural alignment: firstly, a lordotic posture, which tended to produce a forward lean; secondly a backward leaning posture; and thirdly a more typical balanced posture [67]. Variations from normal postural alignment go on to affect the child's dynamic balance responses.

2.3.2 Postural sway in children with cerebral palsy

Postural sway was found to be significantly different in one third of ambulant children with spastic diplegia aged 5-18 years compared to TD children [68]. Where it differs, children with CP show a lower frequency of postural sway and larger radial displacement of the COM compared to TD children [63, 68]. Furthermore, postural sway in children with CP is notably more regular, with less twisting or turning excursions of the COP when compared to TD children [68]. It has been postulated that the rotational synergies contribute significantly to postural stability in quiet standing [69]. Therefore, the apparent reduction in the movement repertoire during postural sway, in children with CP, may reduce stability in standing.

2.3.3 Postural reactions following an external perturbation in children with cerebral palsy

Ambulant children with CP often struggle with balance in functional situations such as riding on a bus. This may be related to the crouched posture of many children with CP. The crouched posture results in delayed responses at the ankles and increased muscle co-contraction in response to external perturbations [70]. Ambulant children with CP lose balance more quickly than TD children. They take a recovery step at a lower perturbation velocity [71] and produce less efficient movements of the COP to recover their balance [72]. This is thought to be due to the presence of co-contraction and insufficient ability to activate agonist postural muscles [73]. However, where TD children assumed crouched postures, their response to platform perturbations were similar to those of children with CP [71]. This suggests that the co-contraction needed to sustain a crouched standing posture, influences the pattern of the initial postural reaction available.

2.3.4 Anticipatory postural adjustments during gait initiation in children with cerebral palsy

Gait initiation usually involves a small posterior and then lateral movement of the COP prior to taking a step. Gait initiation in children with CP is notably different

from TD children and differs in relation to distribution of the impairment in children with CP. Children with unilateral CP tend to have reduced lateral movement of the COM and children with bilateral CP have an increased downward shifting of the body prior to taking a step [74]. In children with unilateral CP, the COM moves forward less efficiently when the affected leg is trailing. This shows that when a step is taken, the trailing leg has a significant part to play in the propulsion of the COM [75]. It has been suggested that children with CP might harness the inherent instability between stance and swing phases as a strategy for forward propulsion during gait, with a potential side effect of loss of postural control [70]. This can be seen in children with CP who find it easier to keep moving than to stop. Here, children are using the relatively heavy head, arms and trunk (HAT) segment to aid forward propulsion. Reduced postural stabilisation and inaccurate foot placement when walking can lead to reduced function and an increase in falls [70].

In conclusion, many children with CP have reduced opportunities for sensorimotor learning early in life and delay in acquiring motor milestones. This, in addition to the underlying physical impairments, affects the development of normal posture and balance. Therapeutic interventions aimed at improving balance need to be evaluated either by taking measures of balance function or by measuring aspects of the balance mechanism.

2.4 Measuring the balance of children with cerebral palsy

2.4.1 Clinical measures of balance

Current outcome measures used to evaluate clinical interventions and trials, such as the Gross Motor Function Measure (GMFM) [76] and Pediatric Balance Scale (PBS) [77] are known to have a ceiling effect and may not detect changes in the highest functioning children. The Balance Evaluation Systems Test (BESTest) has been used to measure balance of children with CP [78]. It includes a comprehensive range of balance activities, which require the participant to be able to follow a series of very complex instructions that may prove difficult for some children. The Star Excursion Balance Test (SEBT) tests high level balance in single leg stance, however it does not distinguish problems of balance from deficits in strength and motor planning [79]. The 10-metre fast walk test and timed 6-minute walk are useful tools when measuring aspects of functional walking [80], however they do not capture how the child moves. They

do not take account of any compensatory strategies, which could have a negative impact on movement in the long term.

Children with CP frequently demonstrate reduced reactive postural control when stepping, often stepping more quickly and with reduced postural stabilisation and inaccurate foot placement (see section 2.3) [72]. Therapists have identified the need to develop tools to measure movement that distinguish compensatory or adaptive strategies [81, 82]. The Quality Function Measure (QFM) was developed in response to this [20]. It tests the quality of motor performance using the 37 items from the Gross Motor Function Measure-66 and is suitable for children with CP GMFCS I-III. The QFM has been shown to have excellent inter and intra-rater reliability [83]. It uses video analysis of these items and scores co-ordination, alignment, dissociated movement, stability and weight shift. However, it is time consuming to test and analyse. The GMFM, PBS, BESTest and QFM all use ordinal rating scales and thus may not be able to quantify subtle changes in balance.

2.4.2 New developments in measuring dynamic balance

There is an emerging body of work using accelerometers to measure stability of the trunk during balance tasks, but as yet these methods of measurement have not been validated against gold standard measures of balance using the COM [84]. The novel Next Step test was created and developed by Prof Marsden and the author and is presented in the following section. It has been designed to address the need for a quantifiable measure of dynamic balance that is simple enough for children with CP to carry out. It measures subtle changes in the movement of the COM during a constrained stepping task [30]. This has the potential to give insight as to the interplay between spasticity, weakness and contracture, which affect postural stability during stepping. It may be able to direct and evaluate therapy interventions more specifically [70].

The test of coordinated stepping has been developed in adults with neurological conditions [30] in order to explore balance and factors affecting balance. It uses 3D motion analysis equipment in a gait laboratory, which is the gold standard way of measuring kinetic and kinematics during stepping. However, this is very time consuming, and expensive and is not easily portable for use in general clinical practice. A portable and inexpensive test of coordinated stepping would be valuable in evaluating clinical interventions, such as orthotic prescription.

The Next Step test uses a portable, scaled down version of the gait laboratory equipment, including the force plate, a small number of markers and one camera, thereby maintaining the accuracy and precision of the full gait laboratory in a smaller stepping task.

In the following sections (2.5 and 2.6) two papers are presented. Paper 1 has been published in Gait and Posture (impact factor 2.7) [85]. It presents the development of a novel outcome measure of dynamic balance in children with CP (Next Step test), in which the APAs prior to taking a step are measured to see how this aspect of dynamic balance differs between children with CP and typically developing children (TD). Paper 2 is presented in 2.6; this paper is in submission to Gait and Posture, and reports on the reliability of this measure. The patient information sheet for these two studies can be found in Appendix 1.

2.5 Paper 1: A cross sectional study investigating dynamic balance when stepping to targets in children with cerebral palsy compared to typically developing children

Authors: Rachel Rapson [1,3], Jos M. Latour [2], Bernie Carter[4], Vasiliki Pitsouni [1], Jon Marsden [1]

[1] School of Health Professions, Faculty of Health, University of Plymouth PL6 8BH [2] School of Nursing and Midwifery, Faculty of Health, University of Plymouth [3] University Plymouth Hospitals NHS Trust Derriford Road [4] Edge Hill University

Keywords: Cerebral Palsy, Balance, Anticipatory Postural Adjustment, Stepping

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2.5.1 Abstract

Background

Children with Cerebral Palsy (CP) have altered anticipatory postural adjustments (APAs) during gait initiation. These APAs may affect dynamic balance in tasks such as stepping.

Research Questions

How are APAs in children with CP affected during stepping to precise targets? How do children with CP modulate APAs when stepping to medial and lateral targets? What is the association between APAs and symptom severity, movement quality and impairment profile?

Method

Children undertook a stepping task to laterally and medially placed targets with either leg, in a randomised order. Movement of the centre of pressure (COP) and markers at the pelvis and foot were measured via a force plate and 3D motion analysis. Motion of the centre of mass (COM) was estimated via pelvic markers. APAs were assessed prior to leading leg lift-off in medio-lateral and antero-posterior directions. Stepping error was calculated. Baseline characteristics of children with CP included Gross Motor Function Measure (GMFM), Quality Function Measure (QFM), leg muscle hypertonia (Tardieu test) and strength (manual dynamometry).

Results

Sixteen ambulant children with CP (12.2 years ± 2.2) and 14 typically developing (TD) children (11.6 years ± 2.9) were assessed. In children with CP, APAs in the medio-lateral direction were 20-30% smaller. Children with CP were less able to modulate their APAs with steps to medial and laterally placed targets, than TD children. Medio-lateral COP motion was associated with movement quality assessed by QFM subsections, GMFM (correlation coefficient r =0.66-0.80) and hip abductor strength (r=0.75). Antero-posterior APAs were significantly smaller when stepping with the non-paretic leg in children with CP. APA size was positively related to the length of the contralateral, paretic gastrocnemius (r=0.77). Stepping error was higher in children with CP and inversely correlated to the size of the medio-lateral APA.

Discussion

Children with CP show smaller medio-lateral APAs especially when stepping to medially placed targets. APA size may be limited by proximal muscle strength and gastrocnemius length.

Key words: Cerebral palsy, Balance, Anticipatory postural adjustment, Stepping

Highlights

- Children with cerebral palsy have significantly greater stepping error
- Children with cerebral palsy show smaller medio-lateral APAs
- Children with cerebral palsy show greater difficulty stepping to a medial target
- Proximal muscle weakness and gastrocnemius length may limit medio-lateral APAs
- Contralateral gastrocnemius range affects antero-posterior motion when stepping

2.5.2 Introduction

Cerebral Palsy (CP) is associated with deficits in posture and balance that affect children's ability to perform the functional activities [86]. Dynamic balance is essential for tasks such as stepping over a toy or manoeuvring in a confined space, such as a shower cubicle. Dynamic balance requires the controlled movement of the centre of mass (COM) within the base of support [87]. It involves a preparatory phase, where anticipatory postural adjustments (APAs) prepare the body for the execution phase when the leg is brought forward to its intended target [40, 42]. Dynamic balance while taking a single step has been modelled as a 'throw and catch' of the COM [88, 89]. This consists of APAs that accurately 'throws' the COM on a ballistic trajectory initially towards the trailing leg and then forwards towards the leading leg [42]. The motion of the body and the stepping leg are coordinated with the COM motion varying with stance width and the position of the stepped foot [30]. Therefore, dynamic balance consists of accurate control and modulation of APAs to control the subsequent trajectory of the body during single leg stance and the positioning of the limb receiving the COM.

Previous work on gait initiation reveals notable differences in APAs related to the severity and laterality of CP [74, 75]. Children with unilateral CP tend to have reduced medio-lateral movement of the COM and children with bilateral CP have an increased

downward shifting of the body prior to initiating gait. In children with unilateral CP, the COM moves forward less efficiently when the affected leg is trailing [75]. However, while this work explores gait initiation, it is unclear how these altered APAs might affect dynamic balance while taking a single step to a precise target. We do not yet know how children with CP modulate APAs according to the target step's location. The motor impairments affecting balance in children with CP include spasticity, contracture, weakness and reduced selective movement [40, 62].It is unknown how APAs associated with stepping to a target vary according to the severity of these impairments.

2.5.3 Aims and Objectives

This study aimed to compare dynamic balance, while stepping to medially and laterally placed targets, in children with CP and TD. The objectives were to assess the APAs by measuring the COP, COM estimate (COM_{est}), and stepping accuracy in a group of children with CP and TD. We tested the hypothesis that APAs will be smaller in children with CP, will be associated with reduced stepping accuracy and show a significant relationship with participants' impairment profile.

2.5.4 Method

A cross-sectional design assessed children on a standardised stepping task.

2.5.5 Participants

Paediatric physiotherapists working in five child development centres in the South West of England recruited children with CP. Children (aged 8-18 years) were eligible if they had a diagnosis of CP, Gross Motor Function Classification System (GMFCS) levels I-III [4], and were able to stand and take up to five steps in bare feet without holding onto equipment. Children were ineligible if they had additional diagnoses affecting balance, were unable to follow complex instructions or were unable to see the illuminated markers on the floor. Using local adverts, we recruited TD children who had no conditions that affected balance or movement.

2.5.6 Sample Size

Previous comparisons of movements of sacral markers as motion of COM_{est} have found differences in COM APA vertical motion between children with CP while stepping (unilateral 26.18mm (±9.11), bilateral 18.08mm (±18.09) and TD 34.31mm (±8.63)) [74]. This produced an average effect size of 1.1. To detect a similar effect size (α=0.05, power=90%) would require a sample size of 30 participants.

2.5.7 **Procedures**

Participant demographics and impairment profile were assessed. Contractures and spasticity of hamstrings (popliteal angle), rectus femoris (Duncan Ely) and gastrocnemius (dorsiflexion with knee extended) were measured using goniometry and the Modified Tardieu Scale [90]. Isometric strength of hip and knee extensors and hip abductors were assessed using a hand-held dynamometer (Lafayette, USA) in standardised positions. The product of the recorded Force x Distance from the point of force application to joint axis provided a measure of the Joint Moment [14, 15]. Gross Motor Function Measure (GMFM) [91] and Quality Functional Measure (QFM) [20] were also determined for children with CP.

Children wore shorts, top, and were barefoot. Children with a leg length shorter than 87cm (measured from the anterior superior iliac spine (ASIS) of the pelvis to the medial malleolus) used stepping targets set at 26cm and children with leg length longer than 87cm used targets set at 35cm. Both target distances were deemed comfortable in pilot studies with TD children. Children stood on a black cardboard platform that housed four targets, which overlaid an embedded portable force plate (Kistler™ 9286BA UK), Figure 3A. The targets consisted of a 2cm diameter circle made of electroluminescent paper. The children stood with their 1st metatarsal-phalangeal joint (MTP) joints 4cm apart. The targets were directly in front (medial target) or 25° to the side (lateral target) for each foot (Figure 1A). Both medial targets overlaid the force plate. Participants had four CODAmotion™ (Leicestershire, UK) electronic markers on each foot, placed at the head of talus, 1st and 5th MTP and on nail of the great toe. A cluster of three markers were placed on a Velcro belt, positioned on a line horizontal with the ASIS's, with the central marker aligned with the midline of the child's body. They were able to hold onto a walking frame/ parallel bars, as needed, when determining the starting position and at the end of a trial when repositioning the foot.

To allow a consistent measure of stepping accuracy, the child was asked to practice stepping their leading foot as accurately as possible onto each target, without rushing, and bring their second foot alongside. This 'best step' target step position was then marked by chalking around the child's leading foot and the position of the leading foot recorded using the 3D motion analysis system (see below) to allow for a calculation of absolute stepping error relative to this position.

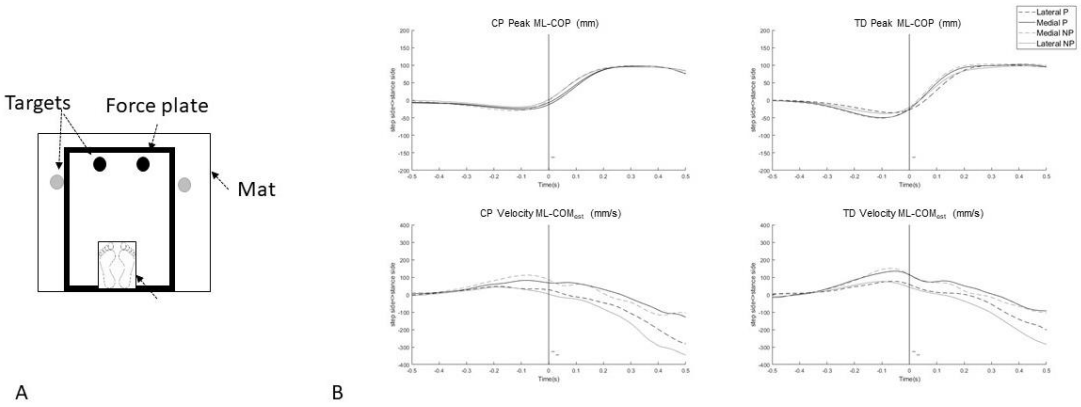

Figure 3 A *Experimental set up showing target location relative to force plate.* ***B*** *Grand average response of medio-lateral centre of pressure (ML-COP (top)) and the centre of mass estimate (COM_{est} (bottom)) for children with cerebral palsy (CP (left)) and typical development (TD (right)).*

The onset of a stepping trial was indicated by a tone whose pitch (high or low) indicated which leg to step with whilst the target simultaneously illuminated. The child was asked to undertake up to four sets of 15 steps with breaks in between sets. This allowed a maximum of 15 trials to each target to be recorded.

Movement data were sampled at 200Hz using the CODAmotion™ minihub and stored using CODAmotion™ software (Leicestershire, UK). Force plate data were sampled at 200 Hz via a power 1401 analogue-digital converter (CED Cambridge UK) and stored using Signal software (CED Cambridge UK). The motion analysis and force plate data were synchronised via transistor-transistor logic pulses generated by the onset of CODAmotion data analysis.

A 0.5-1.5 second random delay occurred between the onset of recording and the onset of the target light/auditory tone. This allowed the collection of baseline data whilst the random delay prevented the child from anticipating the onset of the target.

2.5.8 Data Reduction

Data were exported as text files for secondary analysis in MATLAB™ (Mathworks USA). Target indicator and force plate data were combined with CODAmotion data and grouped according to the target. Data were omitted from all analyses where there was marker drop out. The onset and offset of stepping was automatically determined from the vertical acceleration of the centroid of the foot markers [92] based on data rising above and below the mean baseline level for at least 125ms. If there was an acceleration deviation in a 200ms period after foot offset, this indicated a shuffle in the leading foot

and data were rejected. This accounted for ~5% of records and was similar in children with CP and TD children.

Four APAs were calculated in the 500ms period prior to the onset of stepping with the leading leg: peak medio-lateral (ML), antero-posterior (AP) motion of the COP, ML and AP velocity of the COM estimate (COM_{est}). The peak ML-COP and the peak AP-COP motion in the direction of the leading leg was calculated relative to the baseline period. The midpoint of the ASIS markers was used to calculate the ML and AP velocity of COM_{est} in the direction of the trailing leg relative to the baseline. The fifth outcome was calculated over the 200ms period after foot offset. The accuracy of the end foot position was defined as the endpoint step error to the 'best' foot position target. This was calculated as the absolute distance between the centroid of the foot (defined from the three markers) and the centroid of the target (defined as the centroid of the 'best step' taken at the start of the trial).

2.5.9 Statistical Analysis

Data were arranged as if children were stepping to the left side by inverting the ML related data. Further, the more paretic leg (termed paretic) was compared to stepping with the less paretic leg (termed non-paretic). For TD children the weaker leg was grouped with the paretic leg and the stronger leg with the non-paretic side. Leg strength was determined using the mean of the isometric strength measures. For children with CP and hemiplegia the paretic leg corresponded to the clinical diagnosis with the non-involved side termed the non-paretic leg. Two (out of 6) children with diplegia had a weaker left leg. Seven out of 14 TD children had a weaker left side

Data were assessed for normality using a Shapiro-Wilks test. Data were normally distributed except for AP-COP, AP-COM_{est} and stepping error measures. Differences in demographics and clinical measures between the two groups were compared using an unpaired t-test. Differences between groups (TD vs CP) for the movement-related variables (ML-COP and COM_{est}) were compared using a between groups repeated measures ANOVA with factors leg (left or right) and target (medial or lateral). For non-normally distributed data, group responses (AP-COP and AP-COM_{est} and stepping error) to steps to each target were compared using a Mann Whitney test.

In children with CP, the differences in the mean ML-COP according to GMFCS level (1 vs 2) were compared using unpaired t-tests. Further, in children with CP, a Pearson Correlation coefficient assessed the correlation between mean ML-COP and GMFM

walk and stand scores, Quality Function Measure sub scores (QFM) and selected measures of the child's level of impairment (Hamstring R1 and R2; Gastrocnemius R1 and R2 and Knee extensor strength and Hip abductor strength). A Bonferroni correction was used for multiple comparisons (n=7 GMFM & QFM measures, n=12 for measures of range of motion and n=6 for measures of strength)[93]. Finally, based on previous work [75] the relationship between AP-COP peak when stepping with either leg and the contralateral gastrocnemius passive range was explored.

Permission for this study was granted by South West Frenchay Research Ethics Committee (ref 18/SW/0239). The study is reported according to STROBE guidelines [94].

2.5.10 Results

Participant characteristics and impairment profile: Thirty children were recruited, 14 children with TD (mean age 11.58 years ± 2.91 SD) and 16 children with CP (mean age 12.15 years ± 2.18 SD). The groups had similar gender distribution, weight and height (Table 1). Of the children with CP, six had bilateral CP, (median GMFCS level II, range 1-2). Ten children had unilateral CP, five children had right hemiplegia (median GMFCS level I, range 1-2) and five children had left hemiplegia (median GMFCS level II, range 1-2) (Table 1).

Table 1 *Participant demographics*

Study Group:	Typically Developing n=14	Cerebral Palsy n=16
Gender Male (Female)	6 (8)	7 (9)
Age (years, mean ± SD)	11.6 (2.9)	12.2 (2.2)
Height (cm, mean ± SD)	152.2 (13.3)	147.9 (13.5)
Weight (kg, mean ± SD)	42.7 (10.0)	43.1 (15.3)
GMFCS level (I:II:III)	-	7:8:1
Distribution of Impairment Bilateral	-	6
Distribution of Impairment Unilateral Right: Left	-	5:5

Key- Number (N), Gross Motor Function Classification System (GMFCS), mean (M), standard deviation (SD)

Table 2 Mean measures of leg impairment in children with typical development and cerebral palsy.

Parameter	Cerebral Palsy n=16		Typically Developing n=14	
	P	NP	P	NP
Hamstrings R2 (°)	50.8 (10.4) *	42.3 (7.0)*	33.0 (10.3)	29.6(10.2)
Hamstrings R1 (°)	61.4(16.0)	52.3 (13.6)	34.8(11.3)	36.7(14.1)
Gastrocnemius R2 (°)	2.6(6.8)*	5.3(9.8)	9(5.3)	10.1(4.8)
Gastrocnemius R1 (°)	-4.3(11.1)	-0.5(12.1)	3.9(6.9)	2.5(7.9)
Rectus Femoris R2 (°)	133.4(16.4)	128.3(18.8)	142.9(6.8)	137.2(27.5)
Rectus Femoris R1 (°)	128.1(31.7)	126.4(24.1)	142.5(7.4)	138.3(29.1)
Knee Flexion Moment (Nm)	33.2(13.1)*	39.2(16.0)	50.4(25.5)	54.5(29.7)
Knee Extension Moment (Nm)	48.2(19.1)*	57.8(27.6)	64.9(16.7)	71.7(19.8)
Hip Abduction Moment (Nm)	41.0(23.0)	43.7(24.8)	56.9(20.6)	59.5(20.4)

Key- Mean and standard deviation (SD) indicated, R1= modified Tardieu scale fast stretch, R2=modified Tardieu scale slow stretch, P=paretic leg, NP=Non-paretic leg, * indicates significant difference between CP and TDC group $p<0.05$, Nm=Newton metres

Table 3 Mean Quality Function Measure and Gross Motor Function Measure scores for children with cerebral palsy

Parameter	All n=16	GMFCS I n=7	GMFCS II n=8	GMFCS III n=1
GMFM stand	84.8 (16.4)	93.5(8.9)	84.0 (2.3)	30.8
GMFM walk	79.4(26.6)	95.9(24.4)	72.9(3.3)	15.3
QFM alignment	62.4(19.8)	77.6(11.9)	55.0(10.3)	15.4
QFM co-ordination	67.2(25.3)	84.5(20.6)	59.6(9.0)	7.2
QFM dissociated movement	57.7(23.5)	75.9(17.8)	47.8(9.8)	8.9
QFM stability	63.2(24.5)	80.7(20.5)	54.8(7.4)	7.8
QFM weight shift	61.0(21.4)	78.0(15.3)	51.8(8.1)	15.2

Key- Mean and standard deviation (SD) indicated, Gross Motor Function Classification (GMFCS), Gross Motor Function Measure (GMFM), Quality Function Measure (QFM)

Children with CP had significantly reduced passive range in both hamstrings (as measured by popliteal angle) on the paretic side. Spasticity as measured by the modified Tardieu test (R1) was greater in the CP group in the paretic and non-paretic hamstrings (Table 2). Strength was lower in the CP group for the knee flexors and extensors on the paretic side but this was not significant after a Bonferroni correction (Table 2). Table 3 shows that QFM and GMFM walk and stand scores reduced with increasing GMFCS classification in children with CP.

APA response characteristics: On average 11.7 ± (2.4) steps to each target were analysed (CP 11.4 ±2.6; TD11.9 ±2.3). The grand average response curves are shown in Figure 3B.

ML-COP peak*:* ML-COP peak velocity prior to foot lift of the leading leg (foot-offset) was larger when stepping towards medial compared to lateral targets (Effect of target $F(1, 28) = 15.4$ $p<0.001$, Table 4). ML-COP peak velocity was smaller in the CP group (Effect of Group $F(1, 28) = 9.1$ $p<0.005$) (Table 4, Figure 4A). There were no other significant interaction effects.

ML-COMest peak velocity: ML- COM_{est} peak velocity prior to foot-offset was larger when stepping towards medial compared to lateral targets (Effect of target $F(1, 28) = 123.4$ $p<0.001$). There was a Target x Group interaction, here the increase in ML-COM_{est} peak velocity when stepping to medial compared to lateral targets was larger for the TD group compared to the CP group (Target x Group interaction $F(1,28)=5.4$ $p<0.05$)) (Table 4, Figure 4A). There was a Side x Target interaction with the increase in ML- COM_{est} peak velocity when stepping to medial as opposed to lateral targets being larger when stepping with the non-paretic compared to the paretic leg (Figure 4A). There was no group effect (Group $F(1, 28) = 3.2$ $p=0.09$) or any other interaction effects.

AP-COP peak velocity: The AP-COP moved posterior prior to foot-offset. The data were not normally distributed and groups were compared at each target location using a Mann Whitney test. AP-COP peak was significantly greater when stepping with the non-paretic leg in the TD group ($p<0.05$) (Table 4).

AP-COMest peak velocity: AP- COM_{est} moved forwards prior to foot-offset. AP-COM_{est} peak velocity was not normally distributed, and groups were compared at each

target location using a Mann Whitney test. There were no significant differences between groups (Table 4).

Stepping error and step length: Step length was not significantly different between groups (Table 4). Stepping error was compared at each target location using a Mann Whitney test. Stepping error was significantly higher in children with CP at all target locations ($p<0.05$, table 4). In the CP group there was a significant relationship between stepping error and the size of the ML-COP peak when stepping with both the paretic or non-paretic legs to either the lateral or medial targets ($r=0.55$-0.68 $p<0.05$). Here higher stepping errors were associated with low ML-COP peak values. There was no significant relationship in the TD group ($r=0.14$-0.47 $p>0.05$).

Table 4 *Anticipatory postural adjustment parameters for children with CP and typically developing children. Mean (standard deviation) indicated.*

	Cerebral Palsy				Typically Developing			
	Lateral P	Medial P	Lateral NP	Medial NP	Lateral P	Medial P	Lateral NP	Medial NP
Peak ML-COP (mm)	-35.1 (6.1)	-38.4 (6.0)	-32.0 (5.0)	-43.7 (6.3)	-48.9 (4.1)	-58.4 (5.1)	-52.3 (3.7)	-67.1 (5.3)
Peak AP-COP (mm)	-8.1 (2.1)	-9.2 (2.7)	-7.1 (1.2)	-8.9 (1.8)	-9.52 (1.5)	-12.8 (1.4)	-10.7 (1.3)	-12.2 (1.9)
Peak ML-COM velocity (mm/s)	83.1 (13.1)	121.5 (11.3)	77.5 (10.2)	146.4 (11.8)	89.8 (10.5)	168.8 (13.7)	88.4 (6.9)	173.5 (10.3)
Peak AP-COM velocity (mm/s)	-68.0 (10.6)	-107.1 (14.8)	-63.9 (10.2)	-102.3 (12.2)	-54.7 (11.7)	-95.0 (16.8)	-65.3 (11.5)	-93.1 (11.4)
Stepping error (mm)	30.41 (38.1)	27.7 (37.5)	25.5 (18.9)	23.3 (11.3)	15.3 (9.5)	14.9 (5.7)	15.7 (9.4)	15.0 (7.4)
Step length (cm)	29.3 (4.8)	18.4 (5.2)	23.7 (8.1)	21.9 (8.3)	28.3 (13.9)	18.8 (9.0)	21.0 (11.1)	22.5 (11.5)

Key- Medio-lateral motion of the centre of pressure (ML-COP), Antero-posterior motion of the centre of pressure (AP-COP), P=paretic leg, NP=Non-paretic leg

Effects of condition severity and impairment profile on ML anticipatory postural adjustment size: The mean ML-COP peak averaged across all targets was higher in children GMFCS 1 (n=7; 46.6 mm +/- 15.1) compared to those GMFCS 2 (n=8; 30.1 +/-22.1) but this was not significantly different ($t=-1.7$ $p>0.05$). The participant with GMFCS 3 had a higher level of impairment and balance difficulties. Removal of this participant did not affect the significance of the balance and impairment-based results described above.

The mean ML-COP peak was significantly correlated with the GMFM stand ($r=0.71$, $p<0.002$) and walk scores ($r=0.72$ $p<0.002$), and the QFM subsections alignment ($r=0.66$, $p<0.006$), coordination ($r=0.80$ $p<0.001$), stability ($r=0.80$ $p<0.001$) and weight shift ($r=0.72$ $p<0.005$). In all cases, a lower mean COP peak was associated with a lower score on the QFM. These correlations remained significant after removal of the participant GMFCS III expect for alignment and stability categories.

The mean COP peak was significantly correlated with mean knee extensor strength ($r=0.70$ $p<0.005$) and mean hip abductor strength ($r=0.75$ $p<0.001$) with lower strength being associated with smaller ML-COP peak (Figure 4B). Mean gastrocnemius passive range was also associated with the mean ML-COP peak ($r=0.63$ $p<0.01$) with a lower passive range being associated with smaller ML-COP peak.

In children with CP the AP-COP peak when stepping with the non-paretic leg was associated with the paretic leg gastrocnemius passive range ($r=0.77$ $p<0.01$) (Figure 4 **A** Difference in medio-lateral centre of mass estimate (ML-COMest) velocity when stepping with the paretic (or weaker for TD) and non-paretic sides to lateral and medial targets. **B** Relationship between medio-lateral centre of pressure (ML-COP) peak and mean hip abductor strength. A negative value of ML-COP indicates a greater lateral motion. **C** Relationship between anterior-posterior centre of pressure (AP-COP) and gastrocnemius length. A more negative COP indicates greater posterior motion.). In contrast, there was no significant relationship between the AP-COP peak when stepping with the paretic leg and the non-paretic leg gastrocnemius passive range ($r=0.21$ $p>0.05$). All significant correlations remained after removal of the one participant with GMFCS III.

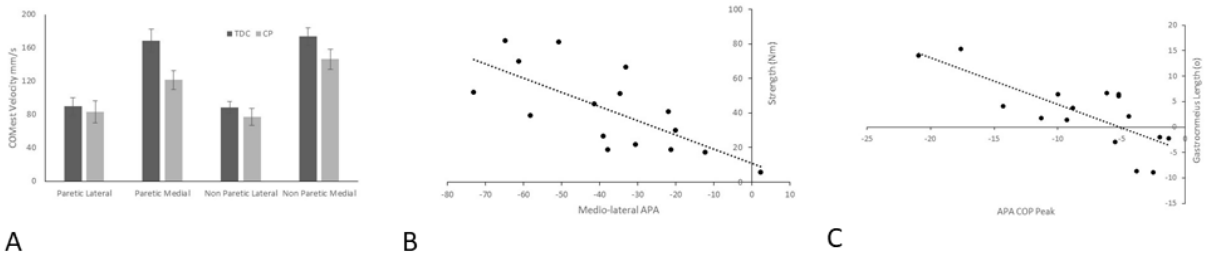

Figure 4 A *Difference in medio-lateral centre of mass estimate (ML-COM$_{est}$) velocity when stepping with the paretic (or weaker for TD) and non-paretic sides to lateral and medial targets.* ***B*** *Relationship between medio-lateral centre of pressure (ML-COP) peak and mean hip abductor strength. A negative value of ML-COP indicates a greater lateral motion.* ***C*** *Relationship between anterior-posterior centre of pressure (AP-COP) and gastrocnemius length. A more negative COP indicates greater posterior motion.*

2.5.11 **Discussion**

This study compared dynamic balance while stepping to medially and laterally placed targets, in children with CP and TD. The objectives were to (a) assess the APAs by measuring the COP, COM estimate (COM$_{est}$), and stepping accuracy in a group of children with CP and TD (b) investigate the relationship between APA and the type and severity of impairment. The study found that children with CP had reduced APAs when stepping and higher stepping inaccuracy. APA modulation with target position was reduced in children with CP and lower APAs were associated with greater stepping error. APA size correlated with functional ability, measures of impairment and stepping error.

Children with CP showed reduced medio-lateral (ML) APAs, similar to that previously described in children with CP [74] and in adults with hemiplegia post stroke [95, 96]. Balance during stepping can be described using the 'throw and catch' model, where the COM is 'thrown' towards the standing leg in a ballistic trajectory towards the target step [30]. Findings from this study agree with previous work showing that the preparatory motion prior to swing leg lift off, varies depending on the position of the intended step target [31]. Stepping laterally can be thought of as a controlled fall towards the target. APAs when stepping laterally were not markedly different between groups and this reduction in the need to accurately programme APAs may be a reason why children with CP often walk with a wide base of support. Stepping medially requires a greater ML and AP movement of the COM$_{est}$. The hip abductors of the leading leg work synergistically with the hip adductors of the stance leg to control the pelvis through this movement [97]. In children with CP, greater knee extensor and hip

abductor strength was associated with greater ML motion of COP suggesting weakness in these muscles may contribute to smaller ML APAs.

The significant association between the APA size (ML-COP) and the stepping error in children with CP suggests that either poorly scaled APAs directly affect the trajectory of the leg movement and/or that alterations in leg motion are required to maintain balance in the face of an inaccurate APA. However, the error in foot placement was still higher in the CP group when stepping to lateral targets despite a non-significant difference in APA size between groups. This suggests that stepping error may be only partly explained by deficits in APA size and that inaccurate programming of leg movements could also occur.

The AP-COP peak when stepping with the non-paretic leg was associated with reduced range in the contralateral paretic gastrocnemius whilst the opposite relationship (that is, paretic leg AP-COP peak and non-paretic leg gastrocnemius length) was not seen. This suggests a limitation in AP motion of the COM; this could be in part due to reduced range in the contralateral gastrocnemius [75].

This study recruited a small number of participants, thereby limiting the ability to analyse sub-groups. The static standing posture prior to taking a step and the asymmetry in standing posture and load taken by each leg were not measured, which may affect the pattern of dynamic balance [96]. Children with CP took shorter steps during the establishment and definition of their individual target step. Although the difference was not significant between the groups, it is a limitation of the study as it may require less dynamic balance to step a shorter distance [31].

2.5.12 Conclusion

Children with CP show smaller ML APAs and this is more marked when they step to more medially placed targets. Medio-lateral APA size may be limited by proximal muscle strength and gastrocnemius length. Limited gastrocnemius range affects AP motion when stepping with the contralateral leg. Targeting such impairments in combination with task related training might help to improve dynamic balance.

2.5.13 Acknowledgements

Thank you to all the children and families who took part in this research and to the NIHR for funding this work through the Clinical Doctoral Fellowship scheme (ICA-CDRF-2017-03-04).

2.6 Paper 2: The novel Next Step test is a reliable measure of anticipatory postural adjustments made by children with cerebral palsy prior to taking a step

Authors Rachel Rapson [1,3], Jos M. Latour [2], Bernie Carter[4], Vasiliki Pitsouni [1], Jon Marsden

[1]School of Health Professions, Faculty of Health, University of Plymouth PL6 8BH [2] School of Nursing and Midwifery, Faculty of Health, University of Plymouth [3] Torbay and South Devon NHS Trust, Torquay, TQ2 7BA [4] Edge Hill University

Key words: Dynamic balance, Measurement, Cerebral palsy, Reliability, Anticipatory postural adjustments, Next Step test.

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2.6.1 Abstract

Background

Children with cerebral palsy (CP) make smaller medio-lateral anticipatory postural adjustments (APAs) than typically developing peers when stepping forward to a medial target. They are also less accurate at reaching the stepping target.

The Next Step test involves the biomechanical measurement of APAs and foot placement error. These may be useful outcome measures to evaluate dynamic balance in a clinical trial. The reliability of the measures must be assessed to establish their reliability as research tools.

Research question

What is the inter-rater and intra-rater reliability of stepping accuracy and measures of APAs made by children prior to taking a step?

Methods

Typically developing (TD n=14) or children with CP (n=16) were recruited from local clinics. Children stepped to electro-luminescent targets placed medially and laterally to each foot. Stepping responses were measured using a force plate and 3D motion analysis of markers placed on the feet and pelvis. The APA was defined as the movement of the centre of pressure (COP) and the centre of mass (COM) estimated via pelvic markers, prior to lifting the lead leg. Stepping accuracy was defined as the absolute distance between the target and end foot position. Participants undertook two data collection sessions separated by at least one week. In session one, the test was measured by rater 1 who repeated this in session two, along with another data collection by rater 2 or rater 3, after a rest period. Where data were normally distributed, they were assessed for inter-rater and intra-rater reliability using an intra-class correlation coefficient (ICC) and Bland-Altman plots. The standard error of measurement was calculated to determine the minimum difference needed to detect true change.

Results

There was no between group differences in-group characteristics (age, weight, height) or in stepping velocity. We found good to excellent reliability when measuring the amplitude and velocity of medio-lateral APAs (ICC range 0.73-0.89). The reliability of antero-posterior APAs was more variable (ICC range 0.08-0.92). The minimum difference to detect a true change for peak medio-lateral motion of COP ranges from

23.7mm to 29.6 mm and for peak velocity of medio-lateral COM estimate 41mm to 61.9 mm. Stepping accuracy was not normally distributed.

Significance

The Next Step test is a reliable measure of dynamic balance. The peak medio-lateral motion of the COP and medio-lateral velocity of the COM estimate are reliable when measured during a constrained stepping task in ambulant children with cerebral palsy.

2.6.2 Introduction

Cerebral palsy (CP) is a childhood disability affecting movement and balance. The motor symptoms of CP include spasticity, co-contraction, muscle weakness and reduced selective movement control, resulting in reduced postural stabilisation and inaccurate foot placement when walking and falling [70]. People with CP fall due to a number of causes. Often falls occur during activities that are voluntarily generated such as stepping in a confined space and turning, as well as in response to an unexpected perturbation [70, 98].

Several tests of balance and motor performance are used in clinical and research practice. The Pediatric Balance Scale (PBS), the Timed Up and Go and Gross Motor Function Measure (GMFM) measure motor function and balance during a range of everyday mobility tasks [77, 99, 100]. Some have ceiling effects which make it difficult to distinguish differences in the balance of more mobile children [101] and none of them are designed to measure the quality of movement while carrying out the tasks. The Quality Function Measure (Quality FM) uses subjective rating to score the performance of the GMFM; however, it is quite time consuming [20]. Therefore, there is a need for a reliable test of balance that can quantify dynamic balance in children with CP.

Previous work shows differences in the way that children with CP make anticipatory postural adjustments (APAs) when initiating walking [74]. Taking a single step to a target is a constrained task that has been frequently assessed in healthy adults and those with neurological conditions [102]. Studies highlight variation in the anticipatory movement of the centre of mass (COM) depending on the medio-lateral location of the forthcoming step [103]. The co-ordinated step of the leg and the motion of the body has been likened to a 'throw and catch' sequence, where the COM of the body is accurately thrown from the stepping leg on a trajectory towards the stance leg and then caught again by the stepping leg, once the foot has been positioned [30]. The initial throw of

the COM is actively controlled with subsequent medio-lateral motion explained by modelling the body as an inverted pendulum pivoting about the ankle.

Inaccuracies in either the positioning of the foot or the throw of the body or the co-ordination of the two result in instability [103]. Biomechanical measurement of APAs reported in the literature include both the displacement and the velocity of COM and Centre of Pressure (COP) [104].

Previous work using the Next Step test (Figure 4A and Figure 5) has established concurrent validity and convergent validity. Children with CP make smaller medio-lateral APAs than typically developing (TD) peers when stepping forward to a medial target (Figure 4B)[85]. However, they do modulate APAs depending on the location of the step in the same way that adults do [74, 103]. Children with CP are less accurate at reaching the stepping target than TD peers [85]. Further, the size of APAs significantly correlate with (a) measures of impairment such as muscle strength and spasticity (b) measures of movement quality assessed using the Quality FM score[85].

The biomechanical measurement of APAs and foot placement error may provide useful clinical outcome measures to evaluate balance that anticipates volitional movement. However, the reliability of the measures over time and between raters must be assessed to establish their reliability as research tools.

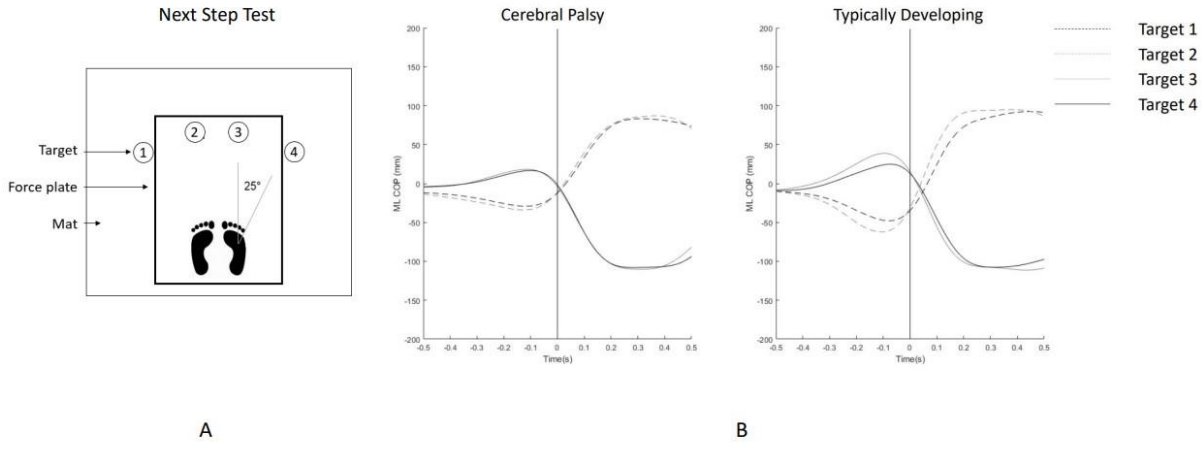
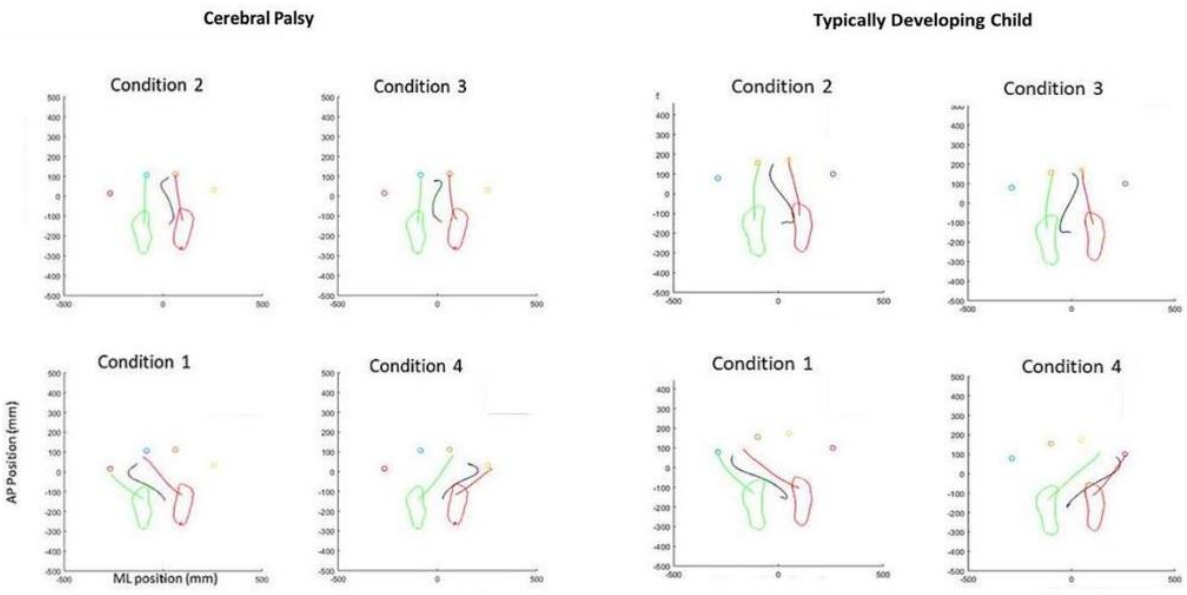


Figure 5 ***A*** *shows the experimental stepping mat overlying the force plate, with four electroluminescent targets set medially and laterally.* ***B*** *shows the differences in pre-step anticipatory postural adjustments between children with cerebral palsy and typically developing children.*



Key- Foot outline and trace of step to target (right=red and left=green), movement of the centre of mass estimate (black line), Antero-posterior (AP), medio-lateral, (ML), millimetres (mm)

Figure 6 *Shows a representative sample picture of a child with cerebral palsy and typical development. There is a larger pre-step movement of the centre of mass estimate to a medial target (conditions 2 and 3) than a lateral target (condition 3 and 4) and small pre-step movement in children with cerebral palsy (left) and typically developing children (right).*

2.6.3 Aims and Objectives

This study aims to measure inter and intra-rater reliability of stepping accuracy, peak medio-lateral (ML) and antero-posterior (AP) motion of the COP, COM estimate (COM_{est}) and the velocity of the COM_{est}. Approval for this study was granted by South-West Frenchay Research Ethics Committee (ref 18/SW/0239).

2.6.4 Methods

Children were recruited, by community paediatric physiotherapists, from five centres in the South-West of England. Children were eligible if they were aged 8-18 years with a diagnosis of CP. Children needed to be able to stand and take five steps independently and had Gross Motor Function Classification System (GMFCS) levels I-III [4]. Typically developing (TD) children were recruited via adverts and had no conditions affecting balance.

2.6.5 Sample Size

An intra-class correlation coefficient (ICC) of 0.7 is deemed to be suitable for outcome measures used in clinical trials [105]. With 14 children in each group, it would be possible to detect a correlation of 0.7 (power =0.85).

2.6.6 Study Procedures

Participants were asked to wear shorts and top and have bare feet. Their height, weight, leg length and pelvic depth were measured. Children with a leg length shorter than 87 cm from the anterior superior iliac spine (ASIS) of the pelvis to the medial malleolus used targets set at 26 cm distance, and children with greater leg length had targets set at 35 cm distance. Pilot work had shown that this was a comfortable stepping distance for both TD and CP children.

Figure 4a shows the "Next Step" board on a Kistler™ (9286BA Kistler, Hampshire, UK) force plate (embedded in a level mat). They had four CODAmotion™ (CODA motion Leicester UK) electronic markers on each foot, placed at the head of Talus, first and fifth metatarsal-phalangeal joint and on the nail of the great toe. A cluster of three markers were placed on a Velcro belt, two positioned in a horizontal line with each ASIS, and one aligned with the midline of the child's body. The midpoint of the ASIS markers gave an estimate of the COM (COM_{est}). This method has been used previously and validated against other models in healthy and neurological populations [106, 107].

The participants were asked to stand with both feet together in a position comfortable to them, holding onto a walking frame as needed. This starting position was marked by drawing round the participant's feet with chalk onto the board. Four electro-luminescent targets were set at the appropriate distance. The targets consisted of a 2 cm diameter circle made of electroluminescent paper that light up when a current is applied. The targets were directly in front or 25° to the side for each foot (Figure 1a). Participants had to step to a medial or laterally placed target with the leg on the same side as the target. An auditory tone signalled the start to each trial step and the foot to step with, followed by a light randomly showing on one of the four targets. Randomisation was achieved using the Spike2 (CED, UK) graphical editor software. The child had two practice steps to each target. The child was then asked to do four sets of 15 steps, with breaks in between sets, as needed. The force plate was reset to zero after each set. Movement and force plate data was sampled at 200Hz using the CODAmotion™ hub / analogue-digital converter (Power 1401 CED, UK) and stored using CODAmotion™ software. Data was exported as text files for secondary analysis in MATLAB™ using customised programmes. The participant was asked to step their foot as accurately as possible onto each target in turn, without rushing, and bring their second foot to follow. The target step was then marked by chalking around the child's foot and the end target position of the foot recorded. This provided the "best foot position" and was used to

determine the endpoint error. The leading leg was the first leg to step whilst the second leg was referred to as the trailing leg.

Differences in demographics between groups (age, weight and height) were assessed using an unpaired t test. A between group analysis of variance assessed the effects of GROUP (n=2) and TARGET (n=4 levels) on step velocity, step length and target position.

The intra-rater reliability of the test was explored by measuring the participants in the same location and circumstances by the same rater, one week later. The inter-rater reliability was explored by the addition of a second rater during the second session. The order of the testing in the second session was randomly varied (via Matperm function in MATLAB™) to avoid the effects of fatigue or potential for motor learning. Each test was separated by a 10-minute minimal break.

Where data was normally distributed, reliability was assessed using an intra-class correlation coefficient (ICC) [108] and Bland Altman Plots [109]. A one-way random effects model was used to assess intra-rater reliability (ICC 1, 1). A two-way random effects model (ICC 2,2) was used to assess inter-rater reliability assuming that raters were selected from a larger population [108]. Absolute agreement between raters / measures was chosen as the definition of reliability. Reliability was interpreted as excellent (ICC >0.9), good (ICC 0.75 to 0.9), moderate (ICC 0.5 to 0.7) or poor (ICC <0.5) [110]. Intra-rater reliability was further assessed using Bland Altman plots to determine limits of agreement. These provide a visual display to compare the variability of each pair of measures using the mean and two standard deviations and the detection of systematic error [111].

Standard Error of Measurement (SEM) was calculated to provide an absolute index of reliability and an estimate of the precision of the scores. This enables the determination of the Minimum Difference needed to detect a true change in the outcome measure. As described by Weir [112] SEM was defined as:

$SEM = SD \sqrt{(1- ICC)}$.

Minimum Difference was defined as:

$MD=SEM \times 1.96 \times \sqrt{2}$

The average of up to 15 steps to each target were calculated. The reliability of the following outcome measures was calculated:

Stepping accuracy was defined as the endpoint step error to the 'best' foot position target. This was calculated as the absolute distance between the centroid of the foot and the centroid of the target. The centroid of the foot was defined from 3 of the 4 markers on the foot. The markers chosen were the same for each participants across all sessions but could vary across participants if there was a marker that showed significant drop out/occlusion. End foot position was defined as the average position calculated over the 200ms period immediately after foot offset.

Peak medio-lateral (ML) and antero-posterior (AP) motion of the COP and COM_{est} were calculated prior to onset of stepping with the leading leg. The peak ML COP motion in the direction of the leading leg and the peak AP COP motion in the 500ms period prior to stepping onset was determined. Peak COP and COM_{est} was calculated relative to the baseline period.

Medio-lateral (ML) and antero-posterior (AP) velocity of the COM_{est} were calculated prior to onset of stepping with the leading leg. The peak ML COM_{est} velocity in the direction of the trailing leg and the peak AP COM_{est} velocity in the 500ms period prior to stepping onset was determined. Peak COM_{est} velocity was calculated relative to the baseline period.

2.6.7 Results

Fourteen typically developing (TD) children and sixteen children with cerebral palsy (CP) aged between 8 and 16 years, were recruited to this study. Their demographic details are shown in Table 5. The groups were similar in age, gender, height and weight ($p>0.05$). Of the children with CP, most were GMFCS I and II and only one child was GMFCS III. They had an even distribution of impairment, six children with bilateral CP and five children with right and left hemiplegia. There was no difference between groups in participant height, target distance or step velocity (Effect of Group $P>0.05$). Medial targets were associated with a faster stepping speed (Effect of TARGET $p<0.05$). Children with CP did show shorter steps across all targets (Effect of Group $P>0.05$).

Reliability was tested where data were normally distributed. Data from stepping accuracy were not normally distributed and not tested for reliability. Intra-class correlation coefficients are shown in Table 6 and Table 7.

In children with CP, good intra-rater reliability was measured in peak ML amplitude of the COP (ICC range 0.79-0.84) and peak ML velocity of the COM_{est} (ICC range 0.73-0.89). Good to excellent reliability was found for AP amplitude of COP (ICC range 0.77-0.92) but intra-rater reliability of the peak AP velocity of COM_{est} (ICC range 0.08-0.51) was poor. Both peak ML and AP COM_{est} had poor intra-rater reliability. Intra-rater reliability was more variable for TD children, ranging from poor to good reliability for each measure (Table 7).

Table 5 *Participant Demographics*

Group	TD	CP
n=	14	16
Gender (Male : Female)	6:8	7:9
Age (years) mean (SD)	11.6 (2.9)	12.2 (2.2)
Height (cm) mean (SD)	152.2 (13.3)	147.9 (13.5)
Weight (kg) mean (SD)	42.7 (10.0)	43.1 (15.3)
Average step length (mm) mean (SD)	311 (69)	250 (69)
Average leg speed (mm/s) mean (SD)	361 (117)	392 (146)
GMFCS level (I:II:III)	-	7:8:1
Distribution of Impairment Bilateral	-	6
Distribution of Impairment Unilateral Right : Left	-	5:5

Typically developing children (TD) and children with cerebral palsy (CP) Number (N) Standard Deviation (SD) Gross Motor Function Classification System (GMFCS)

Inter-rater reliability was good to excellent in children with CP for peak ML COP amplitude (ICC range 0.78-0.93), moderate to excellent for peak AP COP amplitude (ICC range 0.57-0.96), good to excellent for peak ML velocity of COM_{est} (ICC range 0.83-0.92). However, peak AP velocity of COM_{est} had only moderate inter-rater reliability (ICC range 0.56-0.67). In TD children, inter-rater reliability was lower than in children with CP and more variable.

Table 6 *Intra-rater reliability of the Next Step test of dynamic balance during a constrained stepping task in children with cerebral palsy and typical development.*

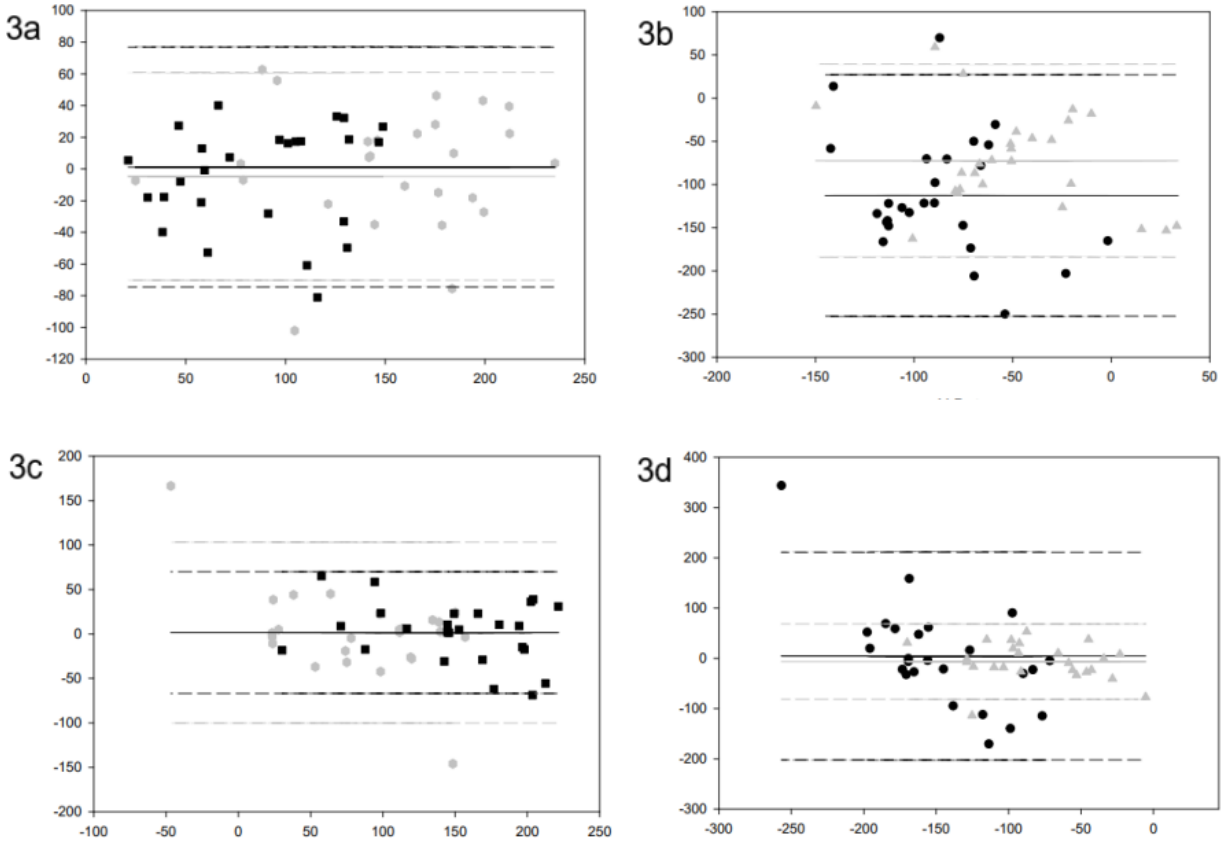
Intra-rater reliability	Target	CP n=16 ICC 95% CI limits	F total	TD n=9 ICC 95% CI limits	F total
Peak ML motion of COP (mm)	1	0.81 (0.46-0.93)	0.64	0.77(0.67- 0.95)	0.32
	2	0.79(0.42-0.93)	0.59	0.51 (-1.01- 0.89)	1.04
	3	0.84(0.56-0.95)	0.02	0.57 (-0.78- 0.90)	0.56
	4	0.81(0.47-0.93)	0.24	0.50 (-1.07- 0.88)	0.44
Peak AP motion of COP (mm)	1	0.77(0.36-0.92)	0.87	0.61(-0.61-0.91)	25.17
	2	0.92(0.76-0.97)	0.50	0.30(-1.87-0.84)	0.33
	3	0.79(0.42-0.93)	0.99	*	
	4	0.85(0.59-0.95)	0.37	0.62(-0.57-0.91)	0.08
Peak Motion of ML COM_{est} (mm)	1	0.52(-0.33-0.83)	0.65	*	
	2	0.51(-0.35-0.83)	0.98	*	
	3	0.42(-0.63-0.79)	0.60	*	
	4	0.39(-0.69-0.79)	0.56	*	
Peak Motion of AP COM_{est} (mm)	1	0.10(-1.52-0.68)	2.37	0.77(0.07-0.95)	0.14
	2	0.424(-0.61-0.80)	1.24	*	
	3	-0.51(-3.21-0.47)	0.51	*	
	4	-0.37(-2.82-0.51)	0.40	*	
Peak velocity of ML COM_{est} (mm/s)	1	0.73(0.26-0.91)	0.47	0.88(0.50-0.97)	3.15
	2	0.88(0.67-0.96)	0.26	0.80(0.17-0.95)	0.17
	3	0.89(0.69-0.96)	1.02	0.60(-0.64-0.91)	0.67
	4	0.83(0.51-0.94)	2.19	0.54(-0.90-0.89)	0.47
Peak velocity of AP COM_{est} (mm/s)	1	0.51(-0.36-0.83)	2.66	0.48(-1.12-0.88)	0.13
	2	*		0.86(0.43-0.97)	0.18
	3	0.18(-1.28-0.71)	1.38	0.88(0.52-0.97)	0.30
	4	0.08(-1.56-0.68)	1.79	*	
Stepping error (cm)	1	*		*	
	2	*		0.81(0.21-0.96)	0.76
	3	*		*	
	4	*		*	

Key-Data not normally distributed (*), Number of participants (n), Intra-class correlation coefficient (ICC), confidence interval (CI), F statistic (F), centre of pressure(COP), centre of mass (COM), Children with cerebral palsy (CP), Typically developing children (TD)

Table 7 *Inter-rater reliability of the Next Step test of dynamic balance during a constrained stepping task in children with cerebral palsy and typical development.*

Inter-rater reliability	Target	CP n=15 ICC 95% CI limits	F total	TD n=11 ICC 95% CI limits	F total
Peak ML motion of COP (mm)	1	0.78(0.35-0.93)	0.00	0.75(0.08-0.93)	0.02
	2	0.91(0.74-0.97)	1.95	0.45(-1.03-0.85)	0.11
	3	0.90(0.71-0.97)	0.05	0.61(-0.45-0.90)	1.85
	4	0.93(0.80-0.98)	0.03	0.40(-1.22-0.84)	0.06
Peak AP motion of COP (mm)	1	0.57(-0.27-0.86)	0.42	0.37(-1.35-0.83)	4.37
	2	0.91(0.73-0.97)	0.58	0.59(-0.54-0.89)	5.65
	3	0.94(0.83-0.98)	0.17	*	
	4	0.96(0.87-0.99)	0.52	0.39(-1.28-0.84)	1.63
Peak Motion of ML COM_{est} (mm)	1	0.59(-0.29-0.86)	1.03	*	
	2	0.58(-0.25-0.86)	1.27	*	
	3	0.57(-0.29-0.85)	0.65	*	
	4	0.57(-0.26-0.86)	0.20	*	
Peak Motion of AP COM_{est} (mm)	1	0.73(0.19-0.91)	0.03	0.70(-0.11-0.92)	1.15
	2	0.54(-0.37-0.85)	0.034	*	
	3	0.57(-0.30-0.85)	0.98	*	
	4	0.34(-0.97-0.78)	0.20	*	
Peak velocity of ML motion of COM_{est} (mm/s)	1	0.83(0.50-0.94)	0.29	0.88(0.53-0.97)	0.00
	2	0.88(0.64-0.96)	0.03	0.71(-0.75-0.92)	0.01
	3	0.92(0.75-0.97)	0.31	0.53(-0.74-0.87)	0.02
	4	0.89(0.68-0.96)	0.07	0.40(-1.22-0.84)	0.66
Peak velocity of AP motion of COM_{est} (mm/s)	1	0.67(0.03-0.89)	0.75	0.24(-1.85-0.79)	0.62
	2	*		0.73(-0.02-0.93)	0.10
	3	0.56(-0.32-0.85)	0.20	0.78(0.18-0.94)	1.08
	4	0.56(-0.30	0.26	*	
Stepping error (cm)	1	*		*	
	2	*		*	
	3	*		0.56(-0.64-0.88)	0.15
	4	*		*	

Key-Data broke rules of normality (*), Number of participants(n), Intra-class correlation coefficient (ICC), confidence interval (CI), F statistic (F), centre of pressure (COP), centre of mass (COM), Children with cerebral palsy (CP), Typically developing children (TD)



Key- Target 1 (grey /hexagon), Target 2 (black /square), Target 3 (black/circle), Target 4 (grey/triangle), mean difference (solid line), 2 standard deviations (broken line)

Figure 7 *Bland Altman plots displaying differences between raters (top row) of the medio-lateral velocity of the centre of mass targets 1 and 2 (3a) and targets 3 and 4 (3b) and differences between sessions (bottom row) of the medio-lateral velocity of the centre of mass targets 1 and 2 (3c) and targets 3 and 4 (3d).*

The Bland Altman plots (Figure 7 Bland Altman plots displaying differences between raters (top row) of the medio-lateral velocity of the centre of mass targets 1 and 2 (3a) and targets 3 and 4 (3b) and differences between sessions (bottom row) of the medio-lateral velocity of the centre of mass targets 1 and 2 (3c) and targets 3 and 4 (3d).) show there were no systematic differences between sessions or raters. Data points outlying two standard deviations were scrutinised and related to four participants with CP and one child with TD.

The minimum difference (MD) to detect a true change for children with CP, ranges from 23.7 to 29.6 mm, for peak ML motion of COP, across the four targets. The MD for peak velocity of ML COM_{est} ranges between 41.3-61.8mm, with a smaller MD for the medial targets (41.0 and 46.3) and a larger MD for lateral targets (61.8 and 54.5).

2.6.8 Discussion

This study tested the reliability of a novel test of dynamic balance in children aged 8-16 years of age with cerebral palsy (CP) GMFCS I-III and typical development. We have shown that there is good to excellent reliability between raters and, over time, for one rater when measuring the amplitude and velocity of medio-lateral (ML) anticipatory postural adjustments (APAs) in children with CP. The reliability of antero-posterior APAs was less reliable. Data for stepping accuracy were not normally distributed and so reliability was not tested.

Clinical measures of functional mobility in children with CP are reliable in younger age groups. The Pediatric Balance Scale (PBS) has good to excellent reliability [113]. However, it has a ceiling effect equivalent to typical development of age 6 years and is only moderately responsive across all GMFCS levels [114, 115]. This is problematic when considering the balance of younger children with CP at GMFCS levels I and II. These children have better balance, walk in their communities and commonly participate in sport. Therefore, although reliable, the PBS may not detect change in these children with higher levels of function. The Timed Up and Go has excellent reliability (ICC>0.9) and can detect minimal important clinical differences from a change of 0.22s in GMFCS I [116]. The test is also reliable in older adolescents [117, 118]. The Gross Motor Function Measure 88 (GMFM88) also has excellent reliability (ICC>0.9) and can detect smallest real difference of between 1.3 and 2 points on the GMFM 88 in children younger than 10 years GMFCS I and II [119]. However, these functional measures neither capture the quality of the movement nor do they exclude compensatory movements, which may be detrimental in the long term.

The Quality Function Measure (Quality FM) was developed to address the need to measure the quality of movement [20]. It tests the quality of motor performance using the 37 items from the GMFM-66 and is suitable for ambulant children with CP. It uses video analysis of these items and the rater ranks co-ordination, alignment, dissociated movement, stability and weight shift on a scale from 0 to 3. The Quality FM has been shown to have excellent inter and intra-rater reliability [83] but is quite time consuming and requires subjective rating. The concurrent validity of the Next Step test has been measured against the Quality FM and GMFM-66 [85]. There is a positive correlation between peak COP motion and GMFM stand and walk and the Quality FM dimensions of alignment, stability and weight shift.

The Next Step test measures anticipatory postural adjustments (APAs) using quantifiable data. We measured COP and COM_{est} motion which have previously been studied in relation to gait initiation [104]. However, we were interested in APAs within a constrained stepping task which demands different control mechanisms from gait initiation [30]. We have shown that we can reliably measure these APA's to both a medial and lateral target step by calculating the velocity of the peak ML COM_{est}. and the peak motion of the COP. This is the first time that the reliability of these measures of dynamic balance have been reported. The TD and CP children showed differences between groups as highlighted in our previous paper on validity [85]. Despite differences in APA size, velocity [85] and in step length the tests were reliable in each group.

We have defined aspects of the Next step test that are reliable for use in clinical trials with children with CP. Measuring the reliability in a group, such as in a clinical trial, reliability of ICC 0.7 is adequate [105]. The medio-lateral (ML) motion of the COP and the peak ML velocity of the COM_{est} are the most reliable measurements of dynamic balance. However, reliability of ICC> 0.9 would be required to use this as a tool for making decision on individual patient outcomes. The measure also involves expensive and cumbersome motion analysis equipment in its current form. Further work is needed to develop the Next Step test as a clinical outcome measurement tool.

A limitation of this study is that we only recruited one child with GMFCS level III. Therefore, we cannot generalise these results to children with this level of motor function. This study measured reliability of the children stepping 60 times per session. It was noted in some younger TD children or children with CP with greater levels of impairment struggled to maintain consistent stepping responses over the whole session, which would have affected the variability of the data.

Stepping accuracy was previously found to be significantly different between TD children and children with CP [85]. Our data was not normally distributed and so we were unable to calculate measures of reliability. Stepping accuracy was calculated as the absolute distance of the centroid of the stepped foot from the centroid of the target step. We asked the children to take 60 steps randomised to the four targets, so that we had 15 trials per target. This number of trials allowed us to capture the variability of each individual during the trial. However, with taking repeated steps during the task there is a risk of a training effect where the child becomes better at the task the more steps they take. We hypothesised that errors in stepping accuracy were due to poor control of the

trajectory of the ‘throw’ of the COM. While this mechanism is responsible for the movement, the factors affecting accuracy may be multifactorial and differ between steps during the same testing session. Every effort was made to define the target step by marking the outline of the child’s foot in the target position. However, the child might have been aiming their step at the light or the marked target.

The youngest TD children tended to be more fidgety and more inclined to vary the way they moved between trials, whereas older children had a more standard and repeatable movement. The maturation of movement during this constrained stepping task is an area that demands further investigation. Stepping error could be also be affected by difficulties in motor planning, reduced visual acuity, fatigue and difficulty with concentration on the task. Further work is required to measure the level of stepping error attributable to other impairments. In addition, an investigation of other measurement properties such as responsiveness is required. This could be established by assessing the change over time that occurs with a known effective intervention or by measuring the natural disease progression in a neurodegenerative condition [120].

2.6.9 Conclusion

We have presented a novel and reliable measure of anticipatory postural adjustments associated with stepping. The peak medio-lateral motion of the centre of pressure and medio-lateral velocity of the estimate of centre of mass have been shown to be reliable when measuring a constrained stepping task in children with cerebral palsy GMFCS I and II. This data, alongside the previous study of the tests’ validity [85], suggests that the Next Step test has potential for use as a measure of dynamic balance in clinical trials.

2.7 Next Step overall conclusion

While the Next Step provides a reliable measure of anticipatory postural adjustments (APAs), it can be difficult to interpret as it measures APAs to four targets. Therefore, it would be useful to have a summary score. Table 8 shows three summary scores. Firstly, the grand average score that takes the mean of the APAs to the four targets. Secondly, the symmetry score measures the ratio of the APAs between right and left sides, where complete symmetry would be zero. Thirdly, the modulation score calculates the ratio of the size of the APAs when stepping to a medial and lateral target. Of the three scores the grand average showed significance between the two groups for medio-lateral (ML) motion of the COP ($p=0.006$) (Table 8).

2.7.1 Summary score equations:

Grand average score= mean (target 1, target 2, -target 3, -target 4)

Symmetry score= $\frac{\text{(mean (target 1, target 2) + mean (target 3, target 4))}}{\text{Grand average score}}$

Modulation right = (target 1-target 2)/(target 1+ target 2)

Modulation left= (target 3-target 4)/(target 3+ target 4)

Modulation ratio= mean (modulation right : modulation left)

Table 8 *Summary scores for the Next Step*

	MLCOMVEL		ML COP		AP COP	
	TD (n=12)	CP (n=16)	TD (n=12)	CP (n=16)	TD (n=12)	CP (n=16)
Grand average	128.4 (33.2)	107.1 (38.3)	56.0 (14.3)	37.3 * (20.5)	-10.8 (4.8)	-8.3 (6.6)
Symmetry	1.1 (0.2)	1.0 (0.5)	1.1 (0.3)	3.1 (7.2)	1.4 (0.4)	1.1 (1.0)
Modulation average	0.32 (0.12)	0.28 (0.21)	0.10 (0.14)	-0.36 (2.02)	0.1 (0.2)	0.2 (0.6)

Key- number (n), medio-lateral velocity of centre of mass (ML COMVEL), medio-lateral motion of the centre of pressure (ML COP), and antero-posterior motion of the centre of pressure (AP COP), Typically developing children (TD), cerebral palsy (CP), significant (*)

In addition, the reliability of these measures was assessed as summarised in Table 9. The ML and AP COP showed excellent intra- and inter-rater reliability. Therefore, the ML COP will be used as the summary measure in the final analysis of the feasibility RCT.

Table 9 *Reliability of the Next Step summary scores using the Intra-class correlation coefficient*

	Intra	Intra	Intra	Inter	Inter	Inter
	Absolute	Symmetry	Modulation Ratio	Absolute	Symmetry	Modulation Ratio
MLCOMVEL	0.86 (0.68-0.94)	0.20 (-0.8-0.64)	0.004 (-1.27-0.56)	-0.04 (-0.13 – 0.14)	-0.17 (-1.7-0.48)	0.061 (-1.00 – 0.57)
ML COP	0.86 (0.69-0.94)	0.32 (-0.54 – 0.7)	0.09 (-1.05 – 0.60)	0.88 (0.74-0.95)	0.44 (-0.25-0.75)	-0.024 (-1.30-0.54)
AP COP	0.86 (0.72-0.95)	0.87 (0.70-0.94)	0.07 (-1.17 – 0.60)	0.87 (0.70-0.94)	0.35 (-0.46-0.71)	-0.11 (-1.18-0.47)

Key- number (n), medio-lateral velocity of centre of mass (ML COMVEL), medio-lateral motion of the centre of pressure (ML COP), and antero-posterior motion of the centre of pressure (AP COP), Typically developing children (TD), cerebral palsy (CP), Intra-rater reliability (Intra), Inter-rater reliability (Inter)

3 Part 3: Feasibility Trial

This section starts with a consensus study to establish usual physiotherapy care for ambulant children with cerebral palsy (CP). This will inform the control intervention for the feasibility trial. The published protocol for the feasibility will be presented followed by the quantitative and qualitative results and a discussion of the feasibility study.

3.1 Introduction

The overall aim of this thesis is to test the feasibility of a randomised controlled trial (RCT) exploring the effectiveness of training for 10 weeks using the Happy Rehab device, in order to improve walking and balance for children with CP. The following consensus study was undertaken to determine usual physiotherapy care. This third paper from the programme of work (Paper 3) was published in Child: Care, Health and Development (January, 2022) [121]. The version of the paper presented in the thesis has an amendment in Table 10 as a [122] publication. The patient information sheet for this study can be found in Appendix 2.

3.1.1 Paper 3: Defining usual physiotherapy care in ambulant children with cerebral palsy in the UK: A mixed methods consensus study.

Short Title: Defining physiotherapy care for children with CP.

Authors: Rachel Rapson [1,3], Jos M. Latour [2], Jon Marsden [1], Harriet Hughes[1], Bernie Carter[4],

[1] School of Health Professions, Faculty of Health, University of Plymouth PL6 8BH [2] School of Nursing and Midwifery, Faculty of Health, University of Plymouth [3] Torbay and South Devon NHS Trust, Torquay, TQ2 7BA [4] Edge Hill University

Key words: Cerebral palsy, Physiotherapy, Consensus, Nominal group, Walking

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Abstract

Background

Ambulant children with cerebral palsy (CP) undertake physiotherapy to improve balance and walking. However, there are no relevant clinical guidelines to standardise usual physiotherapy care in the UK. A consensus process can be used to define usual physiotherapy care for children with CP. The resulting usual care checklist can support the development of clinical guidelines and be used to measure fidelity to usual care in the control groups of trials for children with CP.

Methods

Twelve expert physiotherapists were recruited. In Phase 1, statements on usual care were developed using a survey and two nominal groups. Phase 2 included a literature review to support usual physiotherapy interventions. Phase 3 used a confirmatory survey, which also captured changes to provision during the COVID-19 pandemic. Consensus was calculated by deriving the mean of the deviations from the median score (MDM). High consensus was deemed to be where MDM<0.42.

Results

Physiotherapists reached high consensus on five outcome measures (MDM range 0-0.375) and nine areas of assessment (MDM range 0-0.25). Physiotherapists reached moderate consensus on task specific training (MDM=0.75), delivered at weekly intensity for 4-6 weeks (MDM=0.43). There was high consensus (MDM=0) that children should participate in modified sport and fitness activities and that children with Gross Motor Function Classification System level III should be monitored on long-term pathways (MDM= 0.29).

Conclusions

Physiotherapists reached consensus on two usual care interventions and a checklist was developed to inform the control groups of future randomised controlled trials. Further consensus work is required to establish clinical guidelines to standardise usual physiotherapy care in the UK

3.1.2 Introduction

Cerebral palsy (CP) is an umbrella term describing a group of permanent disorders affecting the development of posture and movement affecting 2.1 per 1000 children [1]. Motor impairments associated with CP make walking more effortful and significantly

limit children's participation at school and in the community [123]. Children with CP can experience primary movement impairments such as spasticity, weakness or reduced selective movement control [124]. The severity of the movement disorder can be described using the Gross Motor Function Classification System (GMFCS)[125]. Children with GMFCS levels I-III are able to walk with varying levels of support or orthoses and tend to achieve their peak motor performance by age nine [126, 127]. However, secondary musculoskeletal impairments can develop during periods of rapid growth, presenting further challenges to walking and balance skills.

Physiotherapists provide advice and therapeutic interventions aimed at addressing primary impairments and preventing secondary complications of CP. Young people with CP and their families want to know which physiotherapy interventions are the most effective, and the frequency and intensity required to achieve optimum mobility [128]. Physiotherapy service provision may vary depending on resources and how emerging evidence [129-131] and national guidance is implemented [132]. Currently, there is no standardisation of physiotherapy care for ambulant children with CP in the UK.

The highest level of evidence for the effectiveness of an intervention is through meta-analysis of randomised controlled trials (RCTs) [133]. In many physiotherapy studies, the control group undertakes 'usual care', but this is often unspecified. Usual care across studies is likely to vary in the frequency and intensity of physiotherapy, and participants in a control group could be undertaking activities similar to the experimental intervention. It is essential to define usual care within the research setting to ensure the effect size of an intervention within a trial is correctly measured. Therefore, a definition of usual care is crucial to ensure robust research findings and to inform the development of evidence-based clinical pathways [134].

3.1.3 Methods

The aim of this study was to reach consensus on current usual physiotherapy care delivered by physiotherapists in the UK, and to develop a usual care checklist to enable measurement of fidelity to usual care in the control group of a forthcoming feasibility RCT. The Health Research Authority and Health and Care Research Wales (reference 254056) granted permission for this study.

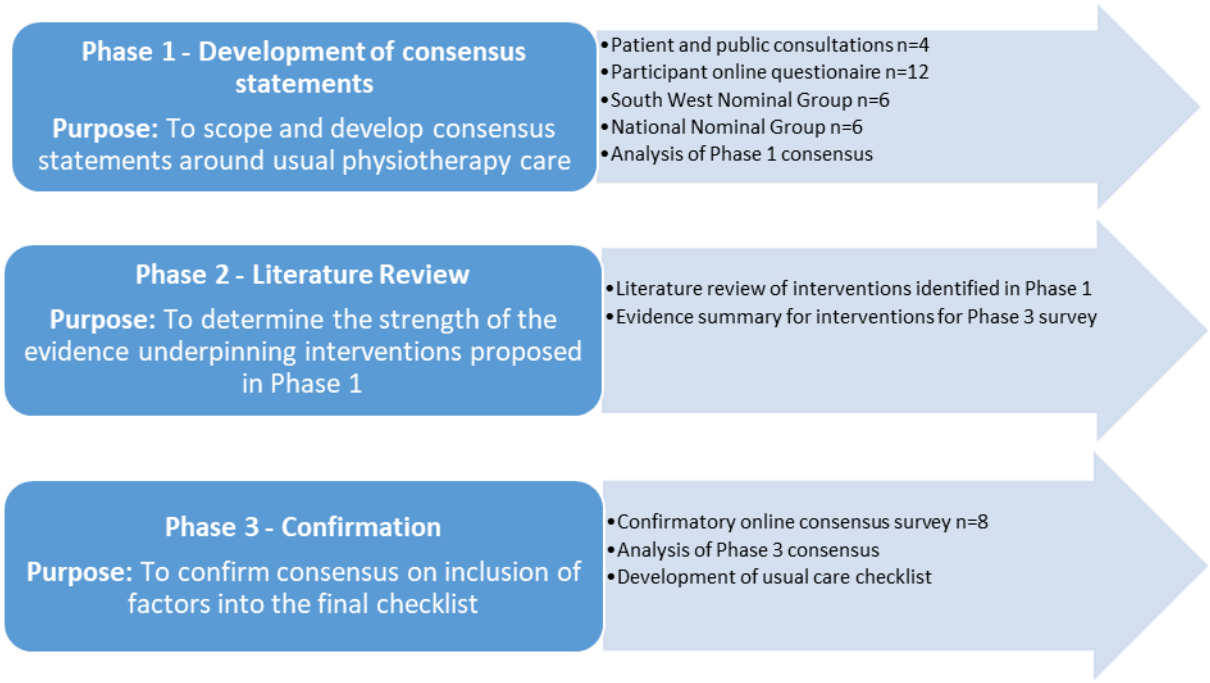

Figure 8 *Flow diagram showing the three phases of the consensus study*

This study adopted a three-phase design (Figure 8). Phase 1 used Idea generation and Nominal Group Technique (NGT) to establish consensus statements on usual physiotherapy care aimed at improving balance and walking in children with CP, GMFCS I-III. Phase 2 was a Literature Review to establish the evidence base underpinning the interventions identified in the consensus statements. Phase 3 used a survey to confirm consensus on the usual care checklist.

3.1.4 Participants

The optimal size for a nominal group (NG) is between 5-12 people [135-137]. Two NGs were established in Phase 1. The first NG consisted of six paediatric community physiotherapists from NHS providers in South West UK. The Physiotherapy Managers of five Child Development Centres recruited participants. They gave information packs to interested clinicians. The manager was asked to nominate one or two staff volunteers to participate, during work time. A National NG was formed with six community physiotherapists from the rest of the UK. Adverts were placed in the Association of Paediatric Chartered Physiotherapists e-bulletin. Interested physiotherapists were invited to respond directly to the chief investigator, who sent them an information pack. Participants were eligible if they had over two years of experience in paediatric physiotherapy and held a current community paediatric caseload in the UK, with a National Health Service (NHS)

provider. In Phase 3, all twelve participants from Phase 1 were invited to complete a confirmatory survey.

3.1.5 Phase 1: Development of consensus statements

Phase 1 employed the Nominal Group Technique (NGT), a consensus process that encourages individual participation and a non-hierarchical exchange of ideas [138]. It has previously been used within physiotherapy to reach consensus on interventions that influence motor development in children with CP [139]. NGT involves a three-stage process of decision making during a structured group meeting led by a skilled, neutral facilitator [139, 140].

Idea Generation

The NGT was modified by using an online questionnaire to develop ideas prior to the NG meetings. In addition to the questionnaire, participants received a clinical scenario, describing a 12-year-old boy with CP (GMFCS level II), to help them frame their responses using an authentic situation. The questionnaire comprised a series of open questions, to explore ideas on what constitutes usual physiotherapy care for him and how it might vary for children of different ages and functional levels. The lead author grouped together the responses generated by participants to form ten statements about usual care. Ideas excluded from the ten statements, where fewer than 20% respondents identified them, were recorded and set aside for discussion and clarification during the NGs.

Nominal Groups

The lead author, an experienced paediatric physiotherapist and researcher, facilitated the NGs. Her position at the group was of a neutral facilitator and other members of the research team supported the process: HH documented notes and JM administered the scoring. Participants were asked to consider the minimal physiotherapy care usually undertaken by a physiotherapist, regardless of NHS setting. Careful consideration was given to the scope of the physiotherapy role. Participants excluded the provision of orthotics, as Orthotists are autonomous practitioners responsible for the assessment and prescription of orthotics.

The statements on usual care were presented to participants at the beginning of the SW NG. Participants scored their level of agreement with each statement using a 5-point Likert type scale (1=strongly disagree, 2=disagree, 3=undecided, 4=agree and 5=strongly agree). The mean group score was calculated for each statement at the end of each scoring round. Participants were presented with the group median score alongside their individual

scores for each statement. The facilitator encouraged a round-robin feedback from the participants for each statement. Participants explored the relative merits of each statement and were able to evaluate their ideas compared to those held by the group. Participants discussed and then revised the statements. The group revisited any ideas previously set aside for further discussion to see if they wished to include them. For example, hydrotherapy was a subject initially set aside, and was revisited by both groups, but remained excluded. Participants re-scored all the statements where consensus was not reached in the previous scoring round.

The statements on usual care developed during the SW NG were presented at the beginning of the National NG, in an iterative process. The National NG decided to include an idea that had been excluded by the SW NG. This was related to the importance of advocating wheelchair mobility for children assessed as GMFCS level III. This was taken forward into Phase 2.

At the end of Phase 1, the levels of consensus for the ten statements on usual physiotherapy care were calculated for each NG. Six physiotherapy interventions were proposed by the NGs as usual care.

3.1.6 Phase 2: Literature review

The aim of the literature review was to appraise the strength of evidence supporting the six interventions proposed for inclusion (in Phase 1) in usual care for ambulant children with CP.

Search strategy

Two researchers (RR and JM) conducted the search for literature systematically. No date limits were set for the search. The initial search took place on 16 December 2019 and was updated as new evidence emerged until 07 July 2020. The databases searched were MEDLINE (EBSCO), EMBASE (EBSCO), PUBMED, The Cochrane Central Register of Controlled Trials, CINAHL, AMED (EBSCO), PEDro, SCOPUS, Google Scholar, ETHOS, PRIMO research outputs and theses.

Initial keywords searched were child OR adolescent AND cerebral palsy AND physiotherapy OR physical therapy AND walking OR gait OR balance AND strength training OR exercise OR progressive resisted exercise OR strengthening OR stretching OR flexibility OR task practice NOT surgical OR Botulinum toxin OR orthotic OR orthoses.

Inclusion and exclusion criteria and study selection

Systematic reviews or studies in the English language were included where they reported physiotherapy interventions with outcomes related to walking and balance. Where no systematic review was found, RCTs and then experimental studies were included. Papers were excluded where the results are reported in a systematic review or were superseded by studies that are more recent. Protocol only publications and papers that did not report an outcome relating to balance or walking were excluded. The results are presented in accordance with PRISMA guidelines [141] shows that of the 670 abstracts reviewed, 105 full papers were retrieved for abstract review; of these, there were 75 systematic reviews, 29 RCTs and 1 experimental design study. Only fifteen papers met the criteria for full review and were assessed for bias using the CASP tool [142]. These comprised twelve systematic reviews [143-155], two RCTs [122, 156] and one non-randomised crossover trial [157]. The strength of evidence for interventions identified as usual care were rated as high, moderate, low or very low levels of evidence [158].

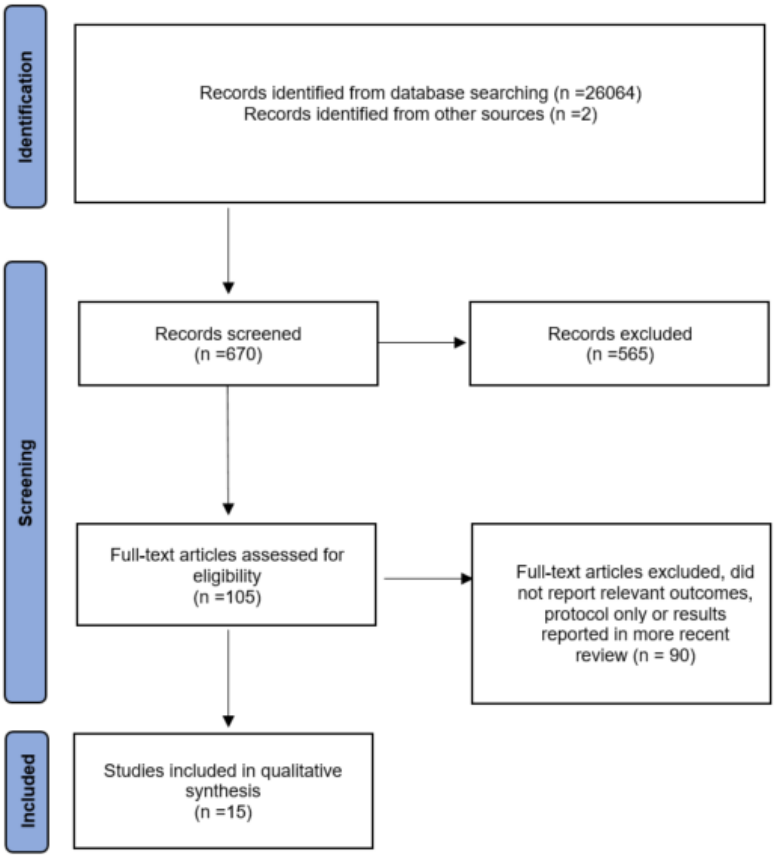

Figure 9 *PRISMA diagram showing the flow of citations reviewed within the literature review.*

3.1.7 Phase 3: Confirmatory Survey

The final online survey allowed participants to score subsections of each statement of usual care in more detail. For example, participants were asked to rate individual assessment tools from the list identified in Phase 1 using the 5-point Likert type scale. Interventions were presented alongside the evidence summary (Table 10) and participants were asked to indicate whether they thought the intervention should be included or excluded as usual care, or if they were undecided. Participants were asked to comment on why they decided to award each score in order to gain more insight into their views and experiences.

3.1.8 Analysis

Consensus was calculated by deriving the mean of the deviations from the median score (MDM) using the following equation [159]:

$$MDM = \frac{Sum\ of\ individual\ deviations\ from\ the\ median}{Number\ of\ participants}$$

High consensus (MDM <0.42) is required for any treatment intervention to be considered important for inclusion e.g. type of exercise, whereas moderate consensus (MDM = 0.42-0.81) is acceptable for other aspects of the programme setting such as method of delivery [137].

Text from the idea generation questionnaire, quotations noted during the nominal groups and responses from the confirmatory survey were transcribed and coded as follows: P representing participant, followed by participant number and either NG=nominal group or S=survey to show at which stage it was said. The confirmatory survey produced anonymous responses from individuals representing both NGs. The text was explored using a framework analysis approach.

3.1.9 Results

Twelve physiotherapists participated across the two NGs in Phase 1. The median age of participants was 43 years (range 28-60) with a median level post qualification of 21.5 years (range 7-38) with 18.5 years (range 3-29) in paediatrics. Table 11 shows the similarity between both NGs. Eight of the twelve participants completed the Phase 3 Confirmatory anonymous survey.

Table 10 *Evidence summary for physiotherapy interventions aimed at improving walking and balance for children with cerebral palsy.*

Intervention	Evidence the intervention improves balance or walking	Evidence strength	Reference
Participation in physical activities	Aerobic and fitness training improves gross motor function.	Moderate	[148]
	Modified sport improves balance and walking.	Low	[148]
Flexibility exercise	No evidence found.	Very low	
Prolonged passive stretching	Serial casting of the ankle improves in gait parameters in the short term (<12 weeks effect) but it is unclear whether there is functional benefit.	Low	[160]
	Serial casting does not improve stride length.	Very low	[149]
	Prolonged standing in a frame or tilt table for 45 mins, 3 times a week may have a short term, positive effect on gait parameters.	Low	[157]
Strength training	Strength training using progressive resisted exercise does not improve gross motor function, gait speed and gait characteristics.	High	[147-149]
	Progressive resisted exercise does not improve postural control in standing.	Moderate	[147]
	Gross motor activity training with progressive resisted training (e.g. loaded sit to stand) does not improve gross motor function and is associated with multiple adverse events.	Moderate	[148]
Task specific training and functional activity training	Gross motor activity training improves gross motor function when undertaken in real world situations with variable practice of skills.	Moderate	[146, 148]
	Gross motor task training of 1 hour, 2-5 times per week for 5- 6 weeks improves postural stability during gait.	Moderate	[147]
	Mobility training, treadmill training, and partial body-weight support treadmill-training increases walking and stride length at a dose of 15-30 mins, 2-7 times per week for 6-8 weeks.	Moderate	[144-146, 148, 149]
	Treadmill training (excluding partial body weight supported) improves balance and postural control.	Moderate	[147]
	Backward gait training improves balance, gross motor function, step length and walking velocity at a dose of 15-25 minutes 3 times per week for 6-12 weeks.	Moderate	[143]
	Partial body-weight support treadmill training improves gross motor function and walking endurance.	Low	[145]
Postural stability and balance activities	Full body vibration training improves gait speed at a dose of 9-18 minutes, 3 times per week for 8 weeks.	High	[149]
	Trunk training on vibration plate improves trunk alignment during gait.	Moderate	[147]
	Neurodevelopmental therapy for 30 mins twice a week for 8 weeks did not improve standing balance in children with spastic diplegia.	Low	[147]

Participants developed ten statements on usual care during the NGs. They described six areas of intervention to be included in the literature review: participation in physical activities, flexibility exercises, prolonged passive stretching; strength training; and task

specific or functional activity training. Participants identified a list of assessment tools and outcome measures to be included in the Confirmatory survey. Both groups reached a high level of consensus (MDM <0.42) for all ten statements on usual care at the end of the Phase 1 process (Table 12). Participants in the SW group tended to award a higher median score for each topic.

Table 11 *Mean age, location and experience of participants*

	All participants n=12	South West NG n=6	National NG n=6
Median participant age (range) years	43 (28-60)	40 (28-60)	45 (31-59)
Median number years (range) qualified as a physiotherapist	21.5 (7-38)	18 (7-39)	23 (7-38)
Median number years (range) working in paediatrics	18.5 (3-29)	15 (3-29)	20.5 (7-25)
Location of NHS Providers represented		Plymouth, Exeter, Torquay, Truro	Chelmsford, Kent, Leicester, London, Medway, Yorkshire,

Key- N=number, NG=Nominal Group

The literature review appraised evidence for the six interventions identified as usual care during Phase 1. Evidence for each intervention was explored in relation to outcomes of walking, balance and gross motor function. The evidence summary (Table 10) shows moderate to low evidence to support fitness training and modified sport. There was low evidence supporting prolonged passive stretching (excluding orthotics) using serial casting or prolonged standing frame use. There was moderate to high evidence against the use of progressive strength training. Strength training did not improve gait characteristics or postural control and was associated with multiple adverse events.

Task specific training, focusing on gait training on the treadmill or on the ground, was supported by a large evidence base, with low to moderate evidence supporting its use. There was moderate to high evidence supporting the use of vibration plate training for postural stability and improving gait, and low evidence against the use of neurodevelopmental therapy for standing balance. There was an absence of literature to support flexibility, postural stability or balance exercises as described by participants.

The results in Table 12, amalgamate the consensus responses with the results of the literature review. Results are presented under two main themes: 'Physiotherapy Service Provision and Structure', and 'Physiotherapy Interventions'. Consensus scores are presented for each statement topic alongside direct quotations from the participants. Where a view was sustained from Phase 1, this is documented to show how the view was developed.

Table 12 *The level of consensus scoring of statements of usual care in Phase 1*

Statement topic	SW group		National group		
	Median score	MDM	Median score	MDM	Level of Consensus
Referral and discharge	5	0.25	4	0.17	High
Location of therapy	4.5	0.38	5	0	High
Frequency and intensity	5	0.25	4.5	0	High
Advice and information	5	0	5	0.33	High
Goals setting	5	0.5	5	0.33	High
Assessment tools	5	0.25	4.5	0	High
Outcome measures	5	0.25	5	0.5	High
Interventions	5	0	4.5	0	High
When frequency and intensity of physiotherapy differs	5	0.25	5	0.33	High
How intervention differs in relation to GMFCS level	5	0	4	0.33	High
How outcome measure differs in relation to GMFCS level	5	0	5	0.5	High
How intervention differs in relation to the child's age	5	0	4.5	0	High
How outcome measure differs in relation to the child's age	5	0	5	0.5	High

Key- GMFCS=Gross Motor Function Classification Scale, SW=South West, MDM=mean deviation from median

3.1.9.1 Physiotherapy Service Provision and Structure

Referral and discharge criteria

There was high consensus (MDM=0.29) that children with GMFCS level III should remain on a long-term pathway, from initial referral until they transition to adult services. This view was sustained from Phase 1 to Phase 3, for example:

> *"Children with GMFCS III are more likely to develop joint contractures and muscle shortening affecting function …. They have on going equipment needs"* (P4-NG).

The pathway should include monitoring schedules for range of motion and hip surveillance, such as the Cerebral Palsy Integrated Pathway (CPIP), and continue until skeletal maturity [161]. There was high consensus (MDM=0.14) that children at GMFCS levels I and II require episodes of care related to individual need as P8 explains:

> *"They may also run into difficulties around growth spurts but can be given red flag information for re-referral"* (P8-NG).

Participants supported the prioritisation of early intervention in younger or newly diagnosed children.

Location of physiotherapy appointments

High consensus established that usual care takes place in a children's outpatient clinic (MDM=0) and that appointments occur at school or home (MDM=0.14) when there are equipment or environmental needs. This is often due to post-surgical rehabilitation programmes or co-morbidities such as learning disability, where treating the child in the context of their usual environment is deemed to be more effective. Physiotherapists frequently visit schools to train support workers to deliver a delegated programme of usual physiotherapy care. Time efficiency was a factor affecting this choice:

> *"It is … more time-efficient to see children in the department. However, we carry out home or school visits if indicated to review equipment or specific activities related to school or home environment"* (P3-S).

Frequency and intensity of physiotherapy input

There was high consensus (MDM=0) that the clinical needs of the children dictate the frequency and intensity of blocks of treatment and reviews There was moderate agreement (MDM=0.43) that blocks occur once per week for 4-6 weeks. This was first identified in Phase 1 and sustained in Phase 3:

> *"4-6 treatments appear to be what is manageable for children and their families to follow a more demanding therapy regime. It allows for review of goals and monitor[ing] progress in a defined timespan"* (P11-NG).

There was high consensus (MDM=0) that children receiving physiotherapy should be routinely reviewed every three to twelve months. There was high consensus (MDM= 0.25) that physiotherapy is needed more often in early years and especially during transition to nursery, school and adult services. Physiotherapy support may be required more frequently when parents have additional needs, such as learning disabilities.

There was high consensus (MDM=0) that intensive blocks of physiotherapy rehabilitation are indicated following procedures (e.g. botulinum toxin injections, orthopaedic surgery and serial casting), during growth spurts and where there are changes in spasticity medications or orthotic provision. There was high consensus (MDM=0.38) that rehabilitation after selective dorsal rhizotomy (SDR) surgery requires a highly intense period of rehabilitation, several times per week over 12 or more months (and requires a specific funding package).

3.1.9.2 Advice, training and information

There was high consensus (MDM=0.29) that physiotherapists play an important role in supporting children and their families to understand the impact of their diagnosis and the prognosis of their condition. Participants reached high consensus (MDM=0.29) on the importance of sharing information across agencies, where parents and children give their consent. This typically includes information in the form reports and Education and Health Care Plans (EHCPs) [162] and training for parents and teaching staff who deliver the child's therapy programme. Physiotherapists also provide information regarding local and national resources, such as the statutory local offer, charitable organisations and support groups. The group emphasised the value of this, with a typical response being:

"We could do more to educate wider school staff and potentially other pupils to help them understand the condition and how it effects an individual" (P3-S).

3.1.9.3 Goal setting

There was high consensus (MDM=0.25) that physiotherapists use the Specific Measurable Achievable Realistic Timed (SMART) goal setting approach. Participants emphasised the need to set goals collaboratively, at the level of participation rather than body structure and function [163]:

"A goal needs to be meaningful to the child/family rather than medical. It can quite often be challenging to make a meaningful goal out of a medical need e.g. better heel strike may be achieved and step length improved but the family struggle to see a functional benefit and we don't spend enough time exploring what this gain means to them in terms of their life demands" (P2-S).

Table 13 The level of consensus on assessment tools for Phase 3

Assessment parameter	Median score	MDM	Level of consensus
Gait analysis (video/observation)	5	0.125	High
Pain	5	0.5	High
Leg length	5	0	High
Spinal posture	5	0.125	High
Muscle tone	5	0	High
Muscle power	5	0	High
Range of movement	5	0	High
Functional task performance	5	0.125	High
Patterns of movement	5	0.25	High
Gross motor function	4	0.75	Moderate
Psychosocial	4	0.75	Moderate

Key- MDM=mean deviation from median

3.1.9.4 Assessment

Participants identified eleven areas of assessment of mobility and balance in Phase 1. In Phase 3, participants reached high consensus (MDM range 0-0.25) for nine areas of assessment covering function, range of movement, muscle tone, gait, posture and pain (Table 13).

Table 14 The level of consensus on outcome measures for Phase 3

Outcome measure	Median score	MDM	Level of consensus
Passive range of motion	4	0.125	High
Modified Ashworth	5	0.375	High
Instrumented gait analysis	5	0.125	High
Gross Motor Function Measure (any)	4	0	High
Observational gait scale	4.5	0.375	High
Patient Reported Outcome Measures	3	0.625	Moderate
Modified Tardieu scale	3.5	0.75	Moderate
Therapy Outcome measures	3	0.75	Moderate
10 metre walk test	3.5	0.75	Moderate
Timed up and go	2.5	1.375	None
Edinburgh gait scale	2	1.875	None
Muscle power sprint test	2.5	1.625	None
Pediatric Balance Scale	3	1.375	None
6 minute walk test	3	0.875	None
Berg balance	3.5	1.625	None
Gross Motor Challenge Module	2.5	1.625	None
Quality Function Measure	3	1.375	None

Key- MDM=mean deviation from median

3.1.9.5 Outcome measurement

In Phase 1, participants developed a list of seventeen outcome measures used to evaluate episodes of care. Table 14 shows the high level of Phase 3 consensus (MDM range 0-0.375) for five individual tools measuring gait, muscle tone, range of movement and motor function. Participants discussed the conflict between wanting to use appropriate tools and barriers to being able to use them, with P5 noting:

> *"Outcome measures used depend on time, space and equipment resources, as well as CYP compliance"* (P5-NG).

3.1.9.6 Equipment advice, provision and referral

There was high consensus (MDM= 0.29) that physiotherapists usually provide mobility equipment and refer onto orthotic and wheelchair providers. There was high consensus (MDM=0) that children with GMFCS level III require a 24-hour postural management plan and assessment for alternate powered or wheelchair mobility to improve participation with school and leisure activities.

Physiotherapists advocate for children to have choice about their mobility, P3 noted that:

> *"Wheelchair mobility [is] considered if it will improve independence and quality of life by improving access to community, reduce fatigue and pain levels. [We] want to encourage weight bearing and mobility but not at detriment to child's independence and participation"* (P3-S).

3.1.9.7 Physiotherapy interventions

In Phase 1 participants reached a high level of consensus on a list of interventions considered as usual care (Table 12). However, after consideration of the evidence summary (Table 10) presented alongside the survey, participants only reached consensus on including two of the six interventions into the usual care position statement (Table 15).

Participation in sport and activity

There was high consensus (MDM=0) that the physiotherapist's role is to encourage physical activities and facilitate children to access school and community resources to develop active lifestyles. Physiotherapists considered that the level of daily activity makes an important difference to the outcomes of children. They recognised that the level of support from home and school is critical, for example:

> *"It is important that the child becomes part of the community and accesses local resources. It is part of a life-long strategy"* (P8-S).

Flexibility exercises

Physiotherapists described active flexibility exercises, that move joints through full range, as usual care In Phase 1. Discussions concerning growth spurts frequently acknowledged that reduced range of movement (ROM) must be addressed in order to maintain the flexibility required for effective walking and balance. P5 noted that flexibility exercises are a:

"useful adjunct in children who have stiff joints, MSK/postural asymmetry or who are tight due to growth spurts, to help to maintain ROM and flexibility, which helps with gait pattern, biomechanics and alignment" (P5-S).

The literature review failed to find evidence that flexibility exercises improve balance and walking. While three respondents wished to include this in usual care, there was low consensus (MDM=0.86) in Phase 3.

Table 15 *The level of consensus on interventions included in the usual care position statement*

Intervention	Median score	MDM	Level of consensus
Participation in sport and activity	5	0	High
Flexibility exercises	3	1	Low
Prolonged passive stretching	4	1.75	Low
Strength training	3	1.5	Low
Task specific training and functional activity	5	0.75	Moderate
Postural stability and balance exercises	3	1	Low

Key-MDM=mean deviation from median

Prolonged passive stretching

In Phase 1 participants reached high consensus (MDM=0) that prolonged passive stretching should be included in the list of usual care interventions. In Phase 3 there was low consensus (MDM=0.86) that it should be included in the final position statement. The evidence summary focused on serial casting and standing frame use as being interventions provided by physiotherapists that deliver prolonged passive stretch. Prolonged passive stretching is more frequently provided using orthotics, a topic excluded in this study. There was divided opinion on inclusion between physiotherapists. While the median score indicated that it should be included, there was low consensus on this. P5 explained how they use serial casting in individual cases, rather than as usual care:

"Serial casting [may be used] on an individual basis e.g. to gain lost dorsiflexion, to enable an optimal AFO (Ankle-foot orthoses) to be provided" (P5-S).

P6 also described a more individual approach to using standing frames, in the presence of a specific risk:

> *"I would only prescribe a standing frame for a child who is clearly at risk of developing knee flexion contractures, not as routine intervention"* (P6-S).

Strength training

Strength training was identified as a key intervention in Phase 1. However, in Phase 3 there was low consensus on including this in usual care. The evidence summary highlighted the adverse events associated with this intervention and the lack of evidence that progressive resisted strength training improves walking and balance. Clinicians discussed integrating different exercise approaches that work through range of motion while working against resistance, for example, P3 reasoned:

> *"Evidence is strong against the use of strengthening exercises. But is this because it was used in isolation, when in usual care we use a combination of different exercises/techniques to improve gait/balance. E.g., strengthening in addition to flexibility and range of movement in ankle/knee"* (P3-S).

Task specific training and functional activity

There was moderate consensus that task specific training should be included in usual physiotherapy care. Task specific training within this context involves treadmill training, gait training and practising balance in functional situations. Participants' reservations over the availability of equipment such as treadmills influenced the consensus score e.g., P4:

> *"Elements of task specific training should be included, when it can be performed at home and school environment. Not all Trusts have access to treadmill training so I would question whether this form of 'task specific training' is usual care"* (P4-S).

Postural stability and balance exercises

There was strong consensus in Phase 1 that postural stability and balance activities are used to improve walking and balance. However, after consideration of the available evidence, there was low consensus on inclusion into usual care (MDM=1). The

literature review found evidence that supported the use of vibration plate training, which does not seem to be widely used in clinical practice, as voiced by P7:

> *"I have not used full body vibration training so cannot comment on this type of therapy intervention*" (P7-S).

P5 talked about how they usually provide postural stability and balance exercise in a clinical setting:

> *"[We] routinely provide postural stability and balance activities e.g. use of balance board"* (P5-S).

P2 was typical of the participants in expressing the way they combine approaches to include exercise targeting balance and posture:

> *"Fun recreational activities are important for compliance and should be incorporated into daily life. Within these there will be elements of flexibility exercise, posture and balance"* (P2-S).

3.1.9.8 The impact of the COVID-19 pandemic on usual care

The final phase of this study was carried out during the COVID-19 pandemic, which may have influenced the results. The Confirmatory survey was expanded to capture how usual care changed due to COVID-19. All respondents reported the swift introduction of virtual appointments by video or telephone. These consultations had both positive and negative consequences, as outlined by P2:

> *"This has not been ideal in terms of assessment of body function but has advantages for functional assessment [of children] in their own environment"* (P2-S).

Participants reported that assessments by virtual consultations were incomplete as they lacked manual assessment of movement quality, which affected clinical analysis and decision-making. Some assessment and outcome measurement tools were not achievable during virtual consultations. Assessment of physical impairment was very limited, as explained by P3:

> *"[We are] unable to ascertain strength/power/tone without hands-on assessment or equipment, [we] can ask parents to measure range of movement but not as reliable as therapist due to angle of camera when carrying out virtual assessments. Parents have been able to*

send us videos of walking/other activities which has allowed us to compare side-by-side and review in slow motion to fully analyse" (P3-S).

All participants said that essential face-to-face visits were possible for some children at home or at COVID-secure premises.

Many respondents reported that they provided an assessment and management programme, but they were unable to offer routine monitoring or blocks of treatment at the height of the pandemic. The overall frequency and amount of contact per child has therefore reduced dramatically. All participants reported that children had reduced levels of activity in lockdown due to lack of access to sports facilities at school and in the community.

3.1.10 Discussion

In this study, we explored ideas of what constitutes usual physiotherapy care to improve walking and balance for ambulant children with CP in the UK. The study used a nominal group consensus process. We examined the evidence supporting the interventions usually employed and developed a checklist of usual physiotherapy care for use in a future RCT (Appendix 3).

We found a high level of consensus among physiotherapists to support the long-term monitoring of children with CP at risk of musculoskeletal decline. This approach is backed by a growing evidence base that advocates routine surveillance of hip migration, joint range of motion and spinal posture for all children with CP [7]. Where services do not currently include all children with CP in surveillance programmes, they give 'red flag' indicators for enabling timely access back into services. Physiotherapists play an essential role in identifying the need for orthotic and postural management equipment to optimise posture and mobility for children with CP.

Physiotherapists use collaborative goal setting to inform the need for treatment blocks usually delivered at an intensity of once per week, for 4-6 weeks. This contrasts with the frequency and intensity of usual physiotherapy care reported in some RCTs as 1-3 sessions of 30-60 minutes per week [164, 165]. Participants reached moderate consensus that task specific functional activity training should be included in usual care to improve balance and mobility. This is supported by both the National Institute of Clinical Excellence guidance [166] and the evidence summary produced from the literature review. However, the reported frequency and intensity falls short of the dose

reported to be effective in the literature. Intensive programmes delivered daily for 2 weeks have been shown to achieve the greatest functional improvements [167]. This level of resourcing for physiotherapy treatment programmes was not found within our study, which brings into question the ecological validity of these studies. Physiotherapy services in the UK might consider the efficiency gains of deploying current resources in a more concentrated way. Physiotherapists in our study applied the principles of research findings by integrating gait training in community, home and school activity programmes.

We found further divergence between the evidence and usual care delivered in the UK. Barriers to implementing evidence included lack of knowledge of new interventions such as vibration therapy. Additionally, physiotherapists reported lack of access to equipment such as body weight support treadmill and vibration plates. Our results show that there is a need for translation of research findings into clinical practice through dissemination of knowledge, appropriate resourcing and prioritising evidenced based interventions. Development of national clinical guidelines for paediatric physiotherapy may help to inform optimal use of precious resources.

Physiotherapy interventions for prolonged passive stretching alone were not considered usual care for all ambulant children. Physiotherapists consider the functional and social impact of using serial casting or standing frames with the child and caregivers and may choose to use them in individual cases. Physiotherapists have an important role in promoting independence and developing self-advocacy in the children that they work with. Sometimes the needs of the child might differ from those of the parents. For instance, some parents request that the focus of therapy should be on improving walking when children with GMFCS III might find that wheeled mobility increases their level of participation with peers. Physiotherapists were strident in promoting participation and emphasising the voice of the child.

The main limitation to this study emerged during Phase 3 of the study. High levels of consensus on interventions were reached during Phase 1 and 2. During Phase 3 participants only reached consensus on two from the initial six interventions considered usual care. This may have been due to the smaller number of respondents in the final confirmatory survey. Furthermore, there was no opportunity at this stage for discussion of what participants understood by the evidence summary or newly emerged ideas, which possibly led to more variation in scoring and lower consensus. Participants in the study did not represent the whole of the UK, despite national advertising during the

recruitment phase. This is a limitation as there may be wider variance from the consensus on usual care across and within the four countries. Another limiting factor of this study was that we only considered physiotherapy as delivered by physiotherapists. However, usual physiotherapy care programmes are delivered by parents and carers. Therefore, it is essential to measure this activity when measuring adherence to usual care in a trial control group.

In 2020, when the study was carried out, the COVID-19 pandemic hugely influenced the provision of usual care for ambulant children with CP. School closure resulted in lack of access to therapeutic classroom support and equipment. It is likely that many parents and guardians were unable to replicate therapy provision at home due to work, other care responsibilities or their own health needs. Children had difficulty accessing usual recreational activities during lockdown and shielding. While the full effect of this pandemic on services for children has yet to be evaluated, this study was able to capture the initial adaptations in the delivery of usual care.

This study used a modified NGT consensus process to develop a position statement and checklist of usual physiotherapy care aimed at improving walking and balance in children with CP in the UK. It is important for RCTs to define the usual care carried out in a control group to measure the effectiveness of a novel intervention. We found that physiotherapists combine heterogeneous approaches and create tailor-made programmes to meet the needs of individual children and families. The frequency and intensity of physiotherapy interventions falls short of dosage reported to be effective in the literature.

3.1.11 Conclusion

Physiotherapists reached consensus on two usual care interventions and a checklist was developed to inform future randomised controlled trials. Further consensus work is required to establish clinical guidelines to standardise usual physiotherapy care in the UK. This study is a first step towards defining physiotherapy care effective at improving balance and walking for ambulant children with CP in the UK.

3.1.12 Key messages

- A checklist of usual physiotherapy care in the UK has been developed for ambulant children with cerebral palsy, to inform the control groups in randomised controlled trials.

- Usual physiotherapy care should include task-focused therapy, facilitation of modified sport and participation in community activity.
- Physiotherapy tools were identified for the assessment of balance and mobility and measurement of treatment outcomes.
- Children with Gross Motor Function Classification System level III should remain on long term monitoring pathways.
- The usual intensity of physiotherapy treatment in the UK is weekly for 4-6 weeks and is lower than that which is reported to be effective in research literature.

A longer table of quotations and themes can be found in Appendix 4.

3.1.13 Reflections on the consensus study

This study set out with the aim of mapping 'usual care' as it is delivered in the UK. In phase one, usual care topics were developed through the nominal group process. However, evidence from the literature changed the consensus scoring on the interventions to such an extent that the nominal group decided to only include two of the six interventions in the final usual care checklist.

This is a weakness in the methodology of this study as the introduction of the evidence base steered the usual care checklist towards developing evidence-based guidelines, rather than reflecting usual care as it is practiced.

3.2 Feasibility Study Protocol

The feasibility study aimed to determine the acceptability of the intervention, to test the processes in the protocol and to examine the appropriateness of the proposed outcome measures, in line with MRC framework for complex interventions [25]. The protocol for this study (Paper 4) was published online in the British Medical Journal in month/year [168]. The participant information sheet, detailing the trial procedures, can be found in Appendix 5. The statistical analysis plan is included as Appendix 6.

3.3 Paper 4: A multicentre, randomised controlled feasibility study to compare a 10-week physiotherapy programme using an interactive exercise training device to improve walking and balance, to usual care of children with cerebral palsy aged 4-18 years: The ACCEPT study protocol.

Authors: Rachel Rapson,[1,2] Jonathan Marsden,[3] Jos Latour,[4] Wendy Ingram,[5] Kara Nicola Stevens,[6] Laura Cocking,[7] and Bernie Carter[8]

[1]Physiotherapy, Children and Family Health Devon, Torbay and South Devon National Health Service Foundation Trust, Torquay, UK

[2]Faculty of Health and human Sciences, University of Plymouth, Plymouth, UK

[3]University of Plymouth Faculty of Health and Human Sciences, Plymouth, UK

[4]University of Plymouth School of Nursing and Midwifery, Plymouth, UK

[5]Peninsula Clinical Trials Unit, Plymouth University Peninsula Schools of Medicine and Dentistry, Plymouth, UK

[6]Medical Statistics, University of Plymouth, Plymouth, UK

[7]Peninsula Clinical Trials Unit at Plymouth University (PenCTU), University of Plymouth, Plymouth, UK

[8]Faculty of Health and Social Care, Edge Hill University, Ormskirk, UK

Keywords: Paediatric neurology, Rehabilitation medicine, Developmental neurology & neurodisability

Date of publication: March 2023

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Rapson, R., Marsden, J., Latour, J., Ingram, W., Stevens, K. N., Cocking, L., & Carter, B. (2022). Protocol: Multicentre, randomised controlled feasibility study to compare a 10-week physiotherapy programme using an interactive exercise training device to improve walking and balance, to usual care of children with cerebral palsy aged 4–18 years: the ACCEPT study protocol. *BMJ Open*, *12*(5).

3.3.1 Abstract

Introduction

Children with cerebral palsy (CP) frequently undertake physiotherapy programmes to improve walking and balance. They often require adult support to exercise in a functional position. A novel interactive exercise trainer has been devised to enable children to exercise with against resistance in a functional position, but its efficacy has yet to be proved. A novel protocol has been developed to determine whether a randomised controlled trial (RCT) is feasible.

Aim

To establish whether it is feasible to conduct an RCT to assess the effectiveness of a ten-week physiotherapy intervention using an interactive trainer in children with CP.

Methods and analysis

This study is multi-centre randomised controlled feasibility trial with an embedded qualitative study. Forty children with cerebral palsy, gross motor function classification system I-III will be recruited from community paediatric physiotherapy caseloads. Participants will be randomised to 10 weeks of training with the interactive training device or to usual physiotherapy care. The medio-lateral motion of the centre of mass estimate and Pediatric Balance Scale will be explored as potential primary outcomes measures, tested at baseline, 10 weeks and follow up at 20 weeks. The views of child participants, their parents and physiotherapists will be gained through e-diaries and qualitative interviews.

Feasibility will be determined by examining recruitment and retention rates, completeness of, adherence to the intervention, appropriateness of outcome measures and effectiveness of blinding. Results will be reported in accordance to CONSORT guidelines.

Ethics and Dissemination

Physiotherapists, children and parents have informed trial design and information leaflets. Results will be disseminated via publications, conferences and to families. This study has approval from North of Scotland Research Ethics Committee (20/NS/0018).

Trials registration number

ISRCTN80878394

www.isrctn.com/ISRCTN80878394

Keywords: Cerebral Palsy, Physiotherapy, Balance, Walking, Quality of Life, Gaming

Strengths and Limitations of this Study

- We use a mixed methods approach to assess the feasibility of a proposed RCT
- This protocol tests the feasibility of two potential primary outcome measures
- This study is not designed to determine differences in outcome measurements

3.3.2 Introduction

Cerebral palsy (CP) is a group of permanent disorders affecting the development of movement and posture that occurs in 2.1 per 1000 children worldwide[1]. Difficulties with walking and balance are common and can limit participation in schooling and functional activities [123, 169-171]. There are multiple causes of walking difficulties in children with CP, including spasticity and weakness, which affects 80% of children [7]. Additionally, children with CP often have poor balance, which further impacts on everyday functional tasks, such as dressing [172].

Walking ability can be classified using the Gross Motor Function Classification system (GMFCS) [173]. Children with GMFCS I-III are the focus of the proposed study. Children with GMFCS classification I-II are able to walk functionally outdoors, while children with grade III GMFCS require walking aids.

Physiotherapists frequently prescribe exercise programmes for children with CP aimed at maintaining range of movement, strengthening weak muscles and developing balance skills. In many cases, the children find it hard to undertake exercises in functional positions such as standing, without support from an adult. The Happy Rehab™ (Innovaid, Denmark) interactive exercise trainer was developed (see Figure 9) to help children exercise more independently in a functional supported standing position. It provides support around the hips and additional assistance and resistance via motors aligned to the ankle and knees. This allows the child to exercise muscles functionally in novel ranges, e.g., strengthening the thigh muscles with the hip and knee in a straighter position. The games-based exercises may increase motivation and require the child to control the games by moving their weight side-to-side, forward and backward. It is proposed that this may improve balance during dynamic tasks such as walking.

A small scale study of the interactive trainer found marked improvements in walking, but had a number of limitations in terms of outcome measures used, lack of follow up and control group [174]. Therefore, evidence is still required to establish the efficacy of the equipment. Initially a study is required to establish the feasibility of such a trial within a community physiotherapy service.

Figure 10 *The Happy Rehab™ interactive exercise gaming device. Permission obtained from Innovaid.*

This study aims to establish whether it is feasible to conduct a randomised controlled trial (RCT) of this complex intervention, and to assess the acceptability of the interactive trainer and the trial protocol to physiotherapists, children and their families.

3.3.3 Objectives

The objectives of the study were to:

1) Determine the feasibility of a definitive trial

2) Determine the acceptability of the intervention

3) Explore the views of a sub-group of study participants.

The trial objectives were measured by the outcomes set out in Table 16.

3.3.4 **Methods and analysis**

3.3.4.1 Trial design and setting

The research question is as follows: Is it feasible to conduct a multi-centre randomised control trial of a physiotherapy programme using an interactive exercise trainer to improve balance in ambulant children with cerebral palsy?

This trial is a single-blinded; multi-centre feasibility randomised controlled trial with an embedded qualitative study. Community paediatric physiotherapists working at Child Development Centres (CDCs) will recruit children from their caseloads. The study will be conducted between 09.02.2021 to 01.08.2022. Participants will be randomly allocated to training with the Happy Rehab™ device, or to the control group of usual physiotherapy care. Both groups will carry out 10 weeks training at home, the clinic or their school. The study will compare the intensity of training in different settings. Qualitative semi-structured interviews with a subgroup of physiotherapists, parents and children will take place to explore their experiences of taking part; interviews will take place in the clinic or child's home.

The research question can be framed in the following way:

P	Population – Children with cerebral palsy aged 4-18 years
I	Intervention – A programme of physiotherapy using the interactive training device
C	Comparison group – Usual care
O	Outcome of interest – Feasibility of the trial and intervention
T	Time – Training three times per week for 10 weeks, plus follow up at week 10 and week 20

The trial flow chart is shown in Figure 11.

3.3.4.2 Participants

The eligibility criteria for participants are shown in Table 17 Eligibility criteria.

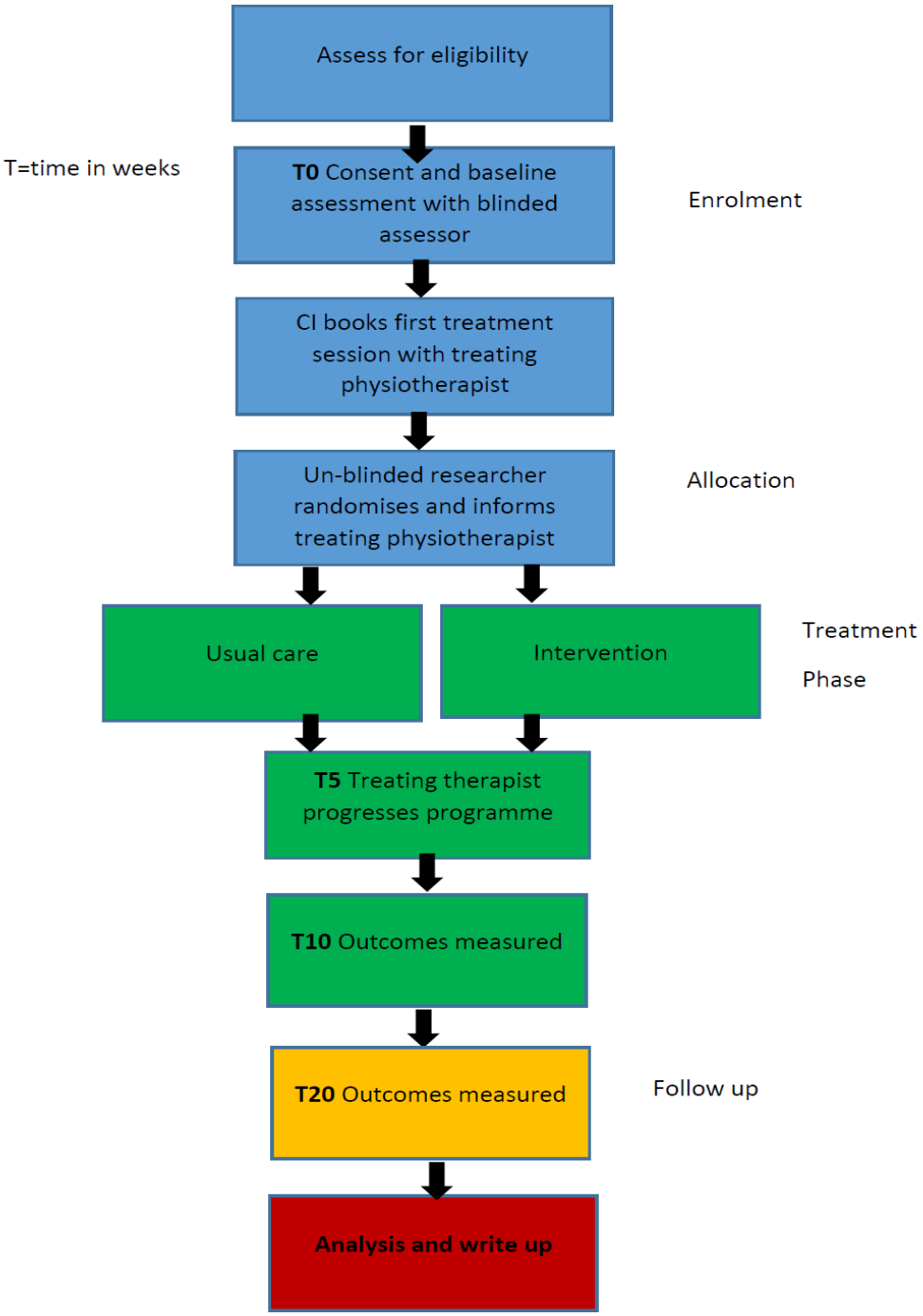

Figure 11 *Study flow diagram*

Table 16 *Objectives of the Feasibility Study*

Objective	Outcome
Focus	Methods
Feasibility of Definitive Trial	
Acceptability of the trial and intervention	Interviews of staff, parents and children
Can we recruit and retain participants?	Number of participants eligible Number recruited and randomised, date of recruitment recorded on study database Recruitment source Number of withdrawals. Number of participants lost to follow-up.
Effectiveness and acceptability of randomisation	Comparison of participant characteristics: severity, distribution of motor impairment, associated impairments at baseline Interviews
Effectiveness of concealment of allocation up to week 10	Number of times Chief Investigator correctly guessed treatment allocation
Concurrence with other surgical and medical interventions	Number of operations or procedures that target balance and walking during the intervention and follow up period.
Change in clinical outcome measures	Change in assessment scores of outcome measures
Assess appropriateness of outcome measures	Number and percentage of outcome measures completed at each time point Interviews
Feasibility of Intervention	
Adherence to treatment	Diary data frequency and duration of training
Acceptability of treatment intervention	Incidence of breakdown of equipment Number of times participants were unable to access equipment Participant view on acceptability of interventions by Interview
Cost of intervention and support needed to use it	Local physiotherapist record of staff time and grade used to support intervention. Travel costs of staff and families. Number and cost of repairs
Safety of intervention	Number and type of SAE and AE
Acceptability of Participation	
Acceptability of participation	Themes identified from interviews/photos

Table 17 Eligibility criteria

Inclusion Criteria
Diagnosis of CP GMFCS I-III.
Aged 4-18 years.
Leg weakness (≤4/5 on the MRC muscle strength rating scale) in at least 1 muscle group
Leg hypertonia (≥1 on the Tardieu scale fast stretch) in at least 1 muscle group
Ability to interact with a computer game using a mouse or joystick.
Exclusion Criteria
Selective dorsal rhizotomy or multi-level orthopaedic surgery within the last 12 months
Soft tissue surgery in lower limbs in last 6 months.
Botulinum toxin injections in the lower limbs within previous 3 months.
Training with the Happy Rehab™ in the last 4 months.

3.3.4.3 Intervention

Four devices will be available and situated in special schools, CDCs or the child's home. The child's physiotherapist will be trained to set up the device targeting exercises to improve range of movement, contracture and muscle weakness. This may include active-assisted hip, knee or ankle movements within specified ranges of movement or side-to-side and forward and back weight transfer. The treating physiotherapist will personalise the exercise programme based upon a standardised assessment, including a discussion with the child and their guardian about their goals and aims of any intervention.

The child will use a pseudonym of their choice to log onto the games, and to maintain confidentiality. Children will play the games within the mid-range of muscle length to begin with so that the games are difficult but achievable. This will aid motivation and adherence. After five weeks, the child's physiotherapist will progress the games by requiring the muscles to work in the inner and outer ranges of movement and/or against increased resistance. Training will build up to 20 minutes per day, 3 days a week over a 2-week period, with progression to a 30-minute programme per day, 3 days a week after five weeks. The child will train with supervision from the child's therapist, teaching assistant, parent or carer. The children will follow a series of games following a 2-minute warm up of continuous passive movement. The interactive trainer records the

training session (duration, games performed games outcomes) to provide a description of the parameters of training.

The control group will receive a usual care physiotherapy programme individualised for each child lasting 20-30 minutes.

Collaborative goal setting combined with an e-diary will allow the children and their parents/carers from both arms of the study to monitor progress over time and record their satisfaction with their exercise programme. The research team will assess fidelity through e-diaries, recording of exercise parameters via the interactive trainer and by observing ten exercise sessions and completing a fidelity checklist, to ensure the intervention follows protocol.

3.3.4.4 Study Procedures

Site set up

Recruitment will take place sequentially in each CDC area in order to ensure that the limited number of training devices are issued in the most efficient way. In preparation for recruitment, the research team will visit each site to familiarise physiotherapists with the eligibility criteria and trial procedures.

Recruitment

The physiotherapist will approach children and families on their caseload and give an information pack. Adverts for the study will be placed in clinic rooms, and on parent forums and social media. Potential participants who respond to the invitation will be screened for suitability using a telephone questionnaire to check diagnosis, age, GMFCS level and ability to play a game using a mouse or joystick. Eligible families will be approached for consent to be recruited to the qualitative study at baseline using a purposive sampling framework.

Potential participants who do not wish to take part in the study or withdraw from the study will be invited to undertake a short (less than five minutes) telephone interview to help understand any barriers and facilitators to participating in the trial, to aid in future recruitment. A separate information sheet will be available for these interviews.

3.3.4.5 Data Collection

Baseline Visit

Written informed consent and assent will be recorded prior to the child and parent undertaking the first baseline measurement session. The following data will be collected at the first visit:

- GMFCS level
- Date of birth
- Medical and surgical history
- Height, weight, pelvic depth
- Frequency and location of usual physiotherapy
- Other sports and social activities

3.3.4.6 Outcome measures

The following assessments will be carried out at weeks 0 and 10, and those indicated with * at 20 weeks follow up. The physical assessments will take 70 minutes followed by goal setting. Qualitative semi-structured interviews will take place after completion of the 10-week training.

Primary Outcomes

- *Medio-lateral motion of the centre of mass estimate [30, 175]
- *Pediatric Balance Scale [77]

Secondary Outcomes

- *Walking kinematics*
- *Muscle strength of quadriceps, hamstrings, and gastrocnemius and hip abductors using a handheld dynamometer (three measurements).
- *Passive range of movement and modified Tardieu scale [176] of quadriceps, hamstrings, gastrocnemius and hip adductors using goniometer (three measurements).
- COPM- Canadian Occupational Performance Measure [177]
- CHU-9D- Paediatric Quality of Life measure [178]

3.3.4.7 Blinding

This will be a single blinded RCT. The assessor will be blinded to allocation while carrying out the assessments at baseline and week 10. It will not be possible to for the assessor to remain blinded to group allocation for the 12 participants taking part in qualitative interviews occurring at week 11. However, the assessor will remain blinded to group allocation at week 20s for the remaining participants who are not undertaking the interviews.

3.3.4.8 Randomisation

Participants will be randomly allocated at a ratio of 1:1 and will be minimised by age (above or below 9 years) and by GMFCS level (level I and II versus level III). This is because acquisition of gross motor ability peaks by age 9, and children above that age plateau or may decline in motor skills [126]. The minimisation sequence and randomised allocations will be computer-generated in conjunction with an independent statistician. The blinded assessor will enter the details required for randomisation into the study website and book the participant's first appointment with the treating therapist.

An email will be generated by the study website to inform the local treating physiotherapist of the participant's allocated group. The treating physiotherapist will reveal group allocation to the participant at the first session. An un-blinded researcher will arrange for the interactive trainer to be transported to the site where the child usually does their physiotherapy e.g. school, CDC, home.

3.3.4.9 Qualitative Assessment

This qualitative study uses novel ways of data collection with the children including semi-structured e-diaries using electronic tablet devices and photo-elicitation interviews. Semi-structured interviews will be undertaken with parents/carers and physiotherapists. Triangulation of the e-diaries and interviews will be used to provide credibility, ensuring that the understanding of the full scope of the experiences related to participating in the trial is as complete as possible from the perspectives of the children, parents and physiotherapists. Twelve parent-child dyads will be recruited (30% of the total sample of the feasibility study). Four physiotherapists, who have delivered the intervention and control treatments in different settings, will be interviewed. Sampling of up to eight parents who declined or withdrew their child from the study will be undertaken.

3.3.4.10 Statistical Analysis Plan

A statistical analysis plan (SAP) will be drafted prior to the final database lock; the SAP will be agreed with the trial steering committee (TSC) in the absence of a data monitoring committee. A CONSORT diagram will be used to present descriptive data on screening, enrolment, intervention allocation, follow-up and assessment.

Completion rates of the intervention and outcomes collected at each time point will be reported with confidence intervals. All analyses and data summaries will be conducted on the intention-to-treat (ITT) population, defined as all participants randomised regardless of non-compliance with the protocol or withdrawal from the study. Participants will be analysed according to the intervention they received.

The baseline characteristics of those lost to follow up will be compared to those who complete the trial in order to identify any potential bias.

3.3.4.11 Proposed Primary and Secondary Outcome Analysis

The planned primary and secondary outcome measures will be reported at each time point using descriptive statistics. As this is a feasibility trial, it is not appropriate to perform a hypothesis test between-group treatment effects [179]. Instead, the difference between allocated groups of the follow-up minus baseline score will be estimated with confidence intervals.

A sample size estimate for a definitive trial will be undertaken for the proposed primary outcome. Estimation of the standard deviation, correlation between baseline and follow-up measures and a clinically meaningful difference will be used in the power calculation.

3.3.4.12 Progression criteria

This is determined in advance of recruitment will include minimum recruitment and retention rates (~70%) and a 90% completion rate of outcome measures. Failure to achieve these will indicate that a full trial is not feasible unless our qualitative study indicates clear means by which the rates may be improved. A recommendation list will be generated to enable refinement of the subsequent RCT protocol.

3.3.4.13 Qualitative Analysis and Data Synthesis

Qualitative data will be analysed using thematic analysis. Results from all aspects of the quantitative and qualitative data will be triangulated and synthesised and will be used to determine the suitability of the protocol for incorporation into the main RCT.

3.3.4.14 Public and Patient Involvement and Engagement

Families and physiotherapists have been consulted on trial design, with a particular focus on the two assessment visits. Documents including the protocol, adverts and patient information sheets were reviewed by an expert parent and a teenager with cerebral palsy and altered to make the information more accessible. Emerging themes from the qualitative analysis will be checked and informed by an invited group of children, parents and physiotherapists with relevant experience.

3.3.4.15 Data Collection and Management

Trial data collected will be recorded on a paper copy of a trial-specific Case Report Form (CRF) and will be considered source data. The blinded assessor will complete the CRFs for all participants. Completeness of data will be maximised by checking all forms at each assessment to ensure there are no missing items. Automatically generated prompts will be sent by email to encourage the participants to return their diaries, should they fail to do so within two weeks of the due date. Double-entered data will be compared for discrepancies using a stored procedure, and discrepant data will be verified using the original paper data forms. Before database lock, a proportion of original paper records will be checked against the database to ensure accuracy of the final dataset.

Confidentiality will be maintained by allocating a participant number to all CRFs and keeping the securely codes stored separately. Audio-recorded interviews will be transcribed and anonymised as soon as practicable. Original recordings will be held securely as an encrypted file on the University of Plymouth server, until completion of the qualitative data analysis process, then deleted.

Data will be collected and stored in accordance with the Data Protection Act 1998/General Data Protection Regulation 2018. Data generated from this trial will be available for inspection on request by the participating research team, University of Plymouth representatives, the REC, local R&D Departments, and the regulatory authorities.

3.3.4.16 Sample Size

As this study is a feasibility trial, it is not appropriate to use a sample size calculation based on considerations of power for detecting between group differences [179]. The feasibility aims are to provide robust estimates of recruitment rate and follow up as well

as estimates of the variability of the outcome measures, which will in turn inform sample size calculations for a full RCT.

A sample size of 40 participants will allow the overall recruitment rate to be estimated. It is anticipated that follow-up of a minimum of 12 participants in each of the intervention and usual care groups would provide sufficient data to inform indicative sample size calculations for the definitive main trial. An estimated recruitment rate of three to four children per month over a 12-month period has been calculated based on population and previous experience.

3.3.4.17 Adverse Events

The risks of taking part in this trial have been assessed to be low. Three adverse events (AEs) that require reporting include aches and pains in the leg muscles following training that last over 1 hour or require pain relief, injury related to the training, and fatigue lasting more than 1 day following training. AEs will be recorded via the online diary. Recorded AEs and serious adverse events (SAEs) will be presented to the monthly trial management group meeting for review.

3.3.4.18 Roles and Responsibilities

The roles and responsibilities are shown in Table 18. The trial management group TMG consists of R Rapson's supervisory team Prof Jos Latour, Prof Bernie Carter, Prof Jonathan Marsden, Rachel Rapson, CTU Trial Manager (Dr Wendy Ingram), CTU Data Manager (Laura Cocking) and trial statistician (Dr Kara Stevens). The trial steering committee consists of an independent chairperson, statistician, PPI representatives, Sponsors representative and local Research and Development manager.

3.3.4.19 Ethics and Dissemination

The child's assent and parental consent for their child's participation will be sought at the start of the study. Rests will be offered during the measurement sessions, which will be conducted at the child's pace. The child and family's involvement is voluntary and they will be reminded that they can refuse any part of the study, or withdraw at any time without consequence to their treatment. This study has approval from North of Scotland Research Ethics Committee (20/NS/0018) and the respective NHS Research and Development departments.

Table 18 *Roles and responsibilities of protocol contributors*

Chief Investigator	Rachel Rapson Peninsula Allied Health Centre University of Plymouth PL6 8BH rachel.rapson@plymouth.ac.uk
Trial Co-ordinator	Jonathan Marsden Peninsula Allied Health Centre University of Plymouth PL6 8BH Jonathan.marsden@plymouth.ac.uk
Sponsor	Sarah.C.Jones University Sponsor Representative Research and innovation Drake Circus Plymouth PL6 8AA Plymouth.sponsor@plymouth.ac.uk
Funder(s)	National Institute of Health Research
Clinical Trials Unit	Wendy Ingram Clinical Trials Manager Peninsula Clinical Trials Unit University of Plymouth Wendy.ingram@plymouth.ac.uk
Data Manager	Laura Cocking Senior Data Manager Peninsula Clinical Trials Unit University of Plymouth Laura.cocking@plymouth.ac.uk
Trial Statistician	Kara Stevens Research Fellow in Medical Statistics, University of Plymouth Kara.stevens@plymouth.ac.uk

The University of Plymouth research team will own the data arising from the trial. On completion of the trial, the data will be analysed and tabulated, and a final trial report prepared. Study findings will be published in peer reviewed academic journals and presented at national and international conferences. NIHR funding will be acknowledged within the publications. The outcomes of the trial will be shared with participants using a lay summary. Anonymised participant level data set will be

available 1 year after the end of the trial via the Rehabilitation Research Group (University of Plymouth) website.

3.3.4.20 Discussion

This study sets out to explore the feasibility of conducting a trial using a complex intervention in a variety of community settings. Using the proposed protocol, we will explore barriers and facilitators to running the trial. The protocol sets out a model of loaning the training device for an intensive ten-week intervention. We will be collecting initial data to indicate cost of the device, transport, repairs and maintenance. We anticipate that the logistics of transporting the devices within the community may prove difficult using existing infrastructures. We will be able to test these procedures to gain realistic timescales for a full trial.

We plan to include children with a range of cognitive, sensory and motor skills and we will examine if our primary outcome will capture change across all participants. By engaging in qualitative interviews, we will be able to determine the children and their parents' experiences of and perspectives on both the intervention and proposed outcomes, gaining important information whether these were acceptable or if outcomes were too difficult or took long. We will be able to examine their perspectives on any impact that the location (school, home or clinics) has on participation, as well the wider impact on their levels of participation and ability to manage their condition.

The main limitation of the trial is the lack of power to determine a significant difference in outcome measures. However, this feasibility study is an important step towards designing a full trial to test the efficacy and economic benefit of using a novel interactive exercise trainer to improve walking and balance in children with cerebral palsy.

Trial registration

ISRCTN80878394

http://isrctn.com/ISRCTN80878394

Protocol versions

ACCEPT Protocol version 1 10.01.2020

ACCEPT Protocol version 2 11.06.2020

ACCEPT Protocol version 3 09.07.2021

Acknowledgements to all the participants

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Contributorship statement

Authors RR, JM, JL, BC, KS, WI, LC contributed to the design, writing, drafting of the protocol. The roles and responsibilities of the authors are detailed in Table 9. All authors gave final approval to the publication of this protocol.

Declaration of interests

The authors declare no competing interests.

3.3.5 Quantitative results of the feasibility RCT

The results were analysed using with reference to the Statistical Analysis Plan (SAP) version 2 (18.08.2022) as approved by the Trial Steering Committee before data lock (Appendix 6). A table of the feasibility outcomes can be seen at the end of this section in Table 43.

3.3.5.1 Recruitment and retention

Recruitment was delayed due to COVID-19 pandemic and so the recruitment period was much shorter than planned. Sixteen children were recruited and 15 randomised. The recruitment rate was 1.2 children per month and this was limited by the number and size of the devices available per site (Figure 12). Table 19 shows the screening, recruitment and retention of participants in the study. Recruitment took place initially at site 1 with 13 children randomised. The number of potential eligible children on the caseload at (Torbay and South Devon NHS Foundation Trust) TSDFT were 47 children, GMFCS I-III. The second site was briefly opened at University Hospitals Plymouth Trust (UHPT), where two children were recruited before the trial was closed to recruitment.

***Figure** 12 Recruitment rate for the ACCEPT study between March 2021 and January 2022*

The CONSORT diagram (*Figure 13*) shows the retention and reason for drop out during the study. Reasons for declining included being too busy and one family not wishing to stop usual care. In the intervention group, one participant did not receive the intervention, as the large Happy Rehab would not fit through an inner doorway. They declined to use the Happy Rehab in the Physiotherapy department due to the required travel and time commitment.

Table 19 *Number of the total children recruited and retained by site*

	TSDFT	UHPT	Total
	n	n	n
Assessed for eligibility	21	3	24
PIS packs given out	21	2	23
Did not meet criteria during caseload screening	1	1	2
Declined to participate	3	0	3
Consented but became ineligible	1	0	1
Participants randomised	13	2	15
Participants allocated to control	6	1	7
Participants allocated to intervention	7	1	8
Participants who received the allocated intervention	5	1	6
Participants who did not receive the allocated intervention	2	0	2
Participants lost to follow up	1	0	1
Participants who discontinued the intervention	0	0	0
Mean (SD) time taken from identification to consent (days)	33.7 (30.6)	50 (49.5)	35.6 (31.7)

Key- Number (n), standard deviation (SD)

One child was randomised to the intervention, but was then offered botulinum toxin in their gastrocnemius, and so became ineligible for the study. They were offered the opportunity to restart the trial three months after the treatment, in December 2021. At that point, the child's mother said that they could not accommodate the Happy Rehab in the lounge as well as a Christmas tree and decided to withdraw. One participant in the intervention group was lost to follow up due to a change in family circumstances and another completed the 10-week assessment but was unable to attend the 20-week assessment before the trial close down date. Of the usual care group, one child became ineligible as their diagnosis changed from cerebral palsy. Another child was followed up at 10 weeks but did not respond to invitations to 20-week assessment or the interviews.

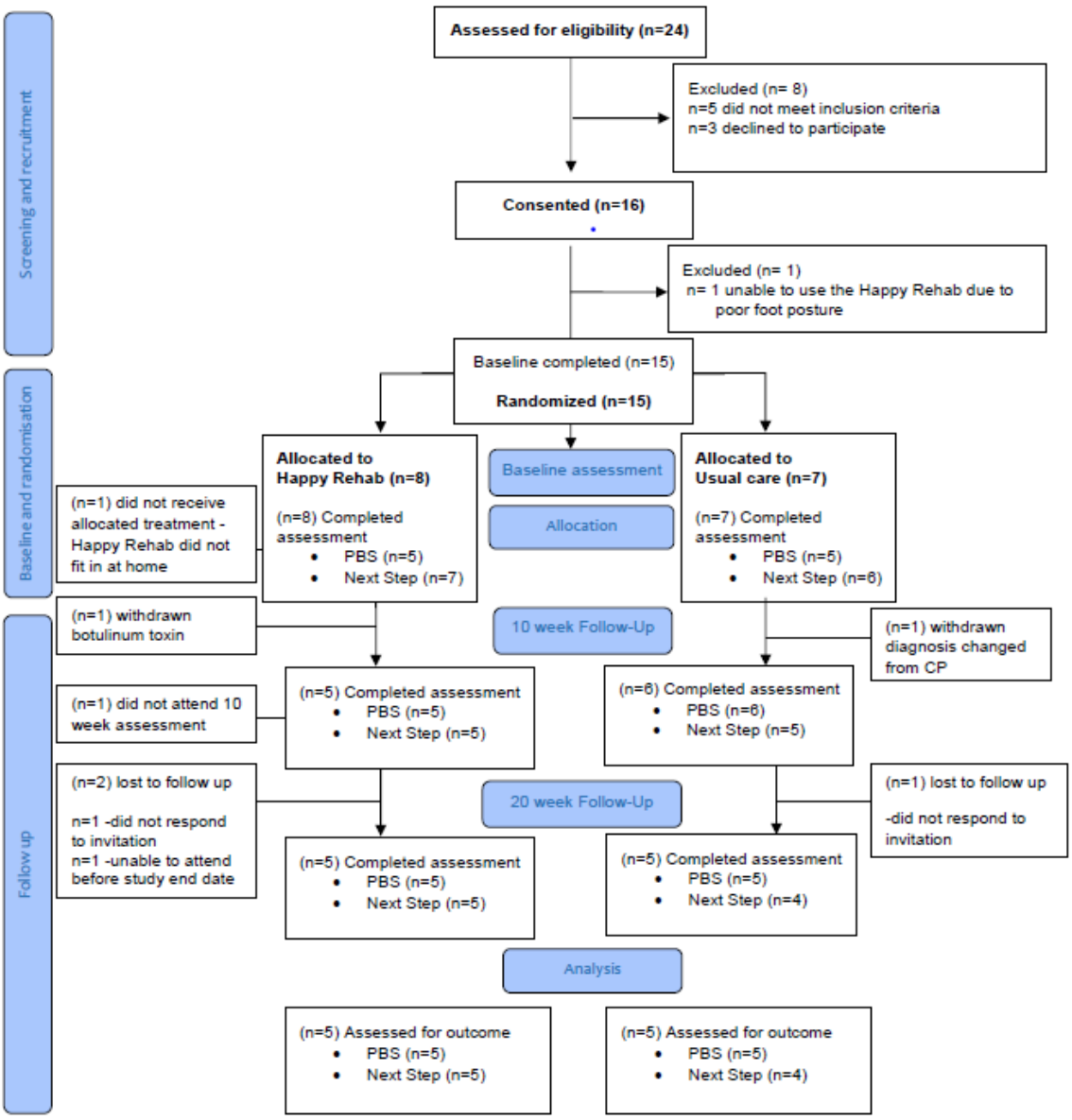


Figure 13- Consort diagram for the ACCEPT study.

3.3.5.2 Participant demographics and clinical characteristics

The participant demographics are shown in Table 20 below. The two groups were similar in gender, hip migration and functional mobility. The usual care group consisted of children with lower GMFCS classification (higher functional ability) and more children with bilateral CP. The intervention group were older and consequently taller and heavier than the usual care group, and had more children with unilateral CP.

Table 20 *Participant Demographics and group allocation*

Patient characteristics	Total n=15	Happy Rehab n=8	Usual Care n=7	Withdrawn n=2
Age (years) Mean (SD)	10 (3.3)	11.0 (3.8)	9.3 (3.1)	8.7 (1.3)
GMFCS level n=1:2:3	6:8:1	1:5:1	3:3:0	2:0:0
Distribution of Impairment Bilateral (n=)	5	1	4	0
Distribution of Impairment Unilateral (n=) right: left	4:6	3:4	1:2	1:1
Gender (n=) m:f	9:6	5:3	4:3	0:2
Height (cm) Mean (SD)	135 (18)	136 (19)	120 (16)	129 (18)
Weight (kg) Mean (SD)	38.9 (17.2)	40.9 (18.1)	33.0 (15.1)	30.2 (12.4)
Hip migration % right: left Mean (SD)	14: 11 (7) (5)	15:11 (8) (5)	11:10 (4) (6)	-
Functional Mobility Scale 5m (0-6) median (IQR)	6 (0)	6 (1)	6 (0)	6 (0)
Functional Mobility Scale 50m (0-6) median (IQR)	6 (0)	6 (1)	6 (0)	6 (0)
Functional Mobility Scale 500m (0-6) median (IQR)	6 (2.5)	5 (2.5)	6 (0)	6 (0)

Key- Number (n), standard deviation (SD), Gross Motor Function Classification Scale (GMFCS), centimetre (cm), kilogram (kg), inter-quartile range (IQR), metre (m), percentage (%)

3.3.5.3 Effectiveness of minimisation and randomisation

Participants were randomised at a ratio of 1:1 using two minimisation criteria: by age (above or below 9 years) and by GMFCS level (level I and II versus level III). Only one child with GMFCS level III was recruited, which may have reduced the effectiveness of the minimisation process. This may explain why there are several differences between the groups.

There was one instance of unblinding where the child told the assessor about their allocation during the assessment. Participants were asked how much activity and physiotherapy exercise they carried out each week.

Table 21 shows that overall children carried out a mean 3.3 (SD 2.5) hours physical activity per week and spent a mean of 2 (SD 2) hours of physiotherapy programmes per week. The intervention group did more exercise and the

control group did more physiotherapy exercises. Participants had a range of additional diagnoses or impairments, which can be seen in Table 22. Notably nine out of fifteen children complained of fatigue, with a higher number being in the intervention group. Four children had a learning disability and two had autistic spectrum disorder. The two children with communication needs used Makaton to support their communication.

Four children (two in each group) recruited to the study had undergone selective dorsal rhizotomy, more than two years previously. Only children in the intervention group had had other surgical interventions. These included botulinum toxin in gastrocnemius and hip adductors and gastrocnemius lengthening (Table 23). In the intervention group, there were two children with epilepsy who took regular medication for seizure management and one child who had heart failure following heart surgery in infancy requiring ACE inhibitors to manage this (Table 24).

Twelve out of the fifteen participants wore ankle-foot orthoses (AFOs), and four used wheeled walkers. Participants using these pieces of equipment were evenly distributed between groups (Table 25).

Two children had concurrent treatment during the intervention phase, which had potential confounding effects. One child had botulinum toxin in her calf muscles, and the second child had a change in diagnosis from CP, which made her ineligible for the study. Both children were withdrawn from the study at the point that these events occurred.

Table 21 *Patient characteristics – Physical activity and physiotherapy*

Participant Characteristic	All Participants n=15	Happy Rehab n=7	Usual Care n=6	Withdraw n=2
Usual amount of sport or physical activity per week (hours) Mean(SD)	3.3 (2.5)	3.5 (2.7)	2.9 (2.3)	2.9 (2.1)
Time spent carrying out physio programme per week (hours) Mean(SD)	2.0 (2.0)	1.6 (0.6)	2.5 (3.1)	1.5 (2.1)

Key- Number (n), standard deviation (SD)

Table 22 *Patient characteristics – Medical history at baseline*

Number of medical conditions	All Participants n=15	Happy Rehab n=7	Usual Care n=6	Withdrawn n=2
Fatigue	9	5	2	2
Congenital /genetic condition	2	1	1	0
Epilepsy	2	0	2	0
Learning Disability	4	2	2	0
Autistic Spectrum Disorder	2	1	1	0
Visual Impairment	1	0	1	0
Communication needs	2	2	0	0
Asthma	3	2	1	0
Constipation	1	0	1	0
sensory	3	3	0	0
Leg cramps	1	1	0	0
Heart failure	1	1	0	0

Key- Number (n)

Table 23 *Patient characteristics - Surgical history*

Number of previous surgical interventions	All Participants n=15	Happy Rehab n=7	Usual Care n=6	Withdrawn n=2
Selective dorsal rhizotomy	4	2	2	0
Botulinum toxin gastrocnemius	4	4	0	0
Botulinum toxin hamstrings	0	0	0	0
Botulinum toxin hip adductors	2	2	0	0
Botulinum toxin tibialis posterior	0	0	0	0
Femoral de-rotation osteotomy	0	0	0	0
Tibial de-rotation osteotomy	0	0	0	0
Gastrocnemius lengthening	2	2	0	0
Hamstring lengthening	0	0	0	0
Adductor lengthening	0	0	0	0
Other	2	2	0	0
Deep Brain Stimulator	0	0	0	0

Key- Number (n)

Table 24 *Patient characteristics - Medications*

Number of medication or class of drug	All Participants n=15	Happy Rehab n=7	Usual Care n=6	Withdrawn n=2
Constipation medication	2	0	2	0
Bronchodilators	3	2	1	0
Anti-epileptics	2	0	2	0
Epilepsy rescue medication	1	0	1	0
Baclofen	0	0	0	0
Melatonin	0	0	0	0
Anti-spasmodic	0	0	0	0
ACE inhibitor	1	1	0	0

Key- Number (n)

Table 25 *Patient characteristics – Orthotics and walking aids*

Number and type of orthotics and walking aids	All Participants n=15	Happy Rehab n=7	Usual Care n=6	Withdraw n=2
Insoles	2	1	1	0
AFOs	12	5	5	2
Dynamic AFOS	1	1	0	0
Ground Reaction AFOs	0	0	0	0
Dynamic Elasticated Fabric Orthosis	0	0	0	0
Functional Electrical Stimulation	0	0	0	0
Wheeled walker	4	2	2	0
1 crutch/stick	0	0	0	0
2 crutches/sticks	1	1	0	0
Manual wheelchair	0	0	0	0
Powered wheelchair	0	0	0	0

Key- Number (n)

3.3.5.4 Descriptive analysis of the primary outcome measures

3.3.5.4.1 Pediatric Balance Scale

The Pediatric Balance Scale (PBS) consists of 14 items scoring 0-4 on an ordinal scale, with a maximum possible score of 56 [77] (see Appendix 7). All participants had maximum scores on the first four items of the PBS. One participant in the Happy Rehab group reached the ceiling of the PBS at 10 weeks. Two participants from the control group reached the maximum PBS

score at baseline. One child with autistic spectrum disorder (ASD) was not able to complete all tasks and so their results were excluded. Change in item scores were seen over the last eight items of PBS.

At baseline, children in the usual care group had a higher median score on the PBS (median 52.7 (IQR 4.2)) compared to the Happy Rehab group (median 50.7 (IQR 2.8)). Both groups had higher median scores at 10 weeks (indicating an improvement in balance); with the Happy Rehab group sustaining a two-point improvement at 20 weeks and the usual care group having a lower median score at 20 weeks (Table 26).

This Minimal Detectable Difference (MDD) needed to detect a true change in the PBS is reported as 1.27 points [180]. A sub analysis of participants who completed the PBS at all three time points showed individual improvements by median 2.3 (1.5) points in the Happy Rehab group at 10 weeks and 1.3 (IQR 2.1) at 20 weeks (Table 26). The participants in the control group showed an improvement of 1.7 (IQR 1.25) points at 10 weeks but a median 0 (IQR 0.38) at 20 weeks compared to baseline scores. Four participants in the Happy Rehab group achieved an increase in score above the MDD at 10 weeks compared to baseline and two participants at 20 weeks. In the control group three participants achieved increase in score above the MDD at 10 weeks compared to baseline and one participant at 20 weeks. Figure 14 shows the group median and range in PBS scores at the different time points.

Table 26 *Median Pediatric Balance Scale score at baseline, 10 weeks, 20 weeks, and median change in scores in the intervention and control group.*

Outcome	Baseline		10 weeks		20 weeks	
	Happy Rehab n=7/8	Usual Care n=6/7	Happy Rehab n=5/8	Usual Care n=6/7	Happy Rehab n=5/8	Usual Care n=5/7
Median (IQR) Pediatric Balance Scale	50.7 (2.8)	52.7 (4.2)	52.7 (4.2)	53.5 (6.3)	52.0 (7.7)	49.0 (5.5)
Median (IQR) change from baseline			2.7* (1)	0.7 (2.4)	2.0 (2.5)	0 (1)
Number of participants who completed all three assessments			n=4/8	n=5/7	n=4/8	n=5/7
Median (IQR) change from baseline score			2.3* (1.5)	1.7* (1.25)	1.3* (2.1)	0 (0.38)

Key- number (n), interquartile range (IQR), percentage (%), score greater than minimal detectable difference (*)

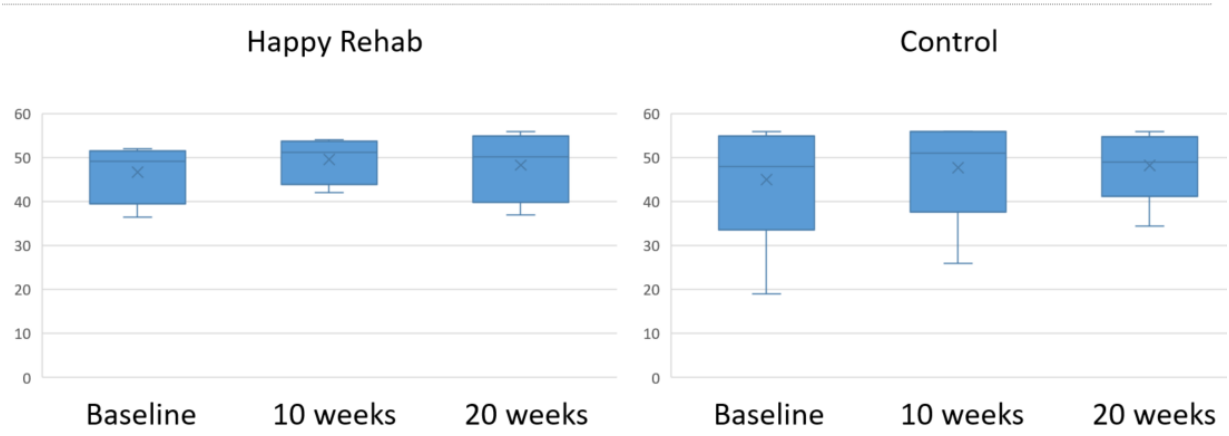


Figure 14 *Boxplots showing the group median and range in Pediatric Balance Scale at baseline 10 weeks and 20 weeks.*

3.3.5.4.2 Next Step measures of balance

Movement of the centre of pressure (COP) and motion of the centre of mass (COM) was estimated via pelvic markers while children stepped to laterally and medially placed targets with either leg. The right leg stepped to targets 1 (lateral) and 2 (medial) and the left leg to targets 3 (medial) and 4 (lateral). The Next step mean values at each time point are shown per target in Table 27. Movements forward and to the right are indicated by positive numbers and movements to the left or backwards are indicated by negative numbers.

The MDD for peak medio-lateral (ML) COP ranges from 23.7-29.6mm and peak ML velocity COM 41.0-61.8 mm/s depending on target. Results which exceed the MDD are indicated with a *. Overall, there was a modulation of the ML COM velocity and peak COP with target position. The ML COM velocity and peak COP was larger for medially placed targets (2 and 3).

The Next Step summary scores are shown in Table 28. Both groups had smaller anticipatory postural adjustments as measured by ML COP than the cohort measured in the Next Step observational study in Part II (mean 37.3 mm SD 20.5). Both groups showed an increase in ML COP grand average scores at 10 weeks with the Happy Rehab group increasing further at 20 weeks. The usual care group decreased to below baseline at 20 weeks. Of participants who completed all three assessments, both groups showed improved change scores at 10 weeks compared to baseline. The Happy Rehab group improved further at 20 weeks compared to baseline. The usual care group decreased ML COP grand average score compared to baseline at 20 weeks.

***Table 27**- Comparison of mean values of Next Step at baseline, 10 weeks and 20 weeks between the intervention and control group. The left footsteps to targets 1 (lateral) and 2 (medial), while the right foot steps to 3 (medial) and 4 (lateral) targets. Movements to the left and posterior are shown as negative numbers*

Outcome		Baseline		10 weeks		20 weeks	
	Target	Happy Rehab n=7/8	Usual Care n=6/7	Happy Rehab n=5/8	Usual Care n=5/7	Happy Rehab n=5/8	Usual Care n=4/7
Peak velocity ML COM Per target (mm/s) Mean (SD)	1	50.1 (34.1)	79.7 (49.4)	57.4 (25.1)	73.2 (40.9)	38.8 (27.3)	59.1 (31.5)
	2	120.9 (44.6)	124.3 (57.2)	108.1 (90.0)	128.4 (64.4)	113.3 (56.3)	95.0 (56.7)
	3	-154.0 (99.4)	-135.8 (68.4)	-87.1* (41.5)	-105.2 (47.5)	-155.7 (53.1)	-93.1 (60.4)
	4	-60.3 (36.6)	-81.1 (53.4)	-44.1 (35.4)	-95.6 (77.5)	-43.3 (50.2)	-54.3 (38.7)
Peak ML COP Per target (mm) Mean (SD)	1	-8.5 (53.1)	-19.9 (27.7)	-21.9 (23.3)	-20.3 (17.4)	-17.8 (29.7)	-26.8 (10.9)
	2	-29.2 (22.6)	-10.3 (31.4)	-31.7 (29.3)	-12.5 (39.7)	-28.6 (44.4)	-18.0 (33.8)
	3	32.3 (22.5)	23.2 (39.7)	24.0 (24.0)	34.9 (14.4)	66.7* (31.6)	25.3 (14.2)
	4	28.1 (36.0)	26.4 (13.8)	18.6 (23.8)	38.9 (24.6)	34.6 (33.2)	0.7 (38.4)
Peak AP COP Per target (mm) Mean (SD)	1	-1.1 (40.5)	-12.0 (5.8)	-14.9 (11.5)	-10.1 (4.1)	-7.7 (5.1)	-6.4 (10.4)
	2	-13.8 (10.9)	-7.20 (9.8)	-13.7 (11.7)	-7.3 (3.6)	-15.0 (9.7)	-11.6 (4.0)
	3	-13.4 (8.3)	-11.6 (12.5)	-9.3 (10.3)	-7.7 (3.6)	-12.8 (8.3)	-10.8 (3.1)
	4	-3.2 (26.9)	-7.35 (5.21)	-8.1 (7.6)	-8.1 (3.2)	-13.7 (8.6)	-8.9 (15.2)

Key- Medio-lateral (ML), antero-posterior (AP). Centre of mass (COM), centre of pressure (COP), standard deviation (SD), millimetres (mm)

3.3.5.5 Descriptive analysis of the secondary outcome measures

3.3.5.5.1 Gait kinematics

Table 29 shows the mean and SD of gait kinematic outcomes. Overall, the usual care group had more knee flexion in midstance, more leg swing (knee peak to trough) and more plantarflexion (indicated by negative numbers) at initial contact than the Happy Rehab group at baseline. Both groups had similar ankle angle at midstance at baseline.

Table 28 *Summary measures of grand average and symmetry of peak ML COP (mm)*

Outcome	Baseline		10 weeks		20 weeks	
	Happy Rehab n=7/8	Usual Care n=6/7	Happy Rehab n=5/8	Usual Care n=5/7	Happy Rehab n=5/8	Usual Care n=4/7
Mean (SD) Next step grand average peak ML COP (mm)	21.3 (32.4)	16.3 (24.5)	24.1 (19.4)	26.6 (15.1)	36.9 (22.6)	17.7 (22.6)
Number of participants who completed all three assessments			n=4/8	n=4/7	n=4/8	n=4/7
Mean (SD) grand average ML COP change from baseline score			7.4 (18.9)	10.4 (16.4)	14.7 (36.3)	2.1 (7.3)

Key- Medio-lateral (ML), centre of pressure (COP), standard deviation (SD), millimetres (mm)

3.3.5.5.2 Comparative knee angle at midstance

The mean knee flexion angle during gait was calculated in comparison to the knee flexion angle in quiet standing. This was greater in the usual care group (Right 7.4 (SD 7.1) and Left 4.0 (SD 3.4) degrees knee flexion) which indicated more crouched gait than the Happy Rehab group (Right mean 0.0 (SD 4.1) and Left 2.4 (SD 13.6) degrees knee flexion) at baseline. The Happy Rehab group showed greater knee extension in midstance of 0.9 (SD 5.4) degrees in the left leg at 10 weeks, but this was not sustained at 20 weeks. The usual care group showed 1.1 (SD 12.4) degree less knee flexion in midstance gain in the right leg at 10 weeks, but at 20 weeks, it was 10.4 degrees knee flexion in midstance.

3.3.5.5.3 Knee angle trough to peak

The knee angle trough to peak is a measure of leg swing during the gait cycle. At baseline the usual care group had a larger range of leg swing 50.6 (SD 6.8) and 52.3 (SD 9.1) degrees in the right and left leg respectively (Table 29). The Happy Rehab group had a mean range of knee movement in swing of 46.0 (18.5) and 46.8 (SD 17.1) degrees in the right and left leg respectively. The only improvement in leg swing was seen in the left leg of the Happy Rehab group at 20 weeks, with an increase to 48.7 (SD 19.4) degrees knee flexion.

3.3.5.5.4 Angle of ankle dorsiflexion at initial contact

The angle of dorsiflexion was measured at initial contact (IC) where a negative value indicates plantarflexion. Both groups, on average, made IC with the ankle in plantarflexion (Table 27). The usual care group had greater mean plantarflexion at IC mean -8.9 (SD7.6) and -7.1 (SD7.4) degrees for right and left legs respectively. In the Happy Rehab group this was mean -3.0 (SD 15.3)

and -6.3 (SD 10.1) degrees for right and left legs respectively (Table 27). There was improvement in the usual care group at 10 weeks only with a slight reduction in plantarflexion to -7.2 (SD 4.8) and -1.8 (4.0) degrees.

Table 29 *Comparison of mean values of gait kinematics at baseline, 10 weeks and 20 weeks between the intervention and control group.*

Outcome	Baseline				10 weeks				20 weeks			
	Happy Rehab n=7/8		Usual Care n=6/7		Happy Rehab n=5/8		Usual Care n=6/7		Happy Rehab n=5/8		Usual Care n=5/7	
leg	R	L	R	L	R	L	R	L	R	L	R	L
Knee angle midstance (°) Mean (SD)	0.0 (4.1)	2.4 (13.6)	7.4 (7.1)	4.0 (3.4)	10.4 (17.2)	-0.9 (5.4)	-1.1 (12.4)	4.8 (4.5)	5.2 (8.1)	8.3 (9.9)	10.4 (2.5)	10.3 (10.4)
Knee angle trough to peak (°) Mean (SD)	46.0 (18.5)	46.8 (17.1)	50.6 (6.8)	52.3 (9.1)	40.3 (7.3)	35.0 (13.3)	43.8 (7.4)	52.7 (9.1)	44.4 (6.2)	48.7 (19.4)	50.6 (2.9)	46.6 (12.9)
Ankle angle initial contact (°) Mean (SD)	-3.0 (15.3)	-6.3 (10.1)	-8.9 (7.6)	-7.1 (7.4)	-3.1 (13.1)	-13.0 (4.2)	-7.2 (4.8)	-1.8 (4.0)	-6.5 (10.3)	-7.5 (6.1)	-7.5 (0.60)	-8.6 (5.0)
Ankle angle midstance (°) Mean (SD)	5.7 (12.5)	2.4 (13.6)	2.8 (3.0)	4.0 (3.6)	8.7 (12.0)	-0.9 (5.4)	-3.1 (4.0)	4.8 (4.5)	1.5 (3.5)	-1.1 (5.6)	5.7 (6.8)	-0.8 (4.1)

Key- Number (n), standard deviation (SD), degree (°)

3.3.5.5.5 Comparative angle of ankle dorsiflexion at midstance

This angle was calculated in comparison to the ankle angle of each participant in quiet standing just before the gait trials. Normal angle of dorsiflexion in midstance would be 0. A negative number indicates plantarflexion. Table 29 shows that both groups have slight ankle dorsiflexion at midstance, which is a possible indication of mild crouch gait. The Happy Rehab group had mean ankle dorsiflexion at midstance of 5.7 (SD 12.5) on the right at baseline. This increased to 8.7 (SD 12.0) degrees at 10 weeks and decreased to 1.5 (SD 3.5) at 20 weeks. The left side was 2.4 (SD 13.6) at baseline decreasing to -0.9 (SD 5.4) at 10 weeks and -1.1 (SD 5.6) degrees at 20 weeks.

The usual care group stood with ankles relatively dorsiflexed at baseline. On the right by mean 2.8 (SD 3.0), increasing to 3.1 (SD 4.0) degrees at 10 weeks and further to 5.7 (SD 6.8) degrees at 20 weeks. On the left side, there was a mean

of 4.0 (SD 3.6) degrees at baseline, increasing to 4.8 (SD 4.5) degrees at 10 weeks and slight plantar flexion of -0.8 (SD 4.1) degrees at 20 weeks.

Table 30 shows the mean changes from baseline for individuals who completed all three assessments at weeks 10 and 20. However, the data set is very small for each of the gait kinematics scores either due to children declining the measure or technical difficulties obtaining the measures.

3.3.5.5.6 Measures of spasticity and range of passive movement measured using the Modified Tardieu Score (MTS)

Table 31 shows the mean values of the Modified Tardieu Score (MTS) at two speeds. R1 indicates the range of motion achieved during a fast. R2 indicates the range of motion achieved during a slow stretch, which measures muscle length. The hamstrings length was determined using the popliteal angle and so a larger number indicates shorter hamstring length. Table 32 shows the changes from baseline for the R1 and R2 for participants with complete data sets. The difference between R1 and R2 is considered to be a measure of spasticity [176]. Changes in spasticity scores are shown in Table 33.

Table 30 *Change in mean values of gait kinematics at 10 weeks and 20 for children who attended all three assessments.*

Number of participants in each group	**10 weeks**				**20 weeks**			
	Happy Rehab n=5		Usual Care n=6		Happy Rehab n=5		Usual Care n=5	
Leg	R	L	R	L	R	L	R	L
Mean (SD) change from baseline of knee angle at midstance (°)	(n=1) -10.3 (0.0)	(n=1) -25.3 (0.0)	(n=2) 1.7 (4.1)	(n=0)	(n=1) 2.4 (0.0)	(n=1) -11.5 (0.0)	(n=2) 5.2 (10.6)	(n=0)
Mean (SD) change from baseline of knee angle trough to peak (°)	(n=3) 41.7 (8.3)	(n=3) -5.4 (22.6)	(n=2) -12.3 (14.1)	(n=2) 10.7 (12.9)	(n=3) 41.3 (6.4)	(n=3) 47.3 (19.0)	(n=2) -1.6 (8.0)	(n=2) 44.0 (16.5)
Mean (SD) change from baseline of ankle angle at initial contact (°)	(n=1) -12.6 (0.0)	(n=1) -22.4 (0.0)	(n=2) 4.6 (1.1)	(n=2) 3.1 (3.1)	(n=1) -4.6 (0.0)	(n=1) -1.4 (0.0)	(n=2) 5.9 (6.5)	(n=2) -5.5 (8.0)
Mean (SD) change from baseline of ankle angle at midstance (°)	(n=1) -4.8 (0.0)	(n=1) -25.3 0.0)	(n=2) -5.7 (3.6)	(n=1) 3.9 (0.0)	(n=1) 6.8 (0.0)	(n=1) -11.5 (0.0)	(n=2) 1.3 (6.3)	(n=1) -6.6 (0.0)

Key- Standard deviation (SD), degrees (°)

Table 31 *Comparison of mean values of the Modified Tardieu Score at baseline, 10 weeks and 20 weeks between the intervention and control group.*

Outcome	Baseline		10 weeks		20 weeks	
	Happy Rehab n=8/8	Usual Care n=7/7	Happy Rehab n=5/8	Usual Care n=6/7	Happy Rehab n=5/8	Usual Care n=5/7
Right hamstrings (°) R1 Mean (SD)	52.7 (18.3)	42.1 (14.4)	61.6 (25.2)	45.4 (14.9)	41.8 (11.3)	48.3 (12.7)
Right hamstrings (°) R2 Mean (SD)	40.5 (13.9)	35.0 (7.8)	51.5 (33.8)	37.1 (8.8)	34.8 (11.5)	43.1 (9.9)
Left hamstrings (°) R1 Mean (SD)	54.0 (6.7)	50.3 (20.4)	64.3 (31.4)	40.8 (16.6)	50.2 (13.9)	43.0 (8.3)
Left hamstrings (°) R2 Mean (SD)	40.2 (10.0)	39.4 (13.1)	55.7 (38.1)	34.5 (9.9)	35.1 (6.0)	40.1 (6.5)
Right hip adductors (°) R1 Mean (SD)	32.3 (12.7)	40.5 (12.2)	34.5 (10.1)	45.6 (2.5)	40.7 (6.4)	38.3 (7.4)
Right hip adductors (°) R2 Mean (SD)	41.9 (10.9)	43.5 (10.3)	41.1 (17.9)	48.1 (7.3)	47.8 (7.3)	38.8 (6.3)
Left hip adductors (°) R1 Mean (SD)	32.5 (10.0)	40.0 (11.1)	35.7 (9.7)	46.9 (8.5)	36.3 (7.8)	40.0 (8.8)
Left hip adductors (°) R2 Mean (SD)	37.7 (7.1)	45.1 (9.4)	43.4 (11.5)	49.2 (4.3)	36.1 (6.3)	41.5 (10.2)
Right gastrocnemius (°) R1 Mean (SD)	-14.5 (3.8)	-9.8 (8.4)	-13.1 (5.3)	-8.8 (9.1)	-6.1 (6.6)	-10.5 (8.2)
Right gastrocnemius (°) R2 Mean (SD)	0.9 (5.0)	4.0 (7.1)	1.3 (4.4)	2.3 (5.6)	4.9 (4.1)	2.3 (5.5)
Left gastrocnemius (°) R1 Mean (SD)	-9.5 (9.1)	-8.9 (5.4)	-12.4 (11.2)	-13.7 (8.1)	-16.8 (15.1)	-9.3 (5.8)
Left gastrocnemius (°) R2 Mean (SD)	7.2 (10.5)	2.5 (8.3)	2.2 (7.6)	1.8 (3.1)	0.1 (11.2)	4.1 (5.2)
Right Duncan Ely (°) R1 Mean (SD)	126.9 (22.4)	140.6 (7.4)	131.9 (11.9)	131.8 (8.6)	135.0 (7.7)	141.4 (12.0)
Right Duncan Ely (°) R2 Mean (SD)	132.3 (7.8)	142.9 (7.2)	136.6 (11.1)	136.8 (9.0)	137.6 (8.8)	141.3 (13.1)
Left Duncan Ely (°) R1 Mean (SD)	131.3 (10.5)	141.4 (9.3)	133.4 (11.8)	135.2 (8.9)	130.7 (11.2)	138.1 (12.5)
Left Duncan Ely (°) R2 Mean (SD)	131.8 (10.1)	142.4 (10.7)	136.5 (8.0)	134.4 (11.1)	127.3 (9.5)	138.4 (9.9)

Key- Number (n), standard deviation (SD), degree (°), Modified Tardieu test fast speed (R1) and slow speed (R2)

Table 32 *Changes from baseline measures of Modified Tardieu Score at 10 and 20 weeks for children who undertook all three assessments*

Outcome	Change at 10 weeks		Change at 20 weeks	
	Happy Rehab n=4	Usual Care n=5	Happy Rehab n=4	Usual Care n=5
Right hamstrings (°) R1 Mean (SD)	9.0 (8.2)	1.5 (6.5)	-0.8 (8.3)	7.2 (5.3)
Right hamstrings (°) R2 Mean (SD)	2.1 (3.5)	0.8 (6.1)	0.1 (9.0)	8.3 (6.9)
Left hamstrings (°) R1 Mean (SD)	-2.4 (10.9)	-7.1 (7.8)	-3.9 (13.7)	-7.9 (20.9)
Left hamstrings (°) R2 Mean (SD)	0.5 (11.7)	-0.8 (7.6)	-3.8 (8.0)	1.9 (8.3)
Right hip adductors (°) R1 Mean (SD)	0.4 (9.0)	4.9 (15.3)	2.1 (12.6)	-2.2 (8.1)
Right hip adductors (°) R2 Mean (SD)	6.8 (9.4)	4.8 (6.8)	6.5 (11.1)	-4.2 (10.9)
Left hip adductors (°) R1 Mean (SD)	4.0 (15.2)	4.7 (14.3)	3.3 (4.4)	-1.4 (12.9)
Left hip adductors (°) R2 Mean (SD)	7.9 (9.5)	2.3 (12.0)	-4.5 (4.4)	-5.6 (14.3)
Right gastrocnemius (°) R1 Mean (SD)	0.3 (4.0)	4.5 (8.2)	6.8 (4.9)	-0.5 (4.9)
Right gastrocnemius (°) R2 Mean (SD)	1.1 (6.8)	-1.3 (6.8)	2.8 (11.4)	-2.6 (4.6)
Left gastrocnemius (°) R1 Mean (SD)	0.2 (2.6)	-4.1 (12.5)	-2.4 (4.5)	1.1 (3.5)
Left gastrocnemius (°) R2 Mean (SD)	0.8 (2.5)	-1.7 (6.5)	-2.3 (5.2)	1.5 (3.9)
Right Duncan Ely (°) R1 Mean (SD)	-3.3 (13.0)	-8.4 (7.9)	-1.8 (7.3)	0.8 (9.3)
Right Duncan Ely (°) R2 Mean (SD)	2.3 (11.0)	-5.8 (4.7)	3.9 (8.3)	-2.6 (13.1)
Left Duncan Ely (°) R1 Mean (SD)	2.0 (6.4)	-3.9 (12.2)	-0.5 (4.3)	-2.1 (14.1)
Left Duncan Ely (°) R2 Mean (SD)	2.3 (2.4)	-5.1 (11.5)	-5.8 (5.3)	-2.0 (7.2)

Key- Number (n), standard deviation (SD), degree (°), Modified Tardieu test fast speed (R1) and slow speed (R2)

Overall, the increase in passive range of motion (R2) was in favour of the Happy Rehab group by an average gain in of 3.1° at 10 weeks and 3.75° at 20 weeks. The change in passive range of motion was greater in the Happy Rehab group compared to the usual care group for 6 out of 8 muscles at both 10 weeks and 20 weeks.

Table 33 *Measures of spasticity calculated by the difference between Tardieu R1 and R2*

							Change in spasticity compared to Baseline			
Outcome	**Baseline**		**10 weeks**		**20 weeks**		**10 weeks**		**20 weeks**	
	Happy Rehab	Usual Care	Happy Rehab	Usual Care	Happy Rehab	Usual Care	Happy Rehab	Usual Care	Happy Rehab	Usual Care
Right hamstrings spasticity (°)	12.2	7.1	10.1	8.3	7	5.2	-2.1	1.2	-5.2	-1.9
Left hamstrings spasticity (°)	13.8	10.9	8.6	6.3	15.1	2.9	-5.2	-4.6	1.3	-8
Right hip adductors spasticity (°)	9.6	3	6.6	2.5	7.1	0.5	-3	-0.5	-2.5	-2.5
Left hip adductors spasticity (°)	5.2	5.1	7.7	2.3	-0.2	1.5	2.5	-2.8	-5.4	-3.6
Right gastrocnemius spasticity (°)	15.4	13.8	14.4	11.1	11	12.8	-1	-2.7	-4.4	-1
Left gastrocnemius spasticity (°)	16.7	11.4	14.6	15.5	16.9	13.4	-2.1	4.1	0.2	2
Right Duncan Ely spasticity (°)	5.4	2.3	4.7	5	2.6	-0.1	-0.7	2.7	-2.8	-2.4
Left Duncan Ely spasticity (°)	0.5	1	3.1	-0.8	-3.4	0.3	2.6	-1.8	-3.9	-0.7

Table 33 shows that spasticity reduced more in the Happy Rehab group in 5 out of 8 muscles at 10 weeks and 6 out of 8 muscles at 20 weeks. Overall, the reduction in spasticity (difference between R1 and R2) was in favour of the Happy Rehab group by an average reduction in spasticity of 0.6° at 10 weeks and 0.6° at 20 weeks.

3.3.5.5.7 Measures of muscle strength

Data were collected for all participants at baseline. The intervention and usual care group were similar in strength measures at baseline (Table 34). Four individuals completed all three assessments of muscle strength in the Happy Rehab group and five in the usual care group (*Table 35).* Overall, the usual care group gained more strength than the Happy Rehab group in 7 out of 8 muscles at 10 weeks, gaining on average 1.6kg more per muscle group. In contrast, the Happy Rehab group increased their strength more than the usual care group in all muscles at 20 weeks, gaining on average 2.5kg more per muscle group.

Table 34 *Comparison of mean values of dynamometry at baseline, 10 weeks and 20 weeks between the intervention and control group.*

Outcome	Baseline		10 weeks		20 weeks	
	Happy Rehab n=8/8	Usual Care n=7/7	Happy Rehab n=5/8	Usual Care n=6/7	Happy Rehab n=5/8	Usual Care n=5/7
Muscle strength right hip abductors (kg) Mean (SD)	11.6 (4.1)	12.0 (6.0)	9.3 (1.4)	14.5 (7.2)	14.3 (10.7)	9.8 (5.4)
Muscle strength left hip abductors (kg) Mean (SD)	11.4 (4.6)	11.2 (5.6)	8.9 (2.9)	12.7 (6.3)	10.9 (5.1)	11.1 (7.5)
Muscle strength right hip adductors (kg) Mean (SD)	9.2 (4.6)	12.1 (6.5)	10.7 (4.1)	12.9 (6.0)	13.9 (7.2)	10.4 (4.5)
Muscle strength left hip adductors (kg) Mean (SD)	9.3 (5.2)	10.4 (4.6)	10.3 (4.3)	13.0 (6.1)	11.9 (5.8)	8.7 (4.6)
Muscle strength right ankle dorsiflexors (kg) Mean (SD)	5.3 (2.2)	7.1 (5.1)	8.3 (3.7)	12.6 (7.3)	6.8 (2.2)	7.2 (5.8)
Muscle strength left ankle dorsiflexors (kg) Mean SD)	9.2 (6.3)	8.3 (5.2)	7.6 (3.3)	12.7 (7.4)	8.8 (6.1)	6.9 (2.8)
Muscle strength right quadriceps (kg) Mean (SD)	13.8 (4.3)	15.1 (8.4)	12.3 (3.7)	15.7 (9.3)	13.2 (5.4)	11.5 (5.8)
Muscle strength left quadriceps (kg) Mean (SD)	16.1 (6.5)	14.0 (8.4)	15.0 (4.3)	16.7 (9.4)	14.1 (4.6)	11.7 (4.2)

Key- Number (n), standard deviation (SD), kilogram (kg)

Table 35 *Changes from baseline of measures of dynamometry at 10 and 20 weeks for children who undertook all three assessments.*

Outcome	Change at 10 weeks		Change at 20 weeks	
	Happy Rehab n=4/5	Usual Care n=5/6	Happy Rehab n=4/5	Usual Care n=5/5
Muscle strength right hip abductors (kg) Mean (SD)	0.2 (2.8)	2.9 (0.9)	0.3 (2.3)	-2.2 (1.4)
Muscle strength left hip abductors (kg) Mean (SD)	-1.3 (4.1)	2.5 (2.8)	0.5 (2.2)	0.3 (2.0)
Muscle strength right hip adductors (kg) Mean (SD)	3.2 (1.9)	1.5 (4.0)	1.8 (2.2)	-1.3 (4.8)
Muscle strength left hip adductors (kg) Mean (SD)	2.1 (3.3)	2.9 (3.6)	1.1 (1.8)	-2.7 (3.3)
Muscle strength right ankle dorsiflexors (kg) Mean (SD)	3.1 (4.1)	4.3 (4.8)	-0.9 (3.9)	-4.3 (2.8)
Muscle strength left ankle dorsiflexors (kg) Mean (SD)	1.4 (3.6)	3.0 (3.0)	-0.3 (3.6)	-3.5 (3.3)
Muscle strength right quadriceps (kg) Mean (SD)	-0.4 (1.8)	1.9 (6.2)	-0.3 (2.2)	-3.3 (5.8)
Muscle strength left quadriceps (kg) Mean (SD)	1.4 (3.6)	3.7 (3.9)	-1.6 (3.7)	-2.3 (4.4)

Key- Number (n), standard deviation (SD), kilogram (kg)

3.3.5.5.8 CHU-9D Quality of life questionnaire

Either young people completed the questionnaire themselves or their parent completed the proxy version of the CHU-9D. One young person completed the questionnaire in each group at both time points. Parents completed the rest of the questionnaires. The CHU-9D score was very similar scores between groups at baseline with no significant change at 10 weeks (Table 36).

Table 36 *CHU-9D Quality of life questionnaire*

CHU-9D	Baseline		10 weeks	
	Happy Rehab n =8/8	Usual Care n=7/7	Happy Rehab n=3/8	Usual Care n=6/7
Mean (SD)	14.5 (4.2)	14.1 (3.4)	14 (5.6)	14 (3.9)

Key- number (n)

3.3.5.5.9 Completeness of outcome measure data

Table 37 shows the proportion of data collected at baseline per outcome measure. It then shows the completeness of data for all possible outcomes, including those who withdrew or did not attend at week 10 and 20. Additionally there are columns showing the completeness of data for those participants who remained in the trial.

At baseline the Pediatric Balance Scale (PBS), Modified Tardieu Score (MTS) and hand-held dynamometry had complete data sets. The Next Step test data was complete in the usual care group at baseline; however, one child declined this test from the Happy Rehab group. Gait kinematic data was incomplete at baseline due to technical difficulties recording the data as well as two children declining to participate in the test. The PBS and MTS had the most complete data sets across all time points.

Table 38 shows the number of assessments unattended by participants remaining in the trial, and the reasons given. One participant was ill and then unable to reschedule attendance at assessment before the end of the trial. Two other participants did not attend assessments due to the time pressure from other family commitments. Several families reported that they had an increase in health appointments following the reopening of clinics following the COVID 19 lockdowns.

Table 37 *Completeness of data- Number of possible outcomes collected at each time point for all participants by group*

Outcome	Baseline		10 weeks				20 weeks			
			Number compared to baseline		Number for participants remaining in trial		Number compared to baseline		Number for participants remaining in trial	
	Happy Rehab n=8	Usual Care n=7	Happy Rehab n=8	Usual Care n=7	Happy Rehab n=5	Usual Care n=6	Happy Rehab n=8	Usual Care n=7	Happy Rehab n=5	Usual Care n=5
Knee angle Midstance (°)	4/8	6/7	4/8	4/7	4/5	4/6	5/8	2/7	5/5	2/5
Knee angle trough to peak (°)	3.5/8	5.5/7	4/8	4/7	4/5	4/6	5/8	2/7	5/5	2/5
Ankle angle at initial contact (°)	5.5/8	6/7	4/8	4/7	4.5/5	4.5/6	5/8	2/7	5/5	2/5
Ankle angle at midstance (°)	5.5/8	6/7	4/8	4/7	4.5/5	4.5/6	5/8	2/7	5/5	2/5
Pediatric Balance Scale	8/8	7/7	5/8	6/7	5/5	6/6	5/8	5/7	5/5	5/5
Next Step peak COP and velocity COM	7/8	7/7	5/8	5/7	5/5	5/6	5/8	4/7	5/5	4/5
Stepping error (mm)	4/8	5/7	5/8	4/7	5/5	4/6	5/8	4/7	5/5	4/5
Tardieu score R1 and R2	8/8	7/7	5/8	6/7	5/5	6/6	5/8	5/7	5/5	5/5
Muscle dynamometry	8/8	7/7	5/8	6/7	5/5	6/6	5/8	5/7	5/5	5/5

Key- Outcome measure (OM), degree (°), Tardieu fast (R1), Tardieu slow (R2), centre of mass (COM), number (n)

Table 38 *Number, proportion and reasons for non-attendance*

Outcome	Baseline		10 weeks		20 weeks		Reasons for non-attendance
	n	%	n	%	n	%	
Intervention participants missed assessments	0	0	1	20	1	20	Ill and unable to attend before end of trial
Control participants missed assessments	0	0	1	17	1	20	Family commitments

Key- number (n)

3.3.5.5.10 Diary data

Children in the Happy Rehab group were asked to exercise for 15-20 minutes, three times per week. Children in the usual care group were not given instruction as to the type or frequency of physiotherapy to undertake.

Some of the children who recorded diaries were also interviewed in the qualitative part of the study, and where appropriate their study pseudonyms (Table 39) are used, in support data triangulation. Five participants from each group completed diary data categorising their feelings about their exercise on each day. Their responses were coded for analysis using a Likert scale of 0-3. The modal response was calculated for these responses over the 10 weeks.

Both groups reported that they felt OK, when asked how tired and how wobbly they felt compared to normal, on the day that they trained. Both groups categorised the level of challenge was ‘just right’. When asked how they found the training, the usual care group ranged between ‘fun’ and ‘boring’. The Happy Rehab responded that it was either ‘fun’ or ‘OK’. All the Happy Rehab group respondents recorded that they trained in the afternoon, whereas three of the five children in the usual care group trained in the morning.

Only one person recorded having a problem while training. Here, Alfie (Table 39), in the Happy Rehab group, had a problem with the machine not working properly. Reasons given for not training included when Alfie was staying at their other parent’s house on six occasions or Gabby was away from home on holiday. Sometimes children did not train because they felt unwell (Gabby once and Alfie four times). Four children reported that they did not train because they were too tired. This included Gabby and Harry on four occasions and Alfie on one occasion and twice for a child from the control group.

Three children did not train when they had injuries such as bruised knees after a fall (Gabby) or having sore toes (Freddie) and a child from the usual care group who fractured his metatarsal. Several children did not train if they had done a lot of sport that day. For example, four children recorded that they had done a lot of exercise at sports day, gymnastics, football or cricket club (Gabby, Freddie, Isaac and Harry).

Table 39 *The number and type of adverse events (AEs) and relatedness to the intervention*

Adverse Events	Number of occurrences of AE	Type	Number Related to Intervention
	1	Bruised getting out of Happy Rehab unsupervised	1
	2	Falls and bruised knees	0
	2	Sore toes	0
Serious adverse events	1	Fractured metatarsal	0

Table 40 *Completion of diaries and adherence to intervention*

	Intervention n=5/8	Control n=4/7
Number of diary entries Mean (SD)	13.2 (13.0)	34.8 (25.3)
% Completed diary returns Mean (SD)	44 (43.4)	115.8 (84.4)
% Adherence to prescribed dose of intervention Mean (SD)	34.2 (28.8)	151.4 (188.7)
Number of participants unable to access the intervention	1	0
Number of photos submitted per participant Mean (SD)	0.6 (0.9)	0.75 (0.96)
Number of exercise sessions per week Mean (SD)	1.32 (1.30)	3.5 (2.5)
Number of minutes of exercise per week Mean (SD)	20.5 (17.3)	35.1 (24.5)
Number of times unable to train due to unavailability of equipment Mean (SD)	0	0
Number of times participant experienced a problem with the equipment Mean (SD)	1 (2)	NA
Amount of NHS staff hours needed to support intervention Mean (SD)	2.3 (2.1)	1.5 (1.9)
Amount travel time (mins) needed access intervention Mean (SD)	0	0
Cost of travel to access intervention Mean (SD)	0	0

Key-number (n), standard deviation (SD), percentage (%)

Table 39 shows the number and type of adverse events recorded in the diaries. There was one serious adverse event. A child from the usual care group had a fractured metatarsal sustained during activities unrelated to the trial. One child sustained bruises getting out of the Happy Rehab by them self, when they were unsupervised. The bruises resolved within a few days and did not require treatment. A child from the intervention group had sore toes related to wearing their leg splints.

Table 40 shows that nine participants completed the diary. Participants were asked to record a diary of their exercise session 15-20 minutes, three times per week, for ten weeks. This was expected to generate an expected 30 entries. The control group recorded a mean 34.8 (SD 25.3) entries, whereas the intervention group recorded a mean 13.2 (SD 13.0) entries.

The intervention group recorded a mean of 20.5 (SD 17.3) minutes per session, undertaking a mean of 1.32 (SD 1.30) sessions per week. The usual care group recorded a mean of 35.1 (SD 24.5) minutes per session, over a mean 3.5 (SD 2.5) sessions per week. The usual care group recorded that they exercised 51% more than the amount of exercise the Happy Rehab group were asked to do. The intervention group only recorded 34.2% adherence to the recommended exercise intensity. One participant was unable to use the Happy Rehab at all. Participants recorded a mean 1 (SD 2) occasions where a problem with the Happy Rehab prevented exercising.

Physiotherapists recorded that the intervention group required a mean 2.3 (SD 2.1) hour support over the 10 weeks compared to 1.5 (SD 1.9) in the usual care group.

3.3.6 **Qualitative findings**

3.3.6.1 Participant demographics

Nine parent and child dyads were recruited: five from the intervention group (n= 3 urban, n=2 rural) and four from the control group (n= 3 urban, n=1 rural). Four children had coexisting additional needs: learning disability, autistic spectrum disorder (ASD) and hearing impairment. One parent consented to be interviewed although he declined consent for his son to participate. One family who had consented withdrew after baseline assessment due to their child receiving botulinum toxin injections, which meant that they no longer met the

criteria for the trial. See Table 41 for the demographics of parent and child participants. One parent and child dyad who consented to be interviewed dropped out due to family and work pressure. Three physiotherapists who delivered the intervention and control arms of the RCT were interviewed (Table 42).

Children were assigned a pseudonym, selected alphabetically (A-I) by the researcher, which is used throughout the findings. Limited demographic details are presented to ensure anonymity. Physiotherapists are referred to as Physio 1, Physio 2, and Physio 3 (Table 42). Two interviews were conducted in a clinical setting, one in the family home and the rest took place over secure video calls with no other people present. One repeat interview was carried out with Bella, as she could not remember the assessments. The repeat interview took place after her final assessment to enable recall of her experience.

Data arising from the physiotherapist, parent and child interviews have been synthesised and are presented together in the text. Additional illustrative quotes can be found in Appendix 3.

3.3.6.2 Photograph data

Four participants uploaded photos to their diaries; Gabby and Freddie had pictures of themselves representing their usual care and Alfie and Caleb using the Happy Rehab at home. Gabby looked happy in her photos exercising against the wall at home and climbing a large wooden obstacle in an outdoor setting. Freddie was in a classroom, surrounded by toys. He looked as though he was concentrating on trying to kick a toy with his foot. Caleb's photo showed him in his kitchen where he had a smile on his face and appeared to be concentrating on the game he was playing using the Happy Rehab. Alfie's photo showed him in the Happy Rehab in his lounge, with an un-smiling look of concentration on his face. Their expressions reflect their opposing views that they expressed during their interviews. Caleb enjoyed the games and found them vey motivating, whereas Alfie only liked one game and found the others too difficult and became frustrated when the Happy Rehab didn't work properly.

Table 41 *Demographics of parent and child participants*

Child's pseudonym	Age (Years)	Group	Impairments affecting the child's communication	Parent	Location
Alfie	10	Intervention	No impairment	Mother	Urban
Bella	16	Intervention	Hearing impairment Learning disability Makaton	Mother	Urban
Caleb	7	Intervention	No impairment	Mother	Urban
Daisy	14	Intervention	No impairment	Father	Rural
Ethan	13	Intervention	Learning disability	Mother	Rural
Freddie	7	Control	Autistic spectrum disorder	Father	Urban
Gabby	7	Control	No impairment	Mother	Rural
Harry	9	Control	Learning disability Autistic spectrum disorder	Mother	Urban
Isaac	11	Control	No impairment	Father	Urban
Joseph	12	Declined to participate	Not recorded	Father	Urban

Table 42 *Demographics of physiotherapist participants*

Physiotherapist code	Physiotherapy experience (years)	Paediatric experience (years)	Location of work
Physio 1	20	18	Community
Physio 2	9	6	Special School
Physio 3	15	6	Community

3.3.6.3 Themes

Five main themes and their subthemes are presented in Figure 15, drawing together the findings arising from the interviews with the children, their parents and physiotherapists.

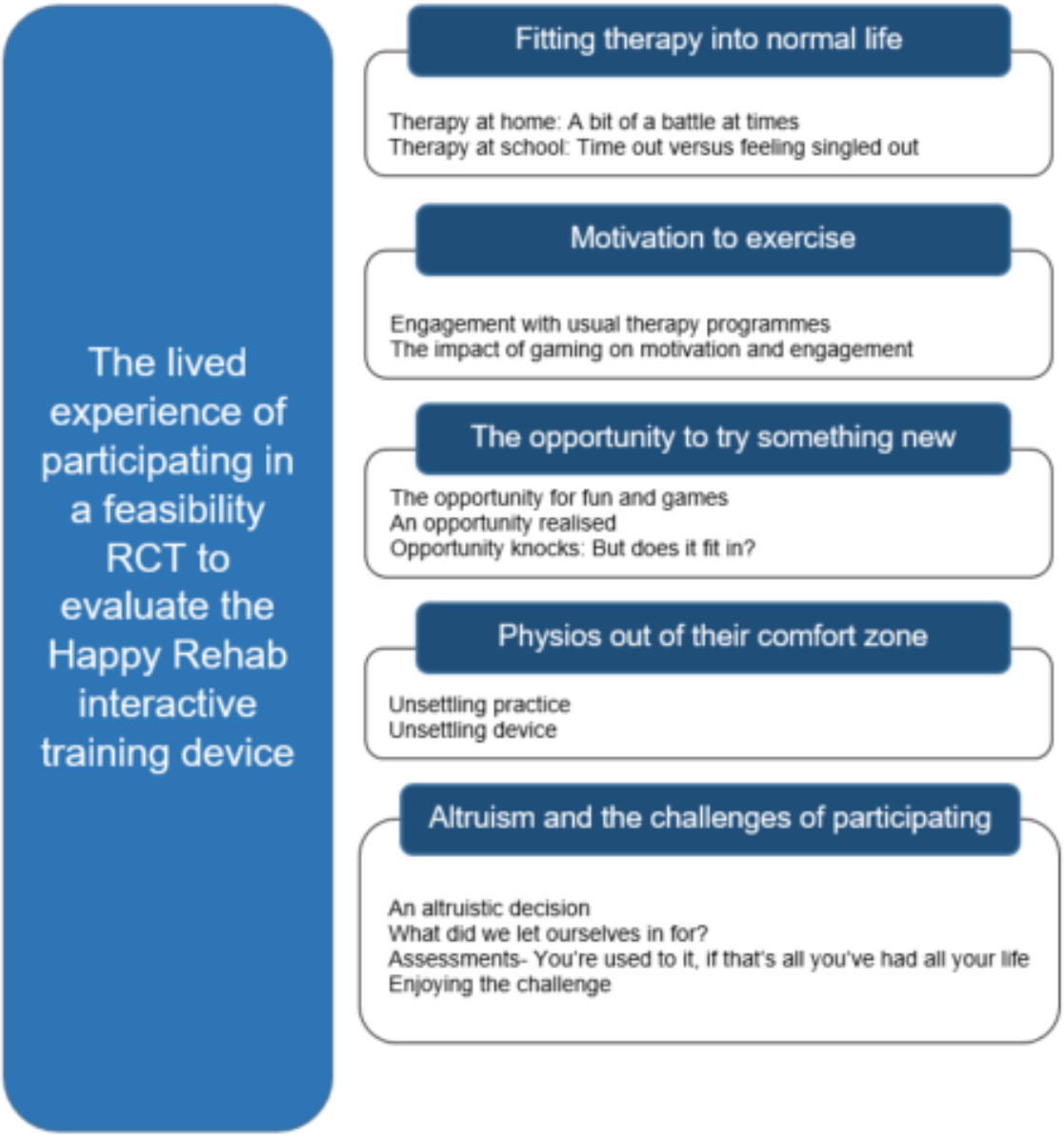


Figure 15 *Themes and subthemes arising from the interviews*

Theme 1, 'Fitting therapy into normal life', explores how the parents and children fit therapy into their lives at home and at school. In theme 2, 'Motivation to exercise', examines what motivates children to adhere to their exercise programmes and the impact gaming might have on motivation and engagement with exercise. In theme 3 'The opportunity to try something new', the opportunity that a new intervention brings is investigated along with whether it delivers on

its promise and how it fits in. In theme 4, ‘Physios out of their comfort zone’, how physiotherapists felt unsettled within their practice by the new device. In theme 5, ‘Altruism and the burden of participation’, the altruistic intent of those taking part in the study and the burden this placed on them is explored. This includes the experiences and feelings of the children being assessed during the study.

3.3.6.3.1 Fitting therapy into normal life

This theme explores the tension between being the child’s parent and the expectations and requirements associated with delivering the child’s physiotherapy at home. Both mothers and fathers talked about the important role that school has in supporting their child’s therapy, but they noted that it was sometimes at the expense of missing time in class.

Therapy at home: A bit of a battle at times

All but one of the parents agreed that it was difficult to find time to fit in therapy at home, with dividing attention between their children. It is also difficult to motivate their children to engage with their exercises, as Isaac’s father explained:

> *“Doing the exercises can be a bit of a battle at times, you know. We normally try and do it of an evening. Um you [Isaac] spend a lot of time with your games consul, so we try to say ‘come on, just take 10 minutes out of that to do your exercises. Do what you’ve got to do’” (Isaac’s father).*

Harry’s mother finds that short exercises fitted into with care routines, are acceptable to her son. Trying to persuade their child to do something that they do not want to do was reported as sometimes creating conflict in the relationship between parent and child. Gabby’s mother described how her daughter was much happier to do therapy at school, where she enjoys time out of class, but strongly resists doing the same exercises with her mother:

> *“It's the opposite of at school - you're getting out doing something boring… that you don't want to do. And so, it's great fun in comparison. Whereas at home, it's like… why do I have to do this? And ‘I hate you mummy’” (Gabby’s mother).*

Therapy at school: Time out versus feeling singled out

All the mothers and fathers of children attending primary or special schools described how much of the therapy programme is carried out at school with the support of the teaching assistant (TA). Gabby talked with excitement about missing class and having fun therapy sessions with her TA:

> *"[It's] quite good… I normally do stretches and exercises. I go out of class. [I feel] a bit pleased because I miss a bit of my learning, and she does things, like she told me how to count to 10 in French whilst doing the exercises" (Gabby, aged 7 years).*

Isaac also talked about the liking to miss lessons. However, most mothers and fathers said that they wanted to minimise the impact of their children missing lessons. Daisy's father was concerned about Daisy having to do things differently from peers, such as taking time out from lessons. He worries that using the Happy Rehab in school is a visible marker of her disability and that this could make his daughter vulnerable to bullying:

> *"…the trouble is it singles her out then, unfortunately.., and she gets enough stick from time to time at school because of obviously the disability and things, so we didn't really want to emphasise that anymore if we could help it?" (Daisy's father)*

This meant that it would only be acceptable to both Daisy and her father to have the Happy Rehab at home, rather than at school.

3.3.6.3.2 Motivation to Exercise

Motivation to exercise was a strong theme that was discussed throughout the interviews. Participants discussed intrinsic and extrinsic rewards, achieving their goals and the impact that gaming has on motivation and engagement.

Engagement with usual therapy programmes

Parents spoke of the difficulty motivating their children to do their physiotherapy programmes at home, especially where they found them boring and repetitive. Gabby and Ethan's mothers spoke about the need to use bribes and rewards to motivate their children. Several children talked about the repetitive nature of

their physiotherapy exercises. For Ethan, this meant that it was “boring”, and for Isaac it was painful at times:

> *“Some moves kind of hurt me, well the one where you have to go on the stairs. It kind of hurts here (shows ankle). [I still do it] because it’s my physio and I have to do it. It’s all right. If I have to do it, then I’ll do it. But if I didn’t have to, then I don’t think I would” (Isaac, aged 11).*

Freddie’s father described how the familiarity and predictability of doing the same exercises was very comforting for his son, because Freddie has autism:

> *“I kind of liked the fact that it was our standard exercises, the same exercises every day, so he knows what he is going to do”* (Freddie’s Father).

Freddie and Bella explained that they find it fun to play with a ball and or sing songs while exercising. Alfie, Ethan and Gabby reflected that they had achieved some of their goals although it was unclear whether the goals themselves motivated them to exercise.

The impact of gaming on motivation and engagement

There was unanimous support from parents, children and physiotherapists for the idea of gaming to motivate children to engage in physiotherapy. Ethan’s mother thought that the distraction of the game would help to take Ethan’s mind off his stretches. Isaac and Daisy agreed that they were likely to do more exercise if there were computer games integrated into the exercise. Caleb’s mother explained what impact gaming had had on Caleb while he was exercising using the Happy Rehab:

> *“He gets tired and he gets like he's had enough. Whereas on this [Happy Rehab] it's like 10 minutes and he's actually enjoyed it and he's actually wanted to do it every day. Whereas obviously if I've had to do his stretches everyday … he hasn't been so forthcoming, like not wanting to do it so much” (Caleb’s Mother).*

Ethan talked of the competitive nature of gaming. He found that he was competitive with himself, and felt compelled to try to beat his previous record:

"And you got to get a certain amount of stars each time… it's quite competitive…you tried to beat your record of like stars each time" (Ethan, aged 13).

This compulsive element of gaming described by Ethan seems to represent something more than just distraction or entertainment.

3.3.6.3.3 The opportunity to try something new

This theme includes thoughts and feelings of the parents and children who took part in the intervention group, and the physiotherapists who delivered the novel intervention. The theme comprises three subthemes, which explore the opportunity for fun and games, whether the Happy Rehab delivered on this promise, and the practicalities of living with the Happy Rehab.

The opportunity for fun and games

The physiotherapists were unanimous in their feeling that the Happy Rehab was likely to offer something different and appealing that might get children exercising more. Physio 2 identified the visual feedback from the games as an important concept that may help stimulate movement:

"The idea of having the screen that they can get that instant feedback, Uh, in a visual way is really good" (Physio 2).

Physio 1 talked positively about the potential benefits of the Happy Rehab in providing support for practicing weight transfer skills in standing:

"… those weight shift games … that's what's hard to do to simulate in sort of hands on therapy with him. So yes, definitely potential for improvement [with his] goals" (Physio 1).

Bella, Ethan, Caleb and Alfie all enjoyed some of the games; however, Alfie found some of the games too hard:

"Well I did [enjoy it] at first, but it … the games were really hard. Except for space invaders. Not all of [the games] were hard. Space invaders was good and that's it!" (Alfie, aged 10).

Alfie was very cross that some of the games were too hard and he ultimately found this demotivating. His physiotherapist (Physio 1) said that of the games

Alfie chose, she was not aware which ones might be the right level of challenge for him. She suggested that in future, a quick guide should be developed to help with choosing and setting the most appropriate games. According to his mother, a factor influencing Alfie's opinion of the Happy Rehab was that he did not like feeling restricted in the device and that he had hurt himself trying to get out of it unaided. He sustained bruises, which resolved within a week. This was reported as an adverse event associated with the intervention.

An opportunity realised

Many of the children found that playing the games with their leg movement was very intuitive. However, Bella needed some additional support to learn and understand that she was controlling the game with her movement. Her mother explained:

> *"she wasn't aware that she was controlling until you know she did a few times and her teaching assistant said 'look, move that, look see this on here' or 'move that, look see that' or 'now you're doing it' and then she got it!" (Bella's Mother).*

Ethan, Caleb and Bella's mothers recognised that the device helped to increase their children's ankle range of movement. Here, Caleb's mother discusses how her son responded to the new device:

> *"Well I do think he has benefited from it? hasn't he? I noticed he wasn't complaining as much that he was aching too much …I thought his flexibilities were a bit better. Like, he wasn't so tight" (Caleb's Mother).*

Physio 2 noticed that the games helped Bella to shift her weight more evenly over each foot. Two of the three physiotherapists raised concern as to whether all the activity was good, for example, Physio 3 was concerned about the level of selective motor control observable in the children while they are playing the games:

> *"Sometimes it's really difficult to know how well the child is actually physically working in the Happy Rehab…because it feels like quite a lot of generalised movement…so I feel like in terms of like isolated muscle*

control and movement, it's really quite difficult to unpick and if it's being that specific" (Physio 3).

Physio 2 observed compensatory movements and expressed the wish to coach the child to learn selective control by using their hands to guide the child to perform more controlled movement.

Three of the four children who trained on the Happy Rehab (Ethan, Alfie and Bella) experienced some issues with either apparent loss of power in the device, or difficulty with controlling the game. Ethan's mother describes how the faulty sensor on the footplate prevented full use of the game:

"We couldn't always get the ship down to refill with bullets, so then we would have to or he would have to wait for him to be like….disappear and then a new one had come, and then he'd start again" (Ethan's Mother).

The loss of power experienced by Alfie and Bella were related to incorrect calibration of the Happy Rehab. The device should be switched on before the child steps in to play the games in order to calibrate itself. This lack of calibration was identified early in the trial as the issue that was causing apparent lack of power; additional training was put in place for physiotherapists delivering the trial.

Opportunity knocks: but does it fit in?

There was consensus from all parents and physiotherapists that although the new device was large and took up a lot of space, they were willing to give up that space for 10 weeks. Alfie's mother represented the view of all the parents:

"I've only got a small house so it's sort of thing where you saw in the corner and he has other machines as well. It's OK if you got space for it like it was out of the way for me. It was in the corner. So, it didn't really bother us really..." (Alfie's Mother).

Alfie's mother also had concerns about Alfie potentially tripping over the wires leading to the plug socket. Physio 2 talked about a child who was in the intervention group but had difficulty accessing the Happy Rehab as it was on a different floor of the house. The insurmountable block to Daisy adhering to the

new intervention, was that the Happy Rehab could not get into the house. Daisy's father explained:

> *"We got a porch door and then internal front door…and it was the internal front door that's too narrow. We could go through the porch door. Yep, we couldn't then get any further into the house from there, so it's a real shame" (Daisy's Father).*

Physio 1 suggested that it could be set up at the CDC for families to come and use, and that a physiotherapy assistant could support these sessions. However, this was not an option for Daisy when she was considering training in the device three times per week:

> *"It's not very easy for us to get there (into the CDC) with like mum and dad in work and me in school and different activities out of school. Yeah, so it wasn't really possible to get across" (Daisy, aged 14).*

Daisy's father and Ethan's mother agreed that the travel and time required to train this frequently was not feasible for their families. The parent of the child who withdrew due to botulinum toxin injections was offered the intervention three months later. She declined to have the Happy Rehab at this point as it would have included the Christmas period, and she felt she would not have room for the device and a Christmas tree.

3.3.6.3.4 Physios out of their comfort zone

This theme explores the experiences of the physiotherapists developing confidence while preparing to use the new device and feeling unsettled when things went wrong.

Unsettling practice

During the trial, the three local NHS physiotherapists were trained how to set up the Happy Rehab for children on their caseloads. They all agreed that the initial training met their needs; however, they were unsettled when having to put this into practice. This had an impact on their levels of confidence. Physio 2 describes her experience with the Happy Rehab, and how unsettling it was not being able to feel which movements the child was making:

> *"I find that [setting the exercises resistance] quite difficult. I think that was probably one of the hardest things that we had to do, 'because I couldn't*

feel it myself and I think we as therapists like being able to feel it ourselves and know what it's like" (Physio 2).

All three physiotherapists found some difficulty with problem solving when things went wrong. Under usual circumstances, they would have contacted a representative from the product distributer to help solve these problems. However, the combined effects of Brexit (Britain's exit from the European Union) and the COVID-19 pandemic meant that the support from the distributor was not available. Two of the physiotherapists spoke of the additional burden they felt participating in the trial, especially due to the increased caseload pressure following the emergence from pandemic lockdowns. Physio 3 suggested a more feasible solution for a full RCT:

"I think it's placing quite a lot of burden on NHS physios if that is who are delivering it [during the trial] when they already have really big caseloads. I think it would be preferable to have a researcher that would go out and set it up" (Physio 3).

Unsettling device

Physio 3 had difficulty getting the right size device for a child on her caseload, even though she had measured him for the device, the child's feet were too big for the footplates. All three physios experienced times either where the device stopped or when the child struggled to operate the games. Physio 1 mentions how this might affect the trusted relationship that she has with the parents. Physio 2 considers the impact this would have if it happened during a treatment session:

"If there's an issue…if you are setting it up … then it suddenly doesn't work. Then do you cancel that session and then try and find someone or …just get on the phone straight away to sort it there and then? Especially if you've bought someone into a centre to do it, if it's not at home. You don't want to waste that appointment for them" (Physio 2).

3.3.6.3.5 Altruism and the challenges of participating

This theme encompasses the selfless act of deciding to do something for the greater good and living with the consequences of that decision. This included

the result of randomisation, the commitment to keep a diary and the challenge of the assessments for the children participating.

An altruistic decision

Three parents (names of children) expressed the desire to contribute to make things better for other families and children in the future. All parents said that they felt that they received enough information to make the decision to participate. Daisy spoke with a pioneering attitude about testing the new device on behalf of her peers:

> *"… you can test it. See if it works and it'll be like a new technology... You can use it for other people. So, if they've got a problem with their legs and stuff...to help it" (Daisy, aged 14).*

The opportunity to try something new excited most parents and children and this may have motivated some to take part in the study. However, all participants spoke about their acceptance of the need for a control group and randomisation process to decide the group allocation. Gabby's mother was typical in saying:

> *"Even if they were just in the control group, it was still nice to know that this study was going on. You know you can't do research without the control, can you?" (Gabby's Mother).*

What did we let ourselves in for?

There were several instances where the consequences of the decision to participate in the trial led to disappointment or frustration. Gabby was disappointed not to have the 'robot physio'. Harry's mother echoed this:

> *"I guess to a point we were a little bit disappointed that we didn't get the other group. But it is what it is, and if it helps you with your study..." (Harry's Mother).*

The online diary proved to be difficult for some parents. Freddie's father found the online diary simple to use, but this was not a view shared by Caleb, Alfie and Gabby's mothers. The diary prompted participants to record all three exercise sessions each week, and for some children this might have occurred at

home and school. Alfie's mother suggested a weekly progress summary would be sufficient and Gabby's mother proposed that a handheld diary would be easier to record in. The form, frequency and likely adherence to recording an exercise diary needs to be established before inclusion in a future trial.

Assessments – "You're used to them, if that's all you've had all your life"

The feasibility RCT included four tests of balance and clinical impairment. The Next Step test involved stepping to a target. Bella's mother described how Bella took a while to understand the test and held herself un-naturally until she felt comfortable with the test. Alfie disliked how The Next Step tested his balance to the limit. Alfie described how this felt:

> *"… the first one where we went step, step, step, [Next step test] … it felt like you were about to fall over" (Alfie, aged 10).*

Harry, Gabby and Freddie disliked how the gait analysis equipment felt. For Freddie and Harry this seemed to be more about having the equipment on their bodies:

> *"I like it without them [coda markers] …because I don't like the thing…. I don't like how it feels" (Freddie, aged 7).*

Gabby was concerned about the sticky tape and how it felt to have it removed from her skin. Gabby's mother noticed that Gabby was much happier when the markers were stuck to her leggings. Freddie and Harry's parents both felt that their children would have benefitted from more detailed information about the tests, so that they could be prepare their children for each stage, sequentially. During assessment sessions, children were invited to tell us to stop if any move became uncomfortable. In practice this rarely happened, however during the interviews, Freddie expressed his dislike of the fast stretches associated with the range of motion measures and Gabby and Isaac talked about aspects of the tests that hurt them. There was a sense of passive acceptance towards tolerating these uncomfortable parts of the assessments. Caleb's mother encapsulated this situation by saying:

"He's used to it, he doesn't like people pushing his legs around, but if that's all you've had all your life…" (Caleb's Mother).

Enjoying the challenge

Physio 3 was concerned about the 90-minute assessments. She thought that they might be too demanding for the children to get through. However, Gabby, Freddie, Caleb and Harry's parents agreed that their children coped well despite the assessment being 90 minutes long and Isaac noted:

"A bit time consuming but it was quite fun" (Isaac, aged 11).

Daisy and Bella's parents considered the length of the assessment acceptable. Caleb's mother thought the sessions were tiring for her son. While she enjoyed seeing Harry undertaking gait analysis, his mother stated that Harry would never be able to do all the tests, because of his autism.

Caleb, Ethan and Daisy enjoyed the Next Step, where a noise sounded before a target randomly lit up that they then stepped onto. Ethan remarked:

"It's quite fun. I liked that. You didn't know what number was going to go off then…It might have helped my, my reaction time" (Ethan, aged 13).

The majority of the children enjoyed most aspects of the assessments, except for Harry and Freddie. Gabby's mother commented that the encouragement that Gabby received during the assessments was received as positive validation of her power and skills, and that this contrasted to the backdrop of her child's disability being perceived as weakness making her somewhat 'less than' her peers.

The one parent who was interviewed after he declined for his child to participate was very supportive of the Happy Rehab in principle. He expressed support for gaming and felt that this would be of huge benefit to his child. He explained that the reason he declined was because of the trial design:

"I think initially there weren't any barriers from our point of view. We've got room in the house for the frame to go, so that wouldn't have been an issue. I think the only concern we had was that it would be in place of his

usual physio. And we've seen in the past, if we don't do daily physio with [my child], he really suffers from that. He really starts getting unbalanced and it's like his legs will be crouching a lot more and he won't be able to stretch them out properly." (Joseph's father).

This father indicated that then he would have consented to his child participating if the trial was designed with the new intervention in addition to his child's usual care.

3.3.7 Synthesis and discussion of the findings of the feasibility study

The results and findings are synthesised in this discussion section. The discussion links to the main aim of the study, which was to establish whether it is feasible to conduct an RCT to assess the effectiveness of a ten-week physiotherapy intervention using an interactive trainer in children with CP. The feasibility outcomes are summarised at the end of the section in Table 43.

The discussion synthesises these findings by addressing the feasibility of the trial, the barriers and facilitators to using the Happy Rehab, the lived experience of usual physiotherapy care and participants' feelings around taking part in research. The strengths and limitations of the trial will be addressed and topics for future research will be suggested.

3.3.7.1 The feasibility of the trial

The trial was initially planned to run between 1/5/2020 to 1/8/2022. However, the COVID-19 pandemic affected the opening date and duration of this feasibility trial. The start of the trial was delayed due to national lockdowns and the partial closure of NHS services. Following the lifting of lockdown and easing of restrictions, there was a considerable delay in NHS research and development departments opening to new studies. This was a challenge as the feasibility trial needed to be completed within the period limited Fellowship. The trial steering committee was consulted and a collaborative decision was reached that a shorter trial should be undertaken, between 1/3/2021 to 1/8/2022. This resulted in a shorter recruitment period, the recruitment target was reduced from 40 to 15, and the number of sites was reduced from five to two.

The lack of availability of Happy Rehab devices and technical support for them was also a factor that affected the trial. Britain's exit from the European Union (Brexit) resulted in the UK distributer withdrawing the Happy Rehab from their product sales. This meant that the trial was run without the expected technical support, and any repairs and support were accessed through the product designer (Innovaid™) in Denmark. Support was accessible via video calls. However, repairs to footplates could only be carried out by staff trained by Innovaid™. Other studies using rehabilitation devices at home have required high levels of technical support [181, 182]. Implementation of the Happy Rehab into clinical practice and a further trial using the device would be unfeasible without the appropriate, timely and effective technical support in place.

During this study, there were four Happy Rehab devices, which meant that recruitment was carried out at each site until saturation was reached. There are three sizes of device with different weight limits. During the baseline assessment, the child's height and weight were measured to determine which frame would be needed. The recruitment rate was limited by the availability of the devices. Potential participants who expressed an interest were held on a waiting list so that they were booked for initial assessment when a device became available.

A randomisation model 1:1 between intervention and control was used. Some consideration was given to contamination such as where several participants could be in in one place, for example, in a special school setting or where there were siblings who were eligible. However, these problems were not encountered in practice. For a larger study, consideration should be given about whether to use cluster randomisation in these situations. Minimisation of age over or under 9 years of age and GMFCS levels I and II versus GMFCS level III was used. However, only one participant of GMFCS level III was recruited so this limited the effect of the minimisation process. In a larger trial, this would still be an appropriate model to use if the eligibility criteria remained GMFCS I-III. The eligibility criteria by GMFCS level is a topic that will be further addressed during the discussion of outcome measures.

Withdrawals and loss to follow up related to services reopening after the COVID-19 pandemic. Families found that there was a sudden increase in appointments and treatment offers from services that had been closed. This put

increased pressure on families and meant that children were missing school more than usual to attend multiple appointments. Physiotherapists delivering the trial experienced increased pressure from the backlog of work following the reopening of services.

3.3.7.2 Barriers and facilitators to using the Happy Rehab

Participants showed support for the Happy Rehab as a tool for motivating children to do therapeutic exercise, and to reduce the burden on parents carrying out manual therapy techniques with their children. Parents and children saw gaming as a usual part of children's life that could be usefully incorporated into physiotherapy.

There is an emerging body of evidence to support 'serious games' in therapy [183]. This topic will be addressed in more depth in Part 4 of the thesis. Similar therapeutic games reported in the literature use relatively low cost and portable virtual reality games that might be used at home, while others focus on expensive lab-based robotic movement training [184-189]. The former may have little guided movement or support, where the latter has fully supported and guided movement. The Happy Rehab sits somewhere in between, as it involves supporting the child in standing position while they operate the games by using movements assisted or resisted by motors in the footplates and kneepads.

Most children were highly motivated by the games, badges and virtual rewards in the Happy Rehab games. Studies have shown that the inclusion of gaming such as Virtual Reality games can increase the repetitions of therapeutic exercise [190]. However, one child did not agree with this view. He was not motivated by the games, in part because he found them too hard and his motivation reduced further after he experienced faults with the device. Stroke patients have also reported loss of motivation with rehabilitation games when there are technical problems or where they needed help to access the device [191, 192]. The insights from the children highlight the need to pitch the challenge of the training at the right level of difficulty, also called the 'just right challenge' [193]. Where the challenge is too high or perceived as unachievable, this can have a de-motivating effect. A review of Serious Games highlighted the need to be able to customise rehabilitation games in order to take into consideration the needs of individual patients [194]. With the correct skill, the therapist can set up the equipment to enable the child to work sufficiently hard

into a therapeutic range while still achieving the goals that make the game motivating.

Nearly all parents were willing to accommodate the device for a 10-week period, except when it was in direct competition with the family's needs for space, such as at Christmas. Parents have previously expressed a willingness to sacrifice space or the aesthetic of their home to accommodate large equipment, where they consider it benefits their child [195]. Several parents felt that their child should not use the Happy Rehab at school as it might single them out. A study exploring the use of standing frames at schools found that they can detract from peer-to-peer interaction [196] when the user is doing something different from the rest of the class. Teachers have also expressed concern that competing demands of rehabilitation devices in the classroom may reduce educational opportunities [192].

Physiotherapists supported the idea of the Happy Rehab as a useful physiotherapy intervention over 10 weeks. They expressed conflicting views between the benefit of supporting children to do more activity, especially activity that might encourage compensatory movements. Traditional practice has included reducing compensatory, undesirable movements and facilitating movement that is more typical. There is growing evidence that suggests that goal-based functional task practice is more effective at improving function than targeting impairments [145, 197]. Children with CP learn to move against a background of neurological impairment. Neuronal group selection theory [198] suggests that while therapy can be targeted at helping younger children to develop a broader repertoire of neuronal networks, therapy in older children should be targeted towards providing ample opportunities to practice activity, which may include compensations for the primary impairments [199]. This is a controversial topic in physiotherapy, which will be explored further in Part 4.

This feasibility study set out to develop and evaluate a complex intervention [25]. A key component of this intervention was the clinician's ability to set up the intervention and coach the child and family how to use it. Physiotherapists in the study were concerned about their own lack of confidence and experience with the Happy Rehab and how it might affect the trusted relationship they have with the families. Effective physiotherapy relies on developing a therapeutic relationship between the therapist and patients and carers [200], and being

confident as an autonomous practitioner. Previous studies have shown the importance of pre-trial training and education for the clinicians and manualised interventions [201]. However, in this instance the physiotherapists did not establish the confidence to set up and tailor the programme for individuals at home. This may have been due to time constraints, competing demands as the service reset after lockdown and the number of physiotherapists delivering the intervention. The trial physiotherapists suggested that a research therapist would be better skilled to do this in a main trial, in order to reduce problems setting up the device. However, the trial protocol aimed to test the intervention as a viable alternative to a block of physiotherapy within the National Health Service. An efficient solution within NHS practice might be to train physiotherapy assistants to become more skilled in using the Happy Rehab so that they can support the registered staff who might want to use it as a modality to treat children on their caseload. Another study found that clinicians adopting novel technology recommended having a 'champion' within their team to facilitate others to become more confident with the device [192]. Therefore, a future trial would need to include a more detailed and flexible training programme for physiotherapists as part of the site set up, as well as supporting the development of a Happy Rehab 'champion' with more in-depth skills who could support their colleagues.

3.3.7.3 Outcome measures

Two potential primary and three secondary clinical outcome measures were used in the trial. Of the two, the Pediatric Balance Scale (PBS) was the more complete than the Next Step test. The PBS, modified Tardieu scores (MTS) and dynamometry had the most complete data sets across all time points. However, two participants who have GMFCS I reached ceiling scores at baseline, and a further one at follow up. For the other participants, the PBS did show group change above minimal detectable difference (MDD). Potential signs of efficacy for the Happy Rehab group were seen in increase in PBS, passive range of motion and spasticity at 10 and 20 weeks, and dynamometry and the Next Step (ML-COP) at 20 weeks. Outcomes used in the trial related to gait, balance, strength and range of movement. However, the ability of children to control selective movement may influence the ability to train on the Happy Rehab and to carry out the Next Step test. This could be included as an outcome measure

in a full RCT using the Selective Control Assessment of the Lower Extremity (SCALE) [202].

Children in the feasibility RCT cohort had smaller APAs at baseline than the children with CP studied in the Next Step observational study, in Part 2. This means that the children recruited into the trial had more impaired balance than the previous group of children with CP. Not all children found the test easy to complete. One of the barriers to completing the Next Step test was that three children found it difficult to tolerate the placement of markers on their limbs. For two children this limited the number of steps for which data was collected and for one child meant that no data was collected for the Next Step. A study of gait analysis with autistic children excluded a child on the basis of not being able to cooperate with the procedure, although they did not report whether this was due to the inability to tolerate the markers [203]. Another study using similar equipment used eligibility criteria stating the child must be able to follow the verbal instructions [204]. Two children found it hard to complete full sets of the Next Step test due to fatigue. Studies show that fatigue is a common problem due to higher energy demands of moving with CP [205-207]. Fatigue may also be related to the motor planning required for the constrained stepping task [208]. The chosen outcome measures are not feasible to use with all children, and therefore eligibility criteria should include the ability to complete the outcome measure at baseline.

The PBS and measures of clinical impairment were more acceptable to all children. However, the Next Step test allows a more physiological and quantifiable analysis of balance mechanisms than the PBS. The PBS has a ceiling effect which was identified in children with GMFCS I in this study. Therefore, it is necessary to weigh up the relative importance of wider eligibility criteria with the type of information and effect size that the two measures of balance can detect. The PBS would be best placed to capture changes in functional balance tasks in children GMFCS II and III, whereas the Next Step test would be preferable in quantifying the mechanism of dynamic balance in children with GMFCS I and II. It is important to note that the Happy Rehab offers potentially unique treatment options for children with GMFCS III, who may not be able to otherwise sustain training in an upright standing position. Therefore, the outcome measure would need to reflect the children included in

the trial. The Happy Rehab itself could be used as an outcome measure of weight shift if used as part of the baseline assessment.

Overall, children tolerated the assessment sessions but many commented on the length and complexity of the tests. It was unclear at the outset of the study which impairments the Happy Rehab might target. This study has shown potential changes in balance strength, range of movement and spasticity. The burden of testing should be reduced in a full trial to capture changes in only the most relevant outcomes.

Children and parents found the structured goal setting used in the Canadian Occupational Performance Measure (COPM) motivating. One young person reported increasing adherence to her usual physiotherapy care because she wanted to achieve her goal. Goal setting combined with activity-focused interventions has been shown to improve outcomes for individuals [92] and may have accounted for this change in adherence. The increased contact with a physiotherapist during the trial may have encouraged greater adherence to usual care.

3.3.7.4 Strengths and limitations

This study used a mixed methods approach to find the feasibility of an RCT to improve balance and walking. This is relatively uncommon within this field but an extremely important part of determining the feasibility of a trial and of a novel intervention [209, 210]. In order to trigger children's engagement in sharing their experiences, child-oriented opportunities and approaches to generating data were utilised [211]. Rather than relying solely on verbal approaches, the triggers for child engagement included the use of electronic tablet devices to encourage digital recording of experiences using a format that is a familiar part of their everyday lives. In addition, the use of photographs within the photo-elicitation interviews provided an additional visual trigger for engagement.

Where children and parents were interviewed together, the interviewer made it clear that they would interview the child first and followed by the parent. This was done to enable the child to discuss the topics with their own thoughts and feelings, unhindered by their parent's ideas. This was a relatively successful strategy, although the parent often contributed to the child's opinion or spoke when the child left a pause for thinking. At times children looked to their parents

for approval before speaking, for example, where Gabby wanted to tell the interviewer that the adhesive tape had hurt her. The interviewer needed to reassure her that it would be helpful to know all the good and bad things about her experience.

A strength of this study was the inclusion of a wide range of children with CP. The eligibility criteria for this study was deliberately broad so that children with CP with additional communication needs could be included. Four children with additional co-morbidities were recruited; these co-morbidities were hearing impairment, epilepsy, learning disability and autistic spectrum condition and these are common in children with CP. Several children needed the support of their parents as communication partners. Bella's mother supported her to use Makaton, and Harry's mother helped interpret his reactions to memories of the trial. This approach allowed the study to be more inclusive of participants, including those with learning disability and autism, who could not have been able to be interviewed without support. These children tended to give more concrete and shorter responses than the children without these co-morbidities, and this affected the depth of reflection on their experience. However, they were also very clear when indicating their like and/or dislike of certain procedures in the trial. This was invaluable when considering the feasibility of including the broadest eligibility criteria for a trial in children with cerebral palsy. One limitation to the study was the relatively low numbers of photographs uploaded via the diaries. During the interviews, two of the children were able to use videos and photos from their parents' phones to stimulate recollections of exercising.

During the interviews, there were several examples of children complying with activities that hurt them. It is interesting to reflect if non-disabled children might not comply in the same way. Physiotherapy tends to be pervasive throughout the childhood of children with physical disabilities, leading children to comply with uncomfortable and time-consuming therapy and assessments [92]. Children have a fundamental right to have their voices heard, particularly in research about them [93]. It is important that children are involved in creating research that is acceptable to them, so as to develop effective therapies that justify the time and effort that they and their families spend on therapy. It is imperative to collaborate with children to develop appropriate ethical research,

with children as partners, rather than the objects of research. The findings from this study reflect the commitment to partnership and highlight children's lived experiences of participating in research.

The qualitative study cannot claim to have reached data saturation from data that was collected. It is contentious subject as to whether data saturation exists, even in larger studies [212, 213] . However, in this study, although there was a constrained number of children in the trial they had range of different experiences, some in the control and some in the intervention group. One participant was unable to carry out the intervention at all. Some topics discussed by participants included repeated ideas that lead to consensus around a theme, such as the struggle to fit therapy into normal life and motivation to exercise.

The position of the interviewer as a paediatric physiotherapist introduced potential bias, as being a paediatric physiotherapist places the interviewer in a position of power over the child. This may have prohibited them from speaking freely about negative views of physiotherapy or the trial. The interviewer had prior knowledge of children's interactions with therapeutic exercise at home and at school, which introduces further potential bias. The interviewer has extensive experience of interacting with children with cerebral palsy that enabled her to pitch conversations in an age-appropriate way. The interviewer attempted to remain conscious of the potential power imbalance during interviews. She gave children encouragement to speak their minds and to tell her what they really thought about physiotherapy and taking part in the trial. As in other studies, children were told that there were no right or wrong answers and that what they said was important for making the trial better for everyone [214]. The interviews were child-led, but the use of topic guides helped to ensure that the interviewer could ask open questions about a similar range of topics in each interview, if there was a lull in the conversation.

One weakness in the protocol was that the Happy Rehab group stopped accessing the intervention at 10 weeks but the Usual Care group were able to continue training. This may have affected the results. One parent declined for his son to participate because he was concerned about stopping his usual care. Children with CP often have well-established exercise programmes at home and school. This interview highlights the ethical dilemma of choosing an RCT

with usual care as the comparator. For this family they would not want to stop therapy that works for their child. Other families were disappointed that they were not allocated to the novel intervention. An alternative method would be to use a best alternative treatment in a superiority trial. The best alternative could have been better defined usual care. This would involve working with the expert physiotherapy group to refine the usual care balance intervention. An alternative evidence-based identified comparator in the literature review was vibration plate therapy. This may have been more appealing for families but would present further feasibility issues in the NHS setting, as it would necessitate training Physiotherapists in two novel device interventions. A recommendation for a full trial would be to amend the protocol to the Happy Rehab intervention plus usual care or to reinstate the usual care programme after the cessation of the Happy Rehab.

3.3.7.5 Recommendations for future research

Other topics, which arose, deserve deeper exploration, such as the struggle between being a parent and having to take the role of therapist when doing home exercise with their child. This tension between the caregiving/protector role and ensuring the child adheres to doing therapeutic exercise is an important topic that deserves further investigation.

The Next step test is unfeasible for roll out to a full trial in its current format due to the length of time for data collection and complexity of the technical set up. A simplified version of the Next Step test may be feasible with further work to validate the test using inertial sensors and a dedicated target mat.

The Happy Rehab has the potential to support children to target multiple impairments. The feasibility trial findings show some indications that balance functions, flexibility and muscle strength may improve with training in the Happy Rehab. However, it is unclear which impairments are the most important to target to improve balance control mechanisms. Further work is needed to explore this, and such work should consider the impact of fatigue and motor planning and selective motor control deficits on the mechanisms of dynamic balance. Future trials for balance in children CP may need to sample children with GMFCS I and II separately from children with GMFCS III, as the outcome measures most appropriate to the child's functional level are different for the

two groups. The Happy Rehab creates a unique training opportunity for children at GMFCS level III and a larger trial is justified within this population of children.

3.3.7.6 Conclusions

This trial tested the feasibility and acceptability of a trial exploring a novel interactive training device. There were multiple feasibility issues related to the COVID-19 pandemic and Brexit. Additionally, some families raised concern about accommodating the device at home or within an educational setting and physiotherapists required more technical support to deliver this complex intervention.

Children found the gaming aspect motivating and enjoyable. Parents and physiotherapists agreed that the exciting potential of gaming to enhance exercise may improve the lived experience of children with CP. The Happy Rehab presents an opportunity for children to undertake interactive training on a device positioned between robotics and virtual reality. The Happy Rehab shows promise in facilitating children to target their impairments while training in a functional position.

Table 43 *Assessment of feasibility objectives to inform a definitive trial.*

Objective	Outcome	Study Results	Suggested modification	Feasible with modification (Y/N)
Focus	Methods			
Feasibility of the trial				
Acceptability of the trial	Number of declines, interviews of physiotherapists, parents and children.	Trial welcomed by children, families and physiotherapists . 3 families declined -one did not want to stop usual care. NHS physiotherapists found implementing the intervention very challenging. 2/15 families unable to accommodate the device at	Intervention plus usual care. Improved package of training and support for the intervention by research therapists. Eligibility criteria need to access a device at home or school.	Y

		home and were unwilling to attend clinic 3 times per week.		
Can we recruit and retain participants?	Number of participants eligible. Number recruited and randomised, date of recruitment recorded on study database. Recruitment source. Number of withdrawals. Number of participants lost to follow-up.	Recruitment rate limited by number of devices and effects of pandemic. Retained 11/15 children in intervention phase.	Need more device availability. Need supplier support in the UK, to reduce wait for device availability.	N No UK supplier at present.
Effectiveness and acceptability of randomisation	Comparison of participant characteristics: severity, distribution of motor impairment, associated impairments at baseline interviews.	Randomisation was acceptable to participants. Minimisation criteria not effective due to low numbers of GMFCS III.	Consider cluster randomisation. Revise minimisation process.	Y
Effectiveness of concealment of allocation up to week 10	Number of times assessor correctly guessed treatment allocation.	Concealment process largely effective.	No modification.	Y
Concurrence with other surgical and medical interventions	Number of operations or procedures that target balance and walking during the intervention and follow up period.	Only 1 child withdrew due to surgical or medical interventions.	No modification.	Y
Change in clinical outcome measures	Change in assessment scores of outcome measures.	Pediatric balance scale detected change for 9/15 children at 10 weeks but ceiling effect reached for 3/15 participants.	Consideration of outcome measures depending on eligibility criteria. PBS suitable for GMFCS II and III. Next Step suitable for GMFCS I and II.	Y N Not yet suitable for multi-centred RCT.
Assess appropriateness	Number and percentage of outcome	Baseline measures	Children should not be randomised	Y

of outcome measures	measures completed at each time point. Participant view on acceptability by interview.	incomplete for some children. Next step and gait analysis unacceptable to some children due to markers on body. Assessment sessions too long. Mobile gait analysis unfeasible due to technical issues. Most complete set of outcomes are for PBS, Modified Tardieu Scale and muscle dynamometry.	unless baseline outcomes are complete. Choice of PBS or Next step related to GMFCS levels inclusion criteria as above. Reduce number of outcomes taken. Exclude gait analysis from main trial. Include PBS, Modified Tardieu Scale and muscle dynamometry in full trial. Next Step not feasible for use in multi-centre trial in current form.	Y Y N Y N
Feasibility of Intervention				
Adherence to treatment	Diary data frequency and duration of training. Poor rate of completion.		Include paper version of diary Reduce the frequency of recording to a weekly summary plus AEs.	Y
Acceptability of treatment intervention	Incidence of breakdown of equipment. Number of times participants were unable to access equipment. Participant view on acceptability of interventions by interview.	Breakdown of equipment was a significant barrier for 1 participant, who disliked the equipment and felt trapped by it. 2 participants enjoyed using the Happy Rehab particularly the gaming aspect of it.	Eligibility criteria should include ability to tolerate being in the equipment without feeling trapped.	Y

		1 participant could not access the treatment as it did not fit into their house.		
Cost of intervention and support needed to use it	Local physiotherapist record of staff time and grade used to support intervention. Travel costs of staff and families. Number and cost of repairs	There is a small increase in staff hours for Happy Rehab.	Support from UK would reduce the amount of staff hours needed. Include physiotherapy assistants in supporting the intervention in the main trial.	Y
Safety of intervention	Number and type of SAE and AE	There was 1 AE related to the Happy Rehab.	Ensure parents are present and supervise the child training in the Happy Rehab.	Y
Acceptability of Participation				
Acceptability of participation	Themes identified from interviews/photos.	Participation was acceptable to children but not for all outcome measures. Physiotherapists required more support to deliver the intervention.	Reduce burden of testing Additional support required from UK supplier Identify a Happy Rehab champion within each team	Y

4 Part 4: Synthesis of the work

This section synthesises the whole thesis. It includes reflections on the strengths and limitations of the work and its relevance to policy and practice. It will address future directions and present the conclusions.

4.1 Strengths and limitations of the programme of work

The work presented in this thesis is novel and innovative. This is evidenced by the publication of three pieces of work in peer-reviewed scientific journals. The work has been improved through scrutiny from supervisors, co-authors, reviewers and journal editors and through discussions with practice colleagues and reflection on my own ongoing practice.

The overall focus of this work has been balance in children with cerebral palsy (CP). A recently published Delphi survey has identified balance and falls as core outcomes to be measured following lower limb surgery in children with CP [215]. The Pediatric Balance Scale (PBS), or elements of PBS, are often used as clinical measures of balance and in research studies. However, the ceiling effect means it is not appropriate for children with higher levels of mobility. The Next Step test showed good to excellent reliability when measuring medio-lateral APAs in children with CP GMFCS levels I and II. This is a useful advancement in knowledge, as the Next Step test provides a reliable and valid test of dynamic balance for children who reach the ceiling of the PBS. It also measures the quality of the child's movement, where the PBS only measures the outcome. In its current form, it is applicable in clinical trials where there is access to 3D motion analysis equipment. Like traditional gait analysis, it is reliant on children being able to tolerate wearing the markers and being able to follow the test instructions. The feasibility study revealed that autistic children found tolerating the markers problematic. This may have been partly because it was novel to them and they lacked experience and trust in the equipment. Parents indicated that with more preparation it might be possible for their autistic children to accept the markers and equipment. Time and resource needs to be allocated to making resources, such as social stories, in order to help children manage their anxieties about novel experiences [216]. This was a weakness in this study as it has been reported that 7% of children with CP may have autism concurrently [217]. This should be taken into consideration in research protocols that include children with autistic traits.

The Next Step test was developed in response to the absence of a suitable clinical and research tool for quantifying dynamic balance. This test allowed

identification that children with CP had reduced anticipatory postural adjustments (APAs) and increased stepping error compared to typically developing peers [218]. Children with CP were able to modulate their APAs depending on the position of the target, in line with previous work in adults [103, 218]. The test was developed from previous work by Lyon and Day, who described a model of dynamic balance during a single step [219]. Dynamic balance during the Next Step test occurs during single leg stance, with controlled carriage of the COM, and therefore the head and trunk, until the stepping leg at the target step receives the COM. The modulation of the trajectory of the step is controlled by the APAs [103, 220] and stepping error during this voluntary task could be the cause of a fall. This is a functionally significant aspect of balance as children with CP frequently fall during volitional movements, such as stepping in confined areas, for example when moving around their desk or chair at school. More evidence for this proposed linkage between reduced APAs and falling in CP is required. Further, this is only one aspect of dynamic balance. Other elements of dynamic balance include reactive balance, where balance is lost during perturbations or where leg movements need to be altered “midstep” to avoid an object [221]. While this is an important aspect of dynamic balance, reactive balance reactions were outside the scope of this programme of doctoral work.

Some children found the Next Step test more tiring than other participants did. One child with higher levels of cognitive and gross motor functioning found the task more tiring than his peers, and it was unclear why this was although they also reported that fatigue limited his daily function at school. A review of fatigue in people with CP reveals that they are actually less likely to experience physiological fatigue during sub-maximal fatiguing tasks than typically developing peers [222]. It is suggested that this finding might be due to the inability of people with CP to fully recruit highly fatigable muscle fibres. Moreover, athletes with and without CP experienced similar levels of muscle fatigue showing that there is an ability to train to overcome muscle fatigue [222]. An alternate explanation for fatigue might be due to problems with motor planning during the constrained stepping task. Differences have been found in the oscillatory responses of neurones in the sensori-motor cortices of children with CP carrying out a constrained knee extension task. The differences

detected in premotor cortical areas during the planning and execution of this task are suggestive of difficulties in motor planning [223]. This area is not well understood but may influence the ability to perform the Next Step task.

A strength of the feasibility study was the Trial Steering committee (TSC) which included parents and a teenager. The TSC along with the Trial Management Group (TMG) and mentorship from the Plymouth Clinical Trials Unit enabled the trial to be structured and planned in a systematic and family friendly way. The TSC gave strong and constructive checks and challenges in several areas. Firstly, the feasibility of the intervention was considered. The original plan had been to test the intervention in a clinic or school setting, but the young person and parents felt that some families would prefer the Happy Rehab at home. The protocol was changed to include this as an option. There was also strong challenge from the statisticians on the feasibility of randomising participants with four devices of three different sizes across five sites. The logistics and time required for transporting, setting up the intervention for 10 weeks and then cleaning and transporting it to the next person was carefully considered to make it as efficient as possible. The limited availability of devices was a limiting factor in the trial. Although recruitment was done systematically to include all eligible children, a waiting list was created for potential participants to wait for a suitable sized device to become available. This strongly influenced the recruitment rate.

Different models of randomisation were considered, such as block randomisation per clinic or school. This could be possible as the devices can easily be set up for multiple users in a home, clinic or special school setting. The TSC proposed a 50:50 randomisation model as it gave participants an equal chance of having the new intervention or usual care. This model raised potential problems. It is not uncommon for families to have siblings with CP or for several potential participants to attend the same school. This would have increased the chance of contamination between groups. It might also have discouraged families with siblings with CP from participating if their children were allocated to different groups. The TSC decided to proceed with the planned randomisation process and to use the protocol to record instances of cross-contamination and use the interviews to capture any thoughts and feelings about the randomisation process.

NHS gave ethics approval for the feasibility study RCT with an embedded qualitative study in February 2020. This was just a few weeks before the first national lockdown during to the COVID-19 pandemic. It was a very uncertain time, and it was unclear whether the study would be able to proceed. Major obstacles included the partial closure of Health and University departments with a redeployment of staff to other departments. University ethics approval was granted in June 2020. However, the local Trust Research and Development department was unable to open the study, as their work was limited to trials related to COVID-19. In October 2020, the TSC discussed whether the intervention could be adapted to minimise the need for face-to-face contacts with participants. This could have been achieved by changing to using video-based outcome measures, home interventions and the use of Personal Protective Equipment (PPE) to manage infection risk for essential contacts. It became clear that the trial could not run for the full period and the recruitment rate would have to be reduced from the planned 40 participants and five sites. A list of feasibility priorities were agreed with the TMG and TSC and major amendments to the protocol were submitted to the Health Research Authority. These included reduction of the recruitment target and number of sites.

During the period before the trial opening, the UK distributor stopped supplying the Happy Rehab and the UK supplier subsequently went out of business because of the pandemic. This created a challenging obstacle, as the company had agreed to loan two Happy Rehabs for the period of the trial. Innovaid, the designer of the Happy Rehab, were keen support the trial and agreed to loan two devices to enable the trial to continue. However, importing the devices was very difficult due to Brexit, which happened on 31st January 2020. There was also a delay in the Director of Innovaid entering the country to support initial training, due to restrictions in international travel associated with COVID-19. The devices eventually arrived safely but could not be used as the new loan arrangement meant that the devices were not insured for use in the trial. Lengthy discussions were facilitated between the University legal department and Innovaid. A risk assessment was undertaken as part of this process in order to allocate responsibility for the equipment at different stages of its use and storage. This enabled the University to insure the equipment for the trial. The loan agreement was finally signed in February 2021. When this was in

place the primary recruitment site had to then confirm their ‘capacity and capability’ to open the site to recruitment. The University Sponsor finally gave the green light to recruit to the trial on 9.2.2021, a year after initial ethics approval. There were so many complicated barriers to setting up and running the trial that it has been a real test of endurance. Running a trial with medical devices raised many more complications than expected. It is a testament to all the teams involved that this small feasibility trial was able to succeed.

The next section addresses the relevance of this work in relation to the wider context of physiotherapy for children with CP.

4.2 Relevance to policy and practice

Children with cerebral palsy (CP) face difficulties with walking and balance that affect their ability to be independent and to participate fully in functional and leisure activities [13, 14]. Children with CP are known to attain their peak gross motor function around the age of nine years [126], and many children experience decline in their skills due to secondary musculoskeletal problems. A growing body of work has identified the need to monitor and manage potential musculoskeletal decline in children with CP [224]. An important question to consider is whether physiotherapy resources should be prioritised towards the maintenance of gross motor function to prevent the predictable musculoskeletal decline over therapy interventions aiming to improve movement and balance function in both childhood and adolescence. Currently the evidence base is stronger for preventing musculoskeletal decline and evidence to support therapy interventions to improve balance is not well established.

Goal setting with the child is an essential part of task-focused training, with the intervention focusing on practicing components of the goal. This feasibility study found that setting goals using the Canadian Occupational Performance Measure (COPM) [225] improved adherence to usual care for one teenager. Children are much more likely to achieve their participation goals where interventions are family centred with intensive blocks of treatment rather than where they are frequent and low dose [226]. It is unclear whether functional gains made during task practice might be achieved by changes at an impairment level. It is possible that improvements could be due to changes in cognitive processes such as memory and planning [227] or in adopting

compensatory motor strategies. When considering whether balance can improve through targeting impairments, it is essential to understand the possible mechanisms for change that interventions may be targeting.

4.2.1 Can the balance of children with CP improve?

In Part 1 of this thesis, the primary motor impairments of children with CP were described as spasticity, weakness and reduced selective motor control. The Happy Rehab provides the opportunity for children to train intensively using lower limb resisted and assisted movement, and weight shift activities to operate the interactive games. Importantly, the Happy Rehab can be tailored to the needs of the child calibrating the equipment to enable the child to work in a therapeutic range of movement and at the right level of resistance. This feasibility RCT suggested an increase in range of movement at the ankles after 10 weeks of training that concurs with the results of previous work [24]. Improvements in muscle strength using dynamometry were measured at 20 weeks. Signals of efficacy for the Happy Rehab as an intervention for balance shown in change scores of the PBS and the Next Step tests were found. However, it is unclear whether balance control improved or just whether just the 'tools' of balance improved i.e. range of movement and muscle strength. For an improvement in balance control to have occurred this would require changes in the central nervous system occurring through "neuroplastic" mechanisms.

Neuroplasticity is a theory used to explain some mechanisms of recovery following neurological injury in adults. However, plasticity in neurological rehabilitation of adults is set upon a backdrop of previously intact sensori-motor pathways. Infants with CP develop their central nervous system (CNS) on a background of neurological injury, therefore mechanisms of neuroplasticity may be different in children with neurodevelopmental disabilities [227]. Early in the developing brain, there is an initial genomic generated approximation of neural networks. These are modified postnatally in response to exposure of the infant to specific environments, such as their specific language context, sensory enhancement or deprivation [228]. Animal studies have shown that the formation and trajectories of dendritic spines can alter during periods of sensory deprivation. In some areas, such as the visual pathways, development is time-critical and can lead to enduring impairments if it is disrupted [228, 229]. Conversely, it is postulated that enhancing stimulation creates opportunities for

neuroplasticity. There are strong indications that therapeutic interventions should be concentrated in the first two years of life in order to capitalise on this intense period of growth and plasticity in the CNS [230, 231].

Importantly, experience-dependent neuroplastic changes can be seen across the lifespan where neurons that are activated together develop stronger connections to become reinforced neural circuits [228]. This has been beautifully described as the ‘sculpting’ of the nervous system where there is an initial over production within regions of the cortex, with dendrite formation and synaptic connections developing. The connections that are reinforced by experience remain, while less used connections shrink back [229]. Plastic adaptation has been measured in the CNS of children with CP with dendrites sprouting projections on the ipsilateral side of the cortex enabling motor control from the uninjured part of the brain. This is illustrated by the ability of children to acquire gross motor skills following hemispherectomy for the treatment of epilepsy [232]. However, a negative functional impact of dendritic sprouting on the ipsilateral side can be seen in involuntary ‘mirror’ movements.

Proponents of neuronal group selection theory suggest that children with CP may lack appropriate activity in the basic neural circuits required for functional postural control. This can lead to stereotyped postures with reduced possibilities or delay in developing a secondary repertoire of movement responses [198]. It has been suggested that therapy in older children should be targeted towards providing ample opportunities to practice activity, which may include compensations for the primary impairments [199]. This opinion places the emphasis on the opportunity for lots of varied practice of skills, which is likely to take place against a background of postural impairment. However, the Happy Rehab enables children to train with their posture supported in a corrected position. It provides many opportunities for repetition of a variety of movements with the avoidance of compensatory or stereotyped postural synergies. It has also been argued that deficits in sensory processing contribute to the inappropriate scaling of motor responses in children with milder CP, and that augmented sensory feedback may help improve the scaling of motor outputs [198]. The Happy Rehab calibrates the sensory feedback from the games with the size and speed of motor responses, for example when steering a car around a racetrack. The repetition and intensity of playing the games provides an

enhanced learning environment on a backdrop of more normal postural activity. It potentially gives children with CP the opportunity to develop a secondary repertoire of neuronal circuits. These can then potentially be adapted to produce more accurate motor responses in everyday situations.

Our understanding of CP is changing. It has been proposed that CP is primarily a problem of disordered movement, resulting from impaired selective motor control with associated spasticity and muscle weakness [233]. The definition of spasticity has been refined following a review of the evidence, as disordered sensori-motor control resulting in the involuntary activation of muscles [234]. Problems with selective motor control (SMC) in children with spastic CP strongly correlate with both gross motor ability and functional balance [233, 235, 236]. Reduced SMC has been defined as the 'impaired ability to isolate the activation of muscles in a selected pattern in response to demands of a voluntary posture or movement' [237]. Disordered SMC can also be described as the obligatory co-activation of synergist muscles, frequently seen in children with CP. Impairment of SMC is linked to damage in the corticospinal tracts with an ensuing loss of normal excitatory and inhibitory activity. Functional balance requires SMC in order to move out of stereotyped synergies in order to produce timely APAs. The Next Step observational study found that children with CP had smaller APAs that may be linked with reduced SMC. It also showed that children with CP had a higher rate of stepping error than that of typically developing children. Problems with SMC by co-activation of synergists in the stepping leg may be responsible for trajectory errors when stepping and requires further exploration.

Training on the Happy Rehab demands distally selective movement to play the games within the set therapeutic range. For example, controlling the position of an avatar while playing a skiing game using ankle dorsiflexion and plantarflexion, and knee flexion and extension. The therapeutic range for this SMC training can be made more specific or more sensitive as the child makes progress. Some of the games use weight shift to control the game, through sensors in both footplates. For example, several scratch-card games require the user to shift their weight in all directions to uncover a complete picture. This has to be done with controlled and sustained weight bearing in each quadrant of the picture. These games relate more directly to balance and weight shift activities,

which are often incorporated into traditional physiotherapy sessions e.g. using a balance board. However, children with poor balance tend to move quickly to compensate for lack of sustained postural control when on a balance board. Physiotherapists appreciated the weight shift games within the Happy Rehab training as a new therapeutic opportunity. This was especially true for those children with GMFCS III who struggle to maintain standing so they can work on dynamic balance skills.

The Happy Rehab provides the ability to train with guided postures and movements within an enhanced, interactive environment. These are all important aspects of gaming and robotics and therefore it is useful to look at the literature to see how it might relate to the Happy Rehab.

4.2.2 Gaming and robotics

Virtual reality (VR) has gained popularity as an adjunct to neuro-rehabilitation in adults following stroke. It is said to drive neuroplasticity by combining opportunities for high repetition of goal-orientated tasks within an enhanced VR environment, thereby providing instant and concrete visuo-motor feedback on task performance [238]. Two small studies have shown changes in cortical organisation and areas of brain activation in children with CP undertaking upper limb training using VR. Furthermore, functional MRI showed that their functional improvements were linked to changes in the contra-lateral primary sensorimotor cortex [239], providing evidence of neuroplasticity. There is a lack of evidence to show that VR alone can stimulate improvement in balance in children with CP [238]. VR headsets provide immersive imagery that that make it easier to harness the principles of mirror training, stimulating recovery by tricking the brain into believing the impaired limb is moving. There is emerging evidence to suggest that interventions such as lower limb mirror therapy could improve balance in children with unilateral CP [235]. This works on changing the sensory feedback to improve movement control, particularly in children with unilateral CP. Some aspects of VR are used within the Happy Rehab, such as the enhanced environment and interactive nature of the games and avatars. There is potential for the Happy Rehab to develop games that use the principles of mirror therapy to improve the sensory learning and therefore movement control.

The sensory feedback from the Happy Rehab games is immediate and heavily incentivised. For example, moving the footplate up and down when controlling a

space invaders game enables the player to win stars and to avoid ‘being killed’ by aliens. These games encourage intensive training with high levels of repetition and activity, with variation in speed and strength of the movement required. If the intensity of training is enough, this neuromuscular activity is likely to stimulate neuroplastic changes, as well as changes in muscle structure and volume associated with resisted exercise training [227].

The Nintendo™ Wii-fit and balance board are commercially available gaming systems marketed with games that help to improve fitness or balance. They have interactive games similar to the Happy Rehab, providing the enhanced sensory learning and feedback experience. However, it is possible to play the games and achieve the goals of the game by using compensatory movements or with poor posture. This could potentially reinforce stereotyped patterns of movement or posture.

A 2017 systematic review showed moderate evidence that a 12-week programme of Wii- fit can improve balance in children with CP. However, the outcome measures used in most of the reviewed studies were unsuitable for measuring change in dynamic balance or excluding compensatory movements. It is also unclear whether any short-term gains were sustained [240]. One RCT found that the postural sway in standing improved in children with unilateral CP after 6 weeks of training on the Wii-balance board, but that this was not sustained after 4 weeks follow up [241]. A more recent feasibility RCT has shown that using a home programme of Wii-Fit is feasible and has the potential to improve lower limb muscle strength and walking; however, it did not include outcomes to measure dynamic balance [242]. Despite the apparent interest in Wii-fit as a therapeutic modality, there is a lack of evidence to demonstrate its ability to improve dynamic balance in the long term.

Robotic gait training provides an opportunity for children to experience and practice normal gait patterns in a robotic exoskeleton, usually using an enhanced sensory learning environment. It has been shown to be effective in improving gait parameters [243] but with limited evidence of improvement in dynamic balance control during gait [244]. However, it uses extremely expensive clinic-based equipment, and children have to travel to a centre to use it. In comparison, the Happy Rehab is more accessible in price and is relatively portable. The Happy Rehab provides support in a standing position through

pads at the hips, knees, back and trunk, which can be adjusted to the individual's height. Like robotic gait exoskeletons, the Happy Rehab uses robotic components that guide movement and provide assistance or resistance from the motors. The training programme during the trial intervention included 6 to 10 short periods of exercise over 20 minutes. This usually included passive games for a warm up and warm down, where children could relax and allow the Happy Rehab to move their legs through their full passive range. This relaxation component allowed children to experience a guided range of movement with enhanced sensory feedback synchronised with the leg movement. Equally when carrying out active and resisted games, the movements were guided by the Happy Rehab to give an experience of more normal movement patterns in the same way as robotic gait trainers do.

Gaming is an aspect of the Happy Rehab that physiotherapists, children and their parents and carers identified as likely to motivate the children and bring about greater adherence to therapy. Gaming is recognised as motivational and, in some cases, even addictive. Motivation through gaming can be due to intrinsic factors such as the joy of sensations derived from the experience of the game, or extrinsic motivators such as in game rewards [245]. The main components of gaming have been categorised as achievement, social and immersion. Male gamers are more likely to be motivated by achievement such as advancement through levels, power, status and accumulation; while female gamers are motivated more by games that involved collaboration and socialisation [246]. The games within the Happy Rehab focus mainly on achievement of tasks with rewards, i.e. extrinsic motivators that may appeal more to boys. In this study, rewards were reported to be an important factor to all of the children discussing their motivation while using the Happy Rehab.

4.3 Future directions

There is a lack of standardised care for children with CP in the UK [121]. This is surprising given that CP is the most common childhood disability seen in community physiotherapy departments across the UK. The 'National clinical guideline for stroke' [247] has been acknowledged as responsible for driving up clinical standards in stroke care. In CP, the relevant NICE guidance is found in two clinical guidelines: 'Cerebral palsy in under 25s: assessment and management' [22] and 'Spasticity management in under 19's' [248]. Neither

guideline addresses physiotherapy management of balance in great depth. The work undertaken in the usual care consensus study has provided essential foundational work towards defining usual care for children with CP [121] and should be further developed towards national standards of practice that include balance re-training.

There are no core outcome sets for children with CP and SMC is not routinely assessed or targeted in usual physiotherapy care aimed at improving balance [121]. The SCALE tool [249] is suitable for clinical use and should be considered as part of core outcomes required to fully assess children with CP. Further work is needed to create a core set of outcomes for physiotherapy aimed at improving SMC, balance and walking for children with CP.

This impact of the trajectory of leg position on stepping error during the Next Step test is yet unknown. Impaired SMC may affect the ability to position the foot during stepping and could be an explanation for the larger foot target error seen in the Next Step observational study [218]. It is unclear how SMC might affect dynamic postural control needed to maintain stability on the COM while stepping. Exploring the relationship between the Next Step measure of dynamic balance, PBS and SMC is an area that requires further research. The Next Step test was validated against the Quality Function Measure. Further work is needed to determine the correlation between the Pediatric Balance Scale (PBS), SCALE and the Next Step.

Training for 10 weeks on the Happy Rehab may be a cost-effective solution as a physiotherapy intervention on the NHS. The equipment can be used by multiple users in a school or for periods of home training. It would require the establishment of a more robust training protocol, and a Happy Rehab champion to support its use. The costs of staff time, transporting the device and technical support as well as any health outcomes needs to be calculated. Further work on the Happy Rehab should include a full Health Economic assessment.

The ideal position would be to have more accessible gaming and rehabilitation equipment available in community spaces. Physiotherapists agreed that they have a role in signposting children and young people with CP to use community facilities to keep fit and active. This often includes support to use traditional gyms, which can sometimes be inaccessible to wheelchair users. The Happy

Rehab requires the device to be calibrated to the user's height, strength and range of movement. As such, it would not be suitable for use by the public in an open access setting, such as a gym. However, it could be useful in a specialist exercise facility set up for people with disabilities who need more tailored or specialist equipment, such has been suggested in the adult neurological rehabilitation population [250]. This type of facility has the advantage of bringing together specialist rehabilitation equipment that can be used by people with disabilities, with the open access of a gym. This encourages an ethos of fitness and active lifestyle and rather than a medical model of rehabilitation. It would allow people to train at a time that is convenient, with other people while maximising the use of expensive equipment. This is particularly relevant given the number of children who are undertaking extensive exercise training following selective dorsal rhizotomy training [251]. Many families fund raise to purchase their own specialist training equipment. However, establishing a separate gym facility creates further segregation and is potentially stigmatising. Further PPIE work needs to be done with stakeholders to determine the implementation of the Happy Rehab within real world setting.

4.4 Conclusion and reflections

This programme of work has contributed considerably and across a range of studies to the body of knowledge on the balance problems of children with cerebral palsy (CP). The results of the Next Step study have highlighted key differences in the dynamic balance of children with CP and has demonstrated new and reliable ways to measure this. The consensus work has revealed the differences in usual care for ambulant children with CP in the UK and has laid the foundations for defining usual care. This is a first step towards developing national standards of care for physiotherapy for ambulant children with CP. The feasibility RCT has identified key factors needed to implement a full RCT and has given voice to the lived experiences of children and families who participated in this research. Their experiences and insights provide essential knowledge, not just for the planned RCT but will also inform other researchers to understand what factors children and families want to be designed into physiotherapy studies.

The fellowship has been a period of intense growth for me. I have enjoyed the ability to be intellectually creative and to work in a self-directed way. I notice a

huge improvement in my organisational skills, my ability to chair meetings and in my presentation skills. Working in research alongside clinical work achieves a perfect balance in my drive to work. I enjoy having the time and space to write up my work for publication.

This PhD fellowship has equipped me with the clinical, research and leadership skills that have enabled me to step into a new clinical academic role as an Associate Clinical Director within the NHS. This new appointment incorporates clinical and professional leadership as well as dedicated time for supervising and carrying out research. I am also a successful co-applicant on two large multi-centred RCTs exploring the effectiveness of stretching and strengthening physiotherapy interventions for children with CP. I will continue work to further validate the Next Step test and develop a more portable version of it for clinical and research use. This PhD was undertaken during the Covid-19 pandemic, which peaked at the most crucial part of this feasibility RCT. I am pleased that the key areas of feasibility of the RCT were evaluated. I plan to conduct further feasibility work with the Happy Rehab when there is technical support for the device in the UK.

I have been very fortunate to feel fully supported during this PhD by parents and young people, as well as my academic and research supervisors. I have been able to publish four papers from this PhD, one is in submission and a further three are planned. My immediate plans are to continue to disseminate the work to academics and clinicians, at national and international conferences, and to children and parents through family support networks and other opportunities, as they arise.

This PhD journey has been exciting and stimulating. I look forward to continuing this clinical academic journey by focusing on developing the evidence for effective physiotherapy interventions and improving the outcomes and quality of life for children with disabilities, and their families.

References

1. Oskoui, M., et al., *An update on the prevalence of cerebral palsy: a systematic review and meta-analysis.* Developmental Medicine & Child Neurology, 2013. **55**(6): p. 509-519.
2. Chounti, A., et al., *Sex differences in cerebral palsy incidence and functional ability: a total population study.* Acta Paediatrica, 2013. **102**(7): p. 712-717.
3. Rosenbaum, P., et al., *A report: the definition and classification of cerebral palsy April 2006.* Dev Med Child Neurol Suppl, 2007. **109**(suppl 109): p. 8-14.
4. Palisano, R.J., *GMFCS-E & R Gross Motor Function Classification System: Expanded and Revised*. 2007: Canchild centre for childhood disability research.
5. Palisano, R.J., et al., *Stability of the gross motor function classification system.* Developmental medicine and child neurology, 2006. **48**(6): p. 424-428.
6. *Prevalence and characteristics of children with cerebral palsy in Europe.* Developmental Medicine & Child Neurology, 2002. **44**(9): p. 633-640.
7. Cans, C., *Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers.* Developmental Medicine & Child Neurology, 2000. **42**(12): p. 816-824.
8. Andersen, G.L., et al., *Cerebral palsy in Norway: Prevalence, subtypes and severity.* European Journal of Paediatric Neurology, 2008. **12**(1): p. 4-13.
9. Lance, J.W., *Pathophysiology of spasticity and clinical experience with baclofen.* Spasticity: disordered motor control, 1980: p. 185-204.
10. Wren, T.A., et al., *Effects of preoperative gait analysis on costs and amount of surgery.* Journal of Pediatric Orthopaedics, 2009. **29**(6): p. 558-563.
11. Park, M.S., et al., *Issues of concern after a single-event multilevel surgery in ambulatory children with cerebral palsy.* Journal of Pediatric Orthopaedics, 2009. **29**(7): p. 765-770.
12. Brunton, L.K. and D.J. Bartlett, *The bodily experience of cerebral palsy: a journey to self-awareness.* Disability and Rehabilitation, 2013. **35**(23): p. 1981-1990.
13. Lindsay, S., *Child and youth experiences and perspectives of cerebral palsy: a qualitative systematic review.* Child: Care, Health and Development, 2016. **42**(2): p. 153-175.
14. Kamp, F.A., et al., *Energy cost of walking in children with spastic cerebral palsy: Relationship with age, body composition and mobility capacity.* Gait and Posture, 2014. **40**(1): p. 209-14.
15. Zwier, J.N., et al., *Physical activity in young children with cerebral palsy.* Disability and rehabilitation, 2010. **32**(18): p. 1501-1508.
16. Saether, R., et al., *Clinical tools to assess balance in children and adults with cerebral palsy: a systematic review.* Developmental Medicine & Child Neurology, 2013. **55**(11): p. 988-999.
17. Dewar, R., et al., *Perspectives on Postural Control Dysfunction to Inform Future Research: A Delphi Study for Children With Cerebral Palsy.* Arch Phys Med Rehabil, 2017. **98**(3): p. 463-479.

18. Rethwilm, R., et al., *Dynamic stability in cerebral palsy during walking and running: Predictors and regulation strategies.* Gait & Posture, 2021. **84**: p. 329-334.
19. Dewar, R., S. Love, and L.M. Johnston, *Exercise interventions improve postural control in children with cerebral palsy: a systematic review.* Dev Med Child Neurol, 2015. **57**(6): p. 504-20.
20. Wright, F.V., et al., *The Quality Function Measure: reliability and discriminant validity of a new measure of quality of gross motor movement in ambulatory children with cerebral palsy.* Developmental Medicine & Child Neurology, 2014. **56**(8): p. 770-778.
21. Pin, T., P. Dyke, and M. Chan, *The effectiveness of passive stretching in children with cerebral palsy.* Developmental Medicine & Child Neurology, 2006. **48**(10): p. 855-862.
22. Shaunak, M. and V.B. Kelly, *Cerebral palsy in under 25 s: assessment and management (NICE Guideline NG62).* Archives of Disease in Childhood-Education and Practice, 2018. **103**(4): p. 189-193.
23. Peplow, U.C. and C. Carpenter, *Perceptions of Parents of Children with Cerebral Palsy About the Relevance of, and Adherence to, Exercise Programs: A Qualitative Study.* Physical & Occupational Therapy In Pediatrics, 2013. **33**(3): p. 285-299.
24. Curtis, D.J., J. Bencke, and B. Mygind, *The effect of training in an interactive dynamic stander on ankle dorsiflexion and gross motor function in children with cerebral palsy.* Dev Neurorehabil, 2014. **17**(6): p. 393-7.
25. Craig, P., et al., *Developing and evaluating complex interventions: the new Medical Research Council guidance.* Bmj, 2008. **337**.
26. O'Cathain, A., et al., *Guidance on how to develop complex interventions to improve health and healthcare.* BMJ open, 2019. **9**(8): p. e029954.
27. Crotty, M.J., *The foundations of social research: Meaning and perspective in the research process.* The foundations of social research, 1998: p. 1-256.
28. Guba, E.G. and Y.S. Lincoln, *Competing paradigms in qualitative research.* Handbook of qualitative research, 1994. **2**(163-194): p. 105.
29. Polgar, S. and S.A. Thomas, *Introduction to research in the health sciences E-Book*. 2011: Elsevier Health Sciences.
30. Lyon, I. and B. Day, *Control of frontal plane body motion in human stepping.* Experimental Brain Research, 1997. **115**(2): p. 345-356.
31. Bancroft, M.J. and B.L. Day, *The Throw-and-Catch Model of Human Gait: Evidence from Coupling of Pre-Step Postural Activity and Step Location.* Frontiers in Human Neuroscience, 2016. **10**(635).
32. Shaw, J.A., D.M. Connelly, and A.A. Zecevic, *Pragmatism in practice: Mixed methods research for physiotherapy.* Physiotherapy theory and practice, 2010. **26**(8): p. 510-518.
33. Plack, M.M., *Human nature and research paradigms: Theory meets physical therapy practice.* The qualitative report, 2005. **10**(2): p. 223-245.
34. Timans, R., P. Wouters, and J. Heilbron, *Mixed methods research: what it is and what it could be.* Theory and Society, 2019. **48**: p. 193-216.
35. Onwuegbuzie, A.J. and N.L. Leech, *On becoming a pragmatic researcher: The importance of combining quantitative and qualitative research methodologies.* International journal of social research methodology, 2005. **8**(5): p. 375-387.

36. Thabane, L., et al., *A tutorial on pilot studies: the what, why and how.* BMC medical research methodology, 2010. **10**: p. 1-10.
37. Eldridge, S.M., et al., *Defining feasibility and pilot studies in preparation for randomised controlled trials: development of a conceptual framework.* PloS one, 2016. **11**(3): p. e0150205.
38. Piitulainen, H., et al., *The gait is less stable in children with cerebral palsy in normal and dual-task gait compared to typically developed peers.* Journal of Biomechanics, 2021. **117**: p. 110244.
39. Latash, M.H.-A., M., *what is posture and how is it controlled?*, in *Postural Control:A key issue in developmental disorders.* 2008, Mac Keith Press: London. p. 3-21.
40. Massion, J., *Postural control system.* Curr Opin Neurobiol, 1994. **4**(6): p. 877-87.
41. Westcott, S.L. and P.A. Burtner, *Postural control in children: implications for pediatric practice.* Phys Occup Ther Pediatr, 2004. **24**(1-2): p. 5-55.
42. Winter, D.A., *Human balance and posture control during standing and walking.* Gait & posture, 1995. **3**(4): p. 193-214.
43. Forssberg, H. and H. Hirschfeld, *Postural adjustments in sitting humans following external perturbations: muscle activity and kinematics.* Experimental Brain Research, 1994. **97**(3): p. 515-527.
44. Takakusaki, K., *Functional Neuroanatomy for Posture and Gait Control.* Journal of movement disorders, 2017. **10**(1): p. 1-17.
45. Peterka, R.J., *Simplifying the complexities of maintaining balance.* IEEE Engineering in Medicine and Biology Magazine, 2003. **22**(2): p. 63-68.
46. Fitzpatrick, R.C., *More pulsating movement.* J Physiol, 2003. **551**(Pt 1): p. 4.
47. Loram, I.D., C.N. Maganaris, and M. Lakie, *Active, non-spring-like muscle movements in human postural sway: how might paradoxical changes in muscle length be produced?* J Physiol, 2005. **564**(Pt 1): p. 281-93.
48. Nashner, L.M., *Fixed patterns of rapid postural responses among leg muscles during stance.* Experimental Brain Research, 1977. **30**(1): p. 13-24.
49. Nashner, L.M., *Adaptation of human movement to altered environments.* Trends in neurosciences, 1982. **5**: p. 358-361.
50. Dietz, V., *Human neuronal control of automatic functional movements: interaction between central programs and afferent input.* Physiol Rev, 1992. **72**(1): p. 33-69.
51. Hadders-Algra, M., *Development of Postural Control*, in *Postural Control: A key issue in developmental disorders*, M. Hadders-Algra and E.B. Carlberg, Editors. 2008, Mac Keith Press: London. p. 22-73.
52. Edelman, G.M., *Neural Darwinism: Selection and reentrant signaling in higher brain function.* Neuron, 1993. **10**(2): p. 115-125.
53. De Vries, J. and B. Fong, *Normal fetal motility: an overview.* Ultrasound in Obstetrics and Gynecology: The Official Journal of the International Society of Ultrasound in Obstetrics and Gynecology, 2006. **27**(6): p. 701-711.
54. Pountney, T., et al., *Chailey approach to postural management*, in *Physical management in neurological rehabilitation*. 2004, Mosby.
55. Wolff, D.R., et al., *Postural balance measurements for children and adolescents.* J Orthop Res, 1998. **16**(2): p. 271-5.

56. Hedberg, Å., et al., *Early development of postural adjustments in standing with and without support.* Experimental Brain Research, 2007. **178**(4): p. 439-449.
57. Sveistrup, H. and M.H. Woollacott, *Longitudinal Development of the Automatic Postural Response in Infants.* Journal of Motor Behavior, 1996. **28**(1): p. 58-70.
58. Conner, B.C., et al., *The cross-sectional relationships between age, standing static balance, and standing dynamic balance reactions in typically developing children.* Gait Posture, 2019. **73**: p. 20-25.
59. Witherington, D.C., et al., *The development of anticipatory postural adjustments in infancy.* Infancy, 2002. **3**(4): p. 495-517.
60. Ledebt, A., B. Bril, and Y. Brenière, *The build-up of anticipatory behaviourAn analysis of the development of gait initiation in children.* Experimental Brain Research, 1998. **120**(1): p. 9-17.
61. Adolph, K.E., et al., *How do you learn to walk? Thousands of steps and dozens of falls per day.* Psychological science, 2012. **23**(11): p. 1387-1394.
62. Hägglund, G., H. Lauge-Pedersen, and P. Wagner, *Characteristics of children with hip displacement in cerebral palsy.* BMC musculoskeletal disorders, 2007. **8**(1): p. 101.
63. Rose, J., et al., *Postural balance in children with cerebral palsy.* Dev Med Child Neurol, 2002. **44**(1): p. 58-63.
64. Lidbeck, C.M., et al., *Postural orientation during standing in children with bilateral cerebral palsy.* Pediatr Phys Ther, 2014. **26**(2): p. 223-9.
65. Galli, M., et al., *Foot pressure distribution in children with cerebral palsy while standing.* Res Dev Disabil, 2015. **41-42**: p. 52-7.
66. Abd El-Nabie, W.A.E. and M.S.M. Saleh, *Trunk and pelvic alignment in relation to postural control in children with cerebral palsy.* J Back Musculoskelet Rehabil, 2019. **32**(1): p. 125-130.
67. Domagalska-Szopa, M. and A. Szopa, *Postural orientation and standing postural alignment in ambulant children with bilateral cerebral palsy.* Clin Biomech (Bristol, Avon), 2017. **49**: p. 22-27.
68. Donker, S.F., et al., *Children with cerebral palsy exhibit greater and more regular postural sway than typically developing children.* Exp Brain Res, 2008. **184**(3): p. 363-70.
69. Ferdjallah, M., et al., *Analysis of postural control synergies during quiet standing in healthy children and children with cerebral palsy.* Clin Biomech (Bristol, Avon), 2002. **17**(3): p. 203-10.
70. Woollacott, M.H. and A. Shumway-Cook, *Postural Dysfunction During Standing and Walking in Children With Cerebral Palsy: What are the Underlying Problems and What New Therapies Might Improve Balance?* Neural Plasticity, 2005. **12**(2-3): p. 211-219.
71. Burtner, P.A., et al., *The capacity to adapt to changing balance threats: a comparison of children with cerebral palsy and typically developing children.* Dev Neurorehabil, 2007. **10**(3): p. 249-60.
72. Woollacott, M.H. and A. Shumway-Cook, *Postural dysfunction during standing and walking in children with cerebral palsy: what are the underlying problems and what new therapies might improve balance?* Neural Plast, 2005. **12**(2-3): p. 211-9; discussion 263-72.
73. Roncesvalles, M.N., M.W. Woollacott, and P.A. Burtner, *Neural factors underlying reduced postural adaptability in children with cerebral palsy.* Neuroreport, 2002. **13**(18): p. 2407-10.

74. Stackhouse, C., et al., *Gait initiation in children with cerebral palsy.* Gait Posture, 2007. **26**(2): p. 301-8.
75. Feng, J., et al., *Motion of the center of mass in children with spastic hemiplegia: balance, energy transfer, and work performed by the affected leg vs. the unaffected leg.* Gait Posture, 2014. **39**(1): p. 570-6.
76. Russell, D.J., et al., *Gross motor function measure (GMFM-66 & GMFM-88) users manual*. 2002: Mac keith press.
77. Franjoine, M.R., J.S. Gunther, and M.J. Taylor, *Pediatric Balance Scale: A Modified Version of the Berg Balance Scale for the School-Age Child with Mild to Moderate Motor Impairment.* Pediatric Physical Therapy, 2003. **15**(2): p. 114-128.
78. Franchignoni, F., et al., *Using psychometric techniques to improve the Balance Evaluation System's Test: the mini-BESTest.* Journal of rehabilitation medicine: official journal of the UEMS European Board of Physical and Rehabilitation Medicine, 2010. **42**(4): p. 323.
79. Kim, D.H., D.H. An, and W.G. Yoo, *Reliability, standard error of measurement, and minimal detectable change of the star excursion balance test in children with cerebral palsy.* J Back Musculoskelet Rehabil, 2020. **33**(6): p. 909-912.
80. Thompson, P., et al., *Test–retest reliability of the 10-metre fast walk test and 6-minute walk test in ambulatory school-aged children with cerebral palsy.* Developmental Medicine & Child Neurology, 2008. **50**(5): p. 370-376.
81. Bernhardt, J., et al., *Moving rehabilitation research forward: developing consensus statements for rehabilitation and recovery research.* Neurorehabilitation and neural repair, 2017. **31**(8): p. 694-698.
82. Bernhardt, J., et al., *Setting the scene for the second stroke recovery and rehabilitation roundtable.* International Journal of Stroke, 2019. **14**(5): p. 450-456.
83. Tustin, K., et al., *Rater reliability and scoring duration of the Quality Function Measure in ambulant children with hyperkinetic movement disorders.* Developmental Medicine & Child Neurology, 2016. **58**(8): p. 822-828.
84. Saether, R., et al., *Clinical tools to assess balance in children and adults with cerebral palsy: a systematic review.* Dev Med Child Neurol, 2013. **55**(11): p. 988-99.
85. Rapson, R., et al., *A cross sectional study investigating dynamic balance when stepping to targets in children with cerebral palsy compared to typically developing children.* Gait & Posture, 2023.
86. Pavão, S.L., et al., *Functionality level and its relation to postural control during sitting-to-stand movement in children with cerebral palsy.* Research in Developmental Disabilities, 2014. **35**(2): p. 506-511.
87. Hof, A.L., M.G. Gazendam, and W.E. Sinke, *The condition for dynamic stability.* J Biomech, 2005. **38**(1): p. 1-8.
88. Yiou, E., et al., *Balance control during gait initiation: State-of-the-art and research perspectives.* World J Orthop, 2017. **8**(11): p. 815-828.
89. Lyon, I.N. and B.L. Day, *Predictive control of body mass trajectory in a two-step sequence.* Exp Brain Res, 2005. **161**(2): p. 193-200.
90. Haugh, A.B., A.D. Pandyan, and G.R. Johnson, *A systematic review of the Tardieu Scale for the measurement of spasticity.* Disabil Rehabil, 2006. **28**(15): p. 899-907.

91. Russell, D.J., et al., *Development and validation of item sets to improve efficiency of administration of the 66-item Gross Motor Function Measure in children with cerebral palsy.* Dev Med Child Neurol, 2010. **52**(2): p. e48-54.
92. O'Connor, C.M., et al., *Automatic detection of gait events using kinematic data.* Gait Posture, 2007. **25**(3): p. 469-74.
93. Weisstein, E.W., *Bonferroni correction.* https://mathworld. wolfram. com/, 2004.
94. Von Elm, E., et al., *The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement: guidelines for reporting observational studies.* Bulletin of the World Health Organization, 2007. **85**: p. 867-872.
95. Delafontaine, A., et al., *Anticipatory Postural Adjustments During Gait Initiation in Stroke Patients.* Front Neurol, 2019. **10**: p. 352.
96. Brunt, D., D.W. Vander Linden, and A.L. Behrman, *The relation between limb loading and control parameters of gait initiation in persons with stroke.* Archives of physical medicine and rehabilitation, 1995. **76**: p. 627-634.
97. Rogers, M.W. and Y.C. Pai, *Organization of preparatory postural responses for the initiation of lateral body motion during goal directed leg movements.* Neuroscience letters, 1995. **187**: p. 99-102.
98. Boyer, E.R. and A. Patterson, *Gait pathology subtypes are not associated with self-reported fall frequency in children with cerebral palsy.* Gait & Posture, 2018. **63**: p. 189-194.
99. Dhote, S.N., P.A. Khatri, and S.S. Ganvir, *Reliability of "Modified timed up and go" test in children with cerebral palsy.* Journal of pediatric neurosciences, 2012. **7**(2): p. 96.
100. Russell, D.J., et al., *THE GROSS MOTOR FUNCTION MEASURE: A MEANS TO EVALUATE THE EFFECTS OF PHYSICAL THERAPY.* Developmental Medicine & Child Neurology, 1989. **31**(3): p. 341-352.
101. Gan, S.-M., et al., *Psychometric properties of functional balance assessment in children with cerebral palsy.* Neurorehabilitation and neural repair, 2008. **22**(6): p. 745-753.
102. Lyon, I.N. and B.L. Day, *Control of frontal plane body motion in human stepping.* Exp Brain Res, 1997. **115**(2): p. 345-56.
103. Bancroft, M.J. and B.L. Day, *The throw-and-catch model of human gait: evidence from coupling of pre-step postural activity and step location.* Frontiers in human neuroscience, 2016. **10**: p. 635.
104. Yiou, E., et al., *Balance control during gait initiation: State-of-the-art and research perspectives.* World journal of orthopedics, 2017. **8**(11): p. 815-828.
105. Fitzpatrick, P., *Evaluating patient-based outcome measures for use in clinical trials.* Health Technol Assessment, 1998. **2**: p. 14.
106. Saini, M., et al., *The vertical displacement of the center of mass during walking: a comparison of four measurement methods.* 1998.
107. Huntley, A.H., et al., *Validation of simplified centre of mass models during gait in individuals with chronic stroke.* Clinical Biomechanics, 2017. **48**: p. 97-102.
108. Shrout, P.E. and J.L. Fleiss, *Intraclass correlations: uses in assessing rater reliability.* Psychological bulletin, 1979. **86**(2): p. 420.

109. Bland, J.M. and D.G. Altman, *Measuring agreement in method comparison studies.* Statistical methods in medical research, 1999. **8**(2): p. 135-160.
110. Koo, T.K. and M.Y. Li, *A Guideline of Selecting and Reporting Intraclass Correlation Coefficients for Reliability Research.* Journal of Chiropractic Medicine, 2016. **15**(2): p. 155-163.
111. Bland, J.M. and D.G. Altman, *Measuring agreement in method comparison studies.* Stat Methods Med Res, 1999. **8**(2): p. 135-60.
112. Weir, J.P., *Quantifying test-retest reliability using the intraclass correlation coefficient and the SEM.* The Journal of Strength & Conditioning Research, 2005. **19**(1): p. 231-240.
113. Ko, J.-Y. and G.-W. Kim, *Test-retest, inter-rater, and intra-rater reliability of a pediatric balance scale in children with cerebral palsy.* The Journal of Korean Physical Therapy, 2010. **22**(4): p. 43-48.
114. Chen, C.-l., et al., *Validity, responsiveness, minimal detectable change, and minimal clinically important change of Pediatric Balance Scale in children with cerebral palsy.* Research in Developmental Disabilities, 2013. **34**(3): p. 916-922.
115. Franjoine, M.R., et al., *The performance of children developing typically on the pediatric balance scale.* Pediatric physical therapy, 2010. **22**(4): p. 350-359.
116. Carey, H., et al., *Reliability and Responsiveness of the Timed Up and Go Test in Children With Cerebral Palsy.* Pediatric Physical Therapy, 2016. **28**(4): p. 401-408.
117. N, D.S. and K.P. A, *Intra-rater reliability of timed 'up and go' test for children diagnosed with cerebral palsy.* International Journal of Therapy and Rehabilitation, 2012. **19**(10): p. 575-580.
118. Nicolini-Panisson, R.D.A. and M.V.F. Donadio, *Timed" Up & Go" test in children and adolescents.* Revista Paulista de Pediatria, 2013. **31**: p. 377-383.
119. Ko, J. and M. Kim, *Reliability and Responsiveness of the Gross Motor Function Measure-88 in Children With Cerebral Palsy.* Physical Therapy, 2013. **93**(3): p. 393-400.
120. Brand, R.A., *Can biomechanics contribute to clinical orthopaedic assessments?* The Iowa orthopaedic journal, 1989. **9**: p. 61.
121. Rapson, R., et al., *Defining usual physiotherapy care in ambulant children with cerebral palsy in the United Kingdom: A mixed methods consensus study.* Child: Care, Health and Development, 2022. **48**(5): p. 708-723.
122. Kimoto, M., et al., *Effect of home-based training focused on increasing maximum step length in walking function of children with cerebral palsy.* Physical Therapy Reviews, 2019. **24**(6): p. 358-365.
123. Kamp, F.A., et al., *Energy cost of walking in children with spastic cerebral palsy: relationship with age, body composition and mobility capacity.* Gait & posture, 2014. **40**(1): p. 209-214.
124. Rosenbaum P Fau - Paneth, N., et al., *A report: the definition and classification of cerebral palsy April 2006.* Dev Med Child Neurol Suppl, 2007. **109**: p. 8-14.
125. Palisano, R., et al., *Gross motor function classification system for cerebral palsy.* Dev Med Child Neurol, 1997. **39**(4): p. 214-23.
126. Hanna, S.E., et al., *Reference Curves for the Gross Motor Function Measure: Percentiles for Clinical Description and Tracking Over Time*

Among Children With Cerebral Palsy. Physical Therapy, 2008. **88**(5): p. 596-607.
127. Palisano, R.J., W.P. Copeland, and B.E. Galuppi, *Performance of physical activities by adolescents with cerebral palsy.* Physical Therapy, 2007. **87**(1): p. 77-87.
128. Morris, C., et al., *Setting research priorities to improve the health of children and young people with neurodisability: a British Academy of Childhood Disability-James Lind Alliance Research Priority Setting Partnership.* BMJ Open, 2015. **5**(1): p. e006233.
129. Novak, I., et al., *A systematic review of interventions for children with cerebral palsy: state of the evidence.* Developmental Medicine & Child Neurology, 2013. **55**(10): p. 885-910.
130. Franki, I., et al., *The evidence-base for basic physical therapy techniques targeting lower limb function in children with cerebral palsy: a systematic review using the International Classification of Functioning, Disability and Health as a conceptual framework.* Journal of rehabilitation medicine : official journal of the UEMS European Board of Physical and Rehabilitation Medicine, 2012. **44**(5): p. 385-395.
131. Hägglund, G., et al., *Prevention of dislocation of the hip in children with cerebral palsy: 20-year results of a population-based prevention programme.* The bone & joint journal, 2014. **96**(11): p. 1546-1552.
132. Mugglestone, M.A., P. Eunson, and M.S. Murphy, *GUIDELINES: Spasticity in children and young people with non-progressive brain disorders: summary of NICE guidance.* BMJ: British Medical Journal, 2012. **345**(7868): p. 43-45.
133. Medicine, O.C.f.E.-B., *OCEBM Levels of Evidence Working Group. The Oxford 2011 levels of evidence.* 2011.
134. Physicians, R.C.o., *National Clinical Guideline for Stroke- SSNAP.* 2016 **5th edition**.
135. Harvey, N. and C.A. Holmes, *Nominal group technique: An effective method for obtaining group consensus.* International Journal of Nursing Practice, 2012. **18**(2): p. 188-194.
136. Potter, M., S. Gordon, and P. Hamer, *The Nominal Group Technique: A useful consensus methodology in physiotherapy research*. Vol. 32. 2004.
137. Allen, J., J. Dyas, and M. Jones, *Building consensus in health care: a guide to using the nominal group technique.* British Journal of Community Nursing, 2004. **9**(3): p. 110-114.
138. Ven, A.H.V.D. and A.L. Delbecq, *The effectiveness of nominal, Delphi, and interacting group decision making processes.* Academy of management Journal, 1974. **17**(4): p. 605-621.
139. Bartlett, D.J. and R.J. Palisano, *Physical therapists' perceptions of factors influencing the acquisition of motor abilities of children with cerebral palsy: implications for clinical reasoning.* Physical therapy, 2002. **82**(3): p. 237-248.
140. Gustafson, D.H., A.L. Delbecq, and A.H. Van de Ven, *Group Techniques for Program Planning-A Guide to Nominal Group and Delphi Processes.* 1986.
141. Moher, D., et al., *PRISMA statement.* Epidemiology, 2011. **22**(1): p. 128.
142. Hannes, K., C. Lockwood, and A. Pearson, *A comparative analysis of three online appraisal instruments' ability to assess validity in qualitative research.* Qualitative health research, 2010. **20**(12): p. 1736-1743.

143. Elnahhas, A.M., S. Elshennawy, and M.G. Aly, *Effects of backward gait training on balance, gross motor function, and gait in children with cerebral palsy: a systematic review.* Clinical Rehabilitation, 2019. **33**(1): p. 3-12.
144. Booth, A.T.C., et al., *The efficacy of functional gait training in children and young adults with cerebral palsy: a systematic review and meta-analysis.* Dev Med Child Neurol, 2018. **60**(9): p. 866-883.
145. Novak, I., et al., *State of the Evidence Traffic Lights 2019: Systematic Review of Interventions for Preventing and Treating Children with Cerebral Palsy.* Current Neurology and Neuroscience Reports, 2020. **20**(2): p. 3.
146. Bania, T., H.-C. Chiu, and E. Billis, *Activity training on the ground in children with cerebral palsy: Systematic review and meta-analysis.* Physiotherapy theory and practice, 2019. **35**(9): p. 810-821.
147. Dewar, R., S. Love, and L.M. Johnston, *Exercise interventions improve postural control in children with cerebral palsy: a systematic review.* Developmental Medicine & Child Neurology, 2015. **57**(6): p. 504-520.
148. Clutterbuck, G., M. Auld, and L. Johnston. *Active exercise interventions improve gross motor function of ambulant/semi ambulant school-aged children with cerebral palsy: A systematic review*. in *Australasian academy of cerebral palsy and developmental medicine*. 2018.
149. Corsi, C., et al., *Effect of physical therapy interventions on spatiotemporal gait parameters in children with cerebral palsy: a systematic review.* Disability and rehabilitation, 2019: p. 1-10.
150. Ritzmann, R., C. Stark, and A. Krause, *Vibration therapy in patients with cerebral palsy: a systematic review.* Neuropsychiatr Dis Treat, 2018. **14**: p. 1607-1625.
151. Galey, S.A., et al., *Effectiveness of surgical and non-surgical management of crouch gait in cerebral palsy: A systematic review.* Gait Posture, 2017. **54**: p. 93-105.
152. Moreau, N.G., et al., *Effectiveness of Rehabilitation Interventions to Improve Gait Speed in Children With Cerebral Palsy: Systematic Review and Meta-analysis.* Physical Therapy, 2016. **96**(12): p. 1938-1954.
153. Franki, I., et al., *The evidence-base for basic physical therapy techniques targeting lower limb function in children with cerebral palsy: a systematic review using the International Classification of Functioning, Disability and Health as a conceptual framework.* J Rehabil Med, 2012. **44**(5): p. 385-95.
154. Anttila, H., et al., *Effectiveness of physical therapy interventions for children with cerebral palsy: a systematic review.* BMC Pediatr, 2008. **8**: p. 14.
155. Tustin, K. and A. Patel, *A Critical Evaluation of the Updated Evidence for Casting for Equinus Deformity in Children with Cerebral Palsy.* Physiother Res Int, 2017. **22**(1).
156. Valentín-Gudiol, M., et al., *Treadmill interventions in children under six years of age at risk of neuromotor delay.* Cochrane Database of Systematic Reviews, 2017(7).
157. Salem, Y., et al., *Effects of prolonged standing on gait in children with spastic cerebral palsy.* Physical & Occupational Therapy in Pediatrics, 2010. **30**(1): p. 54-65.
158. Balshem, H., et al., *GRADE guidelines: 3. Rating the quality of evidence.* Journal of Clinical Epidemiology, 2011. **64**(4): p. 401-406.

159. Gunn, H., et al., *Development of a balance, safe mobility and falls management programme for people with multiple sclerosis.* Disabil Rehabil, 2018. **40**(24): p. 2857-2866.
160. Tustin, K. and A. Patel, *A Critical Evaluation of the Updated Evidence for Casting for Equinus Deformity in Children with Cerebral Palsy.* Physiotherapy Research International, 2017. **22**(1): p. n/a-N.PAG.
161. Wordie, S.J., et al., *Hip displacement and dislocation in a total population of children with cerebral palsy in Scotland: status after five years' hip surveillance.* The Bone & Joint Journal, 2020. **102**(3): p. 383-387.
162. Tutt, R. and P. Williams, *The SEND Code of Practice 0-25 Years: Policy, Provision and Practice*. 2015: Sage.
163. Organization, W.H., *International Classification of Functioning, Disability, and Health: Children & Youth Version: ICF-CY*. 2007: World Health Organization.
164. Scholtes, V.A., et al., *Effect of Multilevel Botulinum Toxin A and Comprehensive Rehabilitation on Gait in Cerebral Palsy.* Pediatric Neurology, 2007. **36**(1): p. 30-39.
165. Scholtes, V.A., et al., *Effectiveness of functional progressive resistance exercise training on walking ability in children with cerebral palsy: a randomized controlled trial.* Research in Developmental Disabilities, 2012. **33**(1): p. 181-188.
166. Mugglestone, M.A., P. Eunson, and M.S. Murphy, *Spasticity in children and young people with non-progressive brain disorders: summary of NICE guidance.* Bmj, 2012. **345**.
167. Bleyenheuft, Y., et al., *Hand and arm bimanual intensive therapy including lower extremity (HABIT-ILE) in children with unilateral spastic cerebral palsy: a randomized trial.* Neurorehabilitation and neural repair, 2015. **29**(7): p. 645-657.
168. Rapson, R., et al., *Multicentre, randomised controlled feasibility study to compare a 10-week physiotherapy programme using an interactive exercise training device to improve walking and balance, to usual care of children with cerebral palsy aged 4–18 years: the ACCEPT study protocol.* BMJ Open, 2022. **12**(5): p. e058916.
169. Morgan, P., R. McDonald, and J. McGinley, *Perceived cause, environmental factors, and consequences of falls in adults with cerebral palsy: a preliminary mixed methods study.* Rehabilitation research and practice, 2015. **2015**.
170. Morgan, P. and J. McGinley, *Performance of adults with cerebral palsy related to falls, balance and function: A preliminary report.* Developmental neurorehabilitation, 2013. **16**(2): p. 113-120.
171. Morgan, P.E., S.-E. Soh, and J.L. McGinley, *Health-related quality of life of ambulant adults with cerebral palsy and its association with falls and mobility decline: a preliminary cross sectional study.* Health and quality of life outcomes, 2014. **12**(1): p. 132.
172. van der Heide, J.C. and M. Hadders-Algra, *Postural muscle dyscoordination in children with cerebral palsy.* Neural Plasticity, 2005. **12**(2-3): p. 197-203.
173. Rosenbaum, P.L., et al., *Development of the gross motor function classification system for cerebral palsy.* Developmental medicine and child neurology, 2008. **50**(4): p. 249.
174. Curtis, D.J., J. Bencke, and B. Mygind, *The effect of training in an interactive dynamic stander on ankle dorsiflexion and gross motor*

function in children with cerebral palsy. Developmental neurorehabilitation, 2014. **17**(6): p. 393-397.
175. Lyon, I.N. and B.L. Day, *Predictive control of body mass trajectory in a two-step sequence.* Experimental brain research, 2005. **161**(2): p. 193-200.
176. Boyd, R.N. and H.K. Graham, *Objective measurement of clinical findings in the use of botulinum toxin type A for the management of children with cerebral palsy.* European Journal of Neurology, 1999. **6**: p. s23-s35.
177. Law, M., et al., *The Canadian occupational performance measure: an outcome measure for occupational therapy.* Can J Occup Ther, 1990. **57**(2): p. 82-7.
178. Canaway, A.G. and E.J. Frew, *Measuring preference-based quality of life in children aged 6–7 years: a comparison of the performance of the CHU-9D and EQ-5D-Y—the WAVES Pilot Study.* Quality of Life Research, 2013. **22**(1): p. 173-183.
179. Thabane, L., et al., *A tutorial on pilot studies: the what, why and how.* BMC Medical Research Methodology, 2010. **10**(1): p. 1.
180. Her, J.-G., J.-H. Woo, and J. Ko, *Reliability of the Pediatric Balance Scale in the Assessment of the Children with Cerebral Palsy.* Journal of Physical Therapy Science, 2012. **24**(4): p. 301-305.
181. Sivan, M., et al., *Home-based Computer Assisted Arm Rehabilitation (hCAAR) robotic device for upper limb exercise after stroke: results of a feasibility study in home setting.* Journal of NeuroEngineering and Rehabilitation, 2014. **11**(1): p. 163.
182. Holland, A.E., et al., *Telerehabilitation for People with Chronic Obstructive Pulmonary Disease: Feasibility of a Simple, Real Time Model of Supervised Exercise Training.* Journal of Telemedicine and Telecare, 2013. **19**(4): p. 222-226.
183. Lopes, S., et al., *Games used with serious purposes: a systematic review of interventions in patients with cerebral palsy.* Frontiers in psychology, 2018. **9**: p. 1712.
184. C. Bryanton, J.B., M. Brien, J. Mclean, A. McCormick, and Dr. H. Sveistru, *Feasibility, Motivation, and Selective Motor Control: Virtual Reality Compared to Conventional Home Exercise in Children with Cerebral Palsy.* CyberPsychology & Behavior, 2006. **9**(2): p. 123-128.
185. Deutsch, J.E., et al., *Use of a low-cost, commercially available gaming console (Wii) for rehabilitation of an adolescent with cerebral palsy.* Physical therapy, 2008. **88**(10): p. 1196-1207.
186. Sajan, J.E., et al., *Wii-based interactive video games as a supplement to conventional therapy for rehabilitation of children with cerebral palsy: a pilot, randomized controlled trial.* Developmental neurorehabilitation, 2017. **20**(6): p. 361-367.
187. Cleary, K., et al. *PedBotHome: Robotically-assisted ankle rehabilitation system for children with cerebral palsy*. in *2019 IEEE 16th international conference on rehabilitation robotics (ICORR)*. 2019. IEEE.
188. Llamas-Ramos, R., J.L. Sánchez-González, and I. Llamas-Ramos, *Robotic systems for the physiotherapy treatment of children with cerebral palsy: a systematic review.* International journal of environmental research and public health, 2022. **19**(9): p. 5116.
189. Ravi, D., N. Kumar, and P. Singhi, *Effectiveness of virtual reality rehabilitation for children and adolescents with cerebral palsy: an*

updated evidence-based systematic review. Physiotherapy, 2017. **103**(3): p. 245-258.
190. Bryanton, C., et al., *Feasibility, motivation, and selective motor control: virtual reality compared to conventional home exercise in children with cerebral palsy.* Cyberpsychology & behavior, 2006. **9**(2): p. 123-128.
191. Standen, P.J., et al., *Patients' Use of a Home-Based Virtual Reality System to Provide Rehabilitation of the Upper Limb Following Stroke.* Physical Therapy, 2015. **95**(3): p. 350-359.
192. Goodwin, J., et al., *Understanding frames: A qualitative exploration of standing frame use for young people with cerebral palsy in educational settings.* Child: Care, Health and Development, 2019. **45**(3): p. 433-439.
193. Proffitt, R., *Gamification in Rehabilitation: Finding the "Just-Right-Challenge"*, in *Handbook of Research on Holistic Perspectives in Gamification for Clinical Practice*. 2016, IGI global. p. 132-157.
194. Bonnechere, B., et al., *Can serious games be incorporated with conventional treatment of children with cerebral palsy? A review.* Research in developmental disabilities, 2014. **35**(8): p. 1899-1913.
195. Mitchell, T.K., et al., *'It doesn't feel like our house anymore': The impact of medical technology upon life at home for families with a medically complex, technology-dependent child.* Health & Place, 2022. **74**: p. 102768.
196. Goodwin, J., et al., *Understanding frames: A qualitative study of young people's experiences of using standing frames as part of postural management for cerebral palsy.* Child: Care, Health and Development, 2018. **44**(2): p. 203-211.
197. Damiano, D.L., *Activity, Activity, Activity: Rethinking Our Physical Therapy Approach to Cerebral Palsy.* Physical Therapy, 2006. **86**(11): p. 1534-1540.
198. Hadders-Algra, M., *The neuronal group selection theory: a framework to explain variation in normal motor development.* Developmental medicine and child neurology, 2000. **42**(8): p. 566-572.
199. Hadders-Algra, M., *Early Brain Damage and the Development of Motor Behavior in Children: Clues for Therapeutic Intervention?* Neural Plasticity, 2001. **8**: p. 813609.
200. Hall, A.M., et al., *The influence of the therapist-patient relationship on treatment outcome in physical rehabilitation: a systematic review.* Physical therapy, 2010. **90**(8): p. 1099-1110.
201. Kunz, R., et al., *A systematic review finds that methodological quality is better than its reputation but can be improved in physiotherapy trials in childhood cerebral palsy.* Journal of clinical epidemiology, 2006. **59**(12): p. 1239. e1-1239. e12.
202. Fowler, E.G., et al., *Selective Control Assessment of the Lower Extremity (SCALE): development, validation, and interrater reliability of a clinical tool for patients with cerebral palsy.* Dev Med Child Neurol, 2009. **51**(8): p. 607-14.
203. Hasan, C.Z.C., et al., *The analysis of three-dimensional ground reaction forces during gait in children with autism spectrum disorders.* Research in Developmental Disabilities, 2017. **66**: p. 55-63.
204. Bruijn, S.M., et al., *Gait stability in children with Cerebral Palsy.* Research in Developmental Disabilities, 2013. **34**(5): p. 1689-1699.
205. Balemans, A.C.J. and E.A.M. Bolster, *Aerobic and Anaerobic Fitness in Children and Youth with Cerebral Palsy*, in *Cerebral Palsy*, F. Miller, et

al., Editors. 2020, Springer International Publishing: Cham. p. 2687-2708.
206. Ettema, S., et al., *The effect of prolonged walking on muscle fatigue and neuromuscular control in children with cerebral palsy.* Gait & Posture, 2022. **93**: p. 7-13.
207. Brunton, L.K., P.G. McPhee, and J.W. Gorter, *Self-reported factors contributing to fatigue and its management in adolescents and adults with cerebral palsy.* Disabil Rehabil, 2021. **43**(7): p. 929-935.
208. Lust, J.M., et al., *Motor planning in children with cerebral palsy: A longitudinal perspective.* J Clin Exp Neuropsychol, 2018. **40**(6): p. 559-566.
209. McCormick, A.M., et al., *A randomized, cross-over trial comparing the effect of innovative robotic gait training and functional clinical therapy in children with cerebral palsy; a protocol to test feasibility.* Contemp Clin Trials, 2023. **127**: p. 107086.
210. Beveridge, B., et al., *"You gotta try it all": Parents' Experiences with Robotic Gait Training for their Children with Cerebral Palsy.* Phys Occup Ther Pediatr, 2015. **35**(4): p. 327-41.
211. Carter, B. and K. Ford, *Researching children's health experiences: The place for participatory, child-centered, arts-based approaches.* Research in Nursing & Health, 2013. **36**(1): p. 95-107.
212. Guest, G., A. Bunce, and L. Johnson, *How Many Interviews Are Enough?:An Experiment with Data Saturation and Variability.* Field Methods, 2006. **18**(1): p. 59-82.
213. Braun, V. and V. Clarke, *To saturate or not to saturate? Questioning data saturation as a useful concept for thematic analysis and sample-size rationales.* Qualitative Research in Sport, Exercise and Health, 2021. **13**(2): p. 201-216.
214. Kortesluoma, R.-L., M. Hentinen, and M. Nikkonen, *Conducting a qualitative child interview: methodological considerations.* Journal of Advanced Nursing, 2003. **42**(5): p. 434-441.
215. Almoajil, H., et al., *A core outcome set for lower limb orthopaedic surgery for children with cerebral palsy: An international multi-stakeholder consensus study.* Developmental Medicine & Child Neurology, 2023. **65**(2): p. 254-263.
216. Gray, C.A. and J.D. Garand, *Social stories: Improving responses of students with autism with accurate social information.* Focus on autistic behavior, 1993. **8**(1): p. 1-10.
217. Christensen, D., et al., *Prevalence of cerebral palsy, co-occurring autism spectrum disorders, and motor functioning – Autism and Developmental Disabilities Monitoring Network, USA, 2008.* Developmental Medicine & Child Neurology, 2014. **56**(1): p. 59-65.
218. Rapson, R., et al., *A cross sectional study investigating dynamic balance when stepping to targets in children with cerebral palsy compared to typically developing children.* Gait & Posture, 2023. **101**: p. 154-159.
219. Lyon, I. and B. Day, *Control of frontal plane body motion in human stepping.* Experimental Brain Research, 1997. **115**: p. 345-356.
220. Rapson, R., Marsden, J., *Intra-rater reliability of clinical measures of leg function, in typically developing children aged 1-4 years.* Association of Paediatric Chartered Physiotherapists, 2016. **16**(1): p. 14-22.

221. Nonnekes, J.H., et al., *Deficits underlying impaired visually triggered step adjustments in mildly affected stroke patients.* Neurorehabilitation and neural repair, 2010. **24**(4): p. 393-400.
222. Puce, L., et al., *Systematic Review of Fatigue in Individuals With Cerebral Palsy.* Frontiers in Human Neuroscience, 2021. **15**.
223. Kurz, M.J., et al., *Neurophysiological abnormalities in the sensorimotor cortices during the motor planning and movement execution stages of children with cerebral palsy.* Developmental Medicine & Child Neurology, 2014. **56**(11): p. 1072-1077.
224. Gaston, M.S., *CPIPS: musculoskeletal and hip surveillance for children with cerebral palsy.* Paediatrics and Child Health, 2019. **29**(11): p. 489-494.
225. Law, M., et al., *The Canadian occupational performance measure: an outcome measure for occupational therapy.* Canadian Journal of Occupational Therapy, 1990. **57**(2): p. 82-87.
226. Novak, I., *Therapy for children with cerebral palsy: who, what, and how much?* Developmental Medicine & Child Neurology, 2020. **62**(1): p. 17-17.
227. Dan, B., *Intensive repetitive motor training: how does it work in children with cerebral palsy?* Dev Med Child Neurol, 2021. **63**(9): p. 1008.
228. Kolb, B., A. Harker, and R. Gibb, *Principles of plasticity in the developing brain.* Developmental Medicine & Child Neurology, 2017. **59**(12): p. 1218-1223.
229. Dan, B., *Neuroscience underlying rehabilitation: what is neuroplasticity?* 2019, Wiley Online Library. p. 1240-1240.
230. Reid, L.B., S.E. Rose, and R.N. Boyd, *Rehabilitation and neuroplasticity in children with unilateral cerebral palsy.* Nature Reviews Neurology, 2015. **11**(7): p. 390-400.
231. Blauw-Hospers, C.H. and M. Hadders-Algra, *A systematic review of the effects of early intervention on motor development.* Developmental medicine and child neurology, 2005. **47**(6): p. 421-432.
232. de Almeida Carvalho Duarte, N., et al., *Motor Cortex Plasticity in Children With Spastic Cerebral Palsy: A Systematic Review.* J Mot Behav, 2017. **49**(4): p. 355-364.
233. Noble, J.J., M. Gough, and A.P. Shortland, *Selective motor control and gross motor function in bilateral spastic cerebral palsy.* Developmental Medicine & Child Neurology, 2019. **61**(1): p. 57-61.
234. Pandyan, A.D., et al., *Spasticity: Clinical perceptions, neurological realities and meaningful measurement.* Disability and Rehabilitation, 2005. **27**(1-2): p. 2-6.
235. Mohammed, A.H., et al., *Correlation between Selective Motor Control of the Lower Extremities and Balance in Spastic Hemiplegic Cerebral Palsy: a randomized controlled trial.* BMC Sports Sci Med Rehabil, 2023. **15**(1): p. 24.
236. Yun, G., et al., *Selective motor control correlates with gross motor ability, functional balance and gait performance in ambulant children with bilateral spastic cerebral palsy.* Gait & Posture, 2023. **99**: p. 9-13.
237. Sanger, T.D., et al., *Definition and classification of negative motor signs in childhood.* Pediatrics, 2006. **118**(5): p. 2159-2167.
238. Amirthalingam, J., et al., *Virtual Reality Intervention to Help Improve Motor Function in Patients Undergoing Rehabilitation for Cerebral Palsy,*

Parkinson's Disease, or Stroke: A Systematic Review of Randomized Controlled Trials. Cureus, 2021. **13**(7): p. e16763.
239. Feitosa, J.A., et al., *Effects of virtual reality-based motor rehabilitation: a systematic review of fMRI studies.* J Neural Eng, 2022. **19**(1).
240. Cooper, T. and J.M. Williams, *Does an exercise programme integrating the Nintendo Wii-Fit Balance Board improve balance in ambulatory children with cerebral palsy?* Physical Therapy Reviews, 2017. **22**(5/6): p. 229-237.
241. Gatica-Rojas, V., et al., *Does Nintendo Wii Balance Board improve standing balance? A randomized controlled trial in children with cerebral palsy.* Eur J Phys Rehabil Med, 2017. **53**(4): p. 535-544.
242. Chiu, H.C., L. Ada, and S.D. Lee, *Balance and mobility training at home using Wii Fit in children with cerebral palsy: a feasibility study.* BMJ Open, 2018. **8**(5): p. e019624.
243. Lefmann, S., R. Russo, and S. Hillier, *The effectiveness of robotic-assisted gait training for paediatric gait disorders: systematic review.* J Neuroeng Rehabil, 2017. **14**(1): p. 1.
244. Wallard, L., et al., *Effect of robotic-assisted gait rehabilitation on dynamic equilibrium control in the gait of children with cerebral palsy.* Gait & Posture, 2018. **60**: p. 55-60.
245. Lafrenière, M.-A.K., J. Verner-Filion, and R.J. Vallerand, *Development and validation of the Gaming Motivation Scale (GAMS).* Personality and individual differences, 2012. **53**(7): p. 827-831.
246. *Motivations for Play in Online Games.* CyberPsychology & Behavior, 2006. **9**(6): p. 772-775.
247. Party, I.S.W., *National clinical guideline for stroke*. 2012, London: Royal College of Physicians.
248. NICE, N.I.f.H.a.C.E. *Spasticity in under 19s: management*. NICE Clinical Guidelines 2012 29/11/2016 05/01/2022

]; Available from: https://www.nice.org.uk/guidance/cg145/chapter/1-Guidance#oral-drugs.
249. Fowler, E.G., et al., *Selective Control Assessment of the Lower Extremity (SCALE): development, validation, and interrater reliability of a clinical tool for patients with cerebral palsy.* Developmental Medicine & Child Neurology, 2009. **51**(8): p. 607-614.
250. Rimmer, J.H., *Getting Beyond the Plateau: Bridging the Gap Between Rehabilitation and Community-Based Exercise.* PM&R, 2012. **4**(11): p. 857-861.
251. Graham, D., et al., *Single-level selective dorsal rhizotomy for spastic cerebral palsy.* J Spine Surg, 2016. **2**(3): p. 195-201.
252. Eldridge, S.M., et al., *CONSORT 2010 statement: extension to randomised pilot and feasibility trials.* bmj, 2016. **355**: p. i5239.
253. Calvert, M., et al., *Reporting of patient-reported outcomes in randomized trials: the CONSORT PRO extension.* Jama, 2013. **309**(8): p. 814-22.
254. Boutron, I., et al., *CONSORT statement for randomized trials of nonpharmacologic treatments: a 2017 update and a CONSORT extension for nonpharmacologic trial abstracts.* Annals of internal medicine, 2017. **167**(1): p. 40-47.
255. Braun, V. and V. Clarke, *Reflecting on reflexive thematic analysis.* Qualitative Research in Sport, Exercise and Health, 2019. **11**(4): p. 589-597.

Appendices

Appendix 1- Participant information sheet- The Next Step Study: What is the validity and reliability of a novel test of coordinated stepping in children with cerebral palsy aged 8-18years?

Parent/Guardian Information Sheet for Child to Participate

IRAS 247623

We would like to invite your child to take part in our research study. Before you decide whether to let them take part, we would like you to understand why the research is being done and what it will involve.

One of our team will go through this information with you and answer any questions you may have. Talk to others about the study if you wish. Take time to decide whether or not you want your child/ward to take part.

The main researcher is Rachel Rapson, who is a physiotherapist who is highly experienced in working with children. She is doing this study as part of her PhD and is working with a supervisory team of Prof Jon Marsden and Prof Jos Latour (University of Plymouth) and Prof Bernie Carter (Edge Hill University).

What is the purpose of the study?

Children with cerebral palsy frequently have difficulties with balance and walking. Some clinical tests may measure speed of walking or stamina but may not be able to show improvements in the quality of balance and walking. A measure of the quality of a movement would help professionals decide if a treatment has helped.

We have developed a new test called the 'Next Step' test. This measures the balance and coordination of children's stepping. In this research study we will compare the 'Next Step' test with other measures in children with and without cerebral palsy. We will then explore the reliability of the test results over time and between testers. Finally, we will use the 'Next Step' test in clinical interventions aimed at improving walking in children with cerebral palsy.

Why has my child/ward been chosen?

We are inviting your child as they are aged between 8-18 years old and they have typical development. If you are interested in letting them take part, a

researcher will contact you to ask you some questions about your child to see if they are eligible.

Does my child/ward have to take part?

No. It is up to you and your child (wherever possible) to decide to join the study. We will explain the study and go through this information sheet with you. If you and your child decide to take part, we will ask you to sign a consent form allowing them to participate. If your child is able to understand the research and is happy to take part and can write their name, they will be asked to sign an assent form, if they want to.

You will be given a copy of the information sheet and the signed consent/assent forms to keep for your records.
You and your child are free to withdraw at any time, without giving a reason. This will not affect the standard of care your child receives in the future.

What will my child have to do if we choose to take part?

A member of the research team will contact you and ask for more information about your child. If you decide to take part the researcher will arrange a convenient time for you and your child to come into the Human Movement Laboratory at the Peninsula Allied Health Centre, Derriford Road, Plymouth, PL6 8BH or your local physiotherapy department in Devon or Cornwall. The tests will take place over two sessions, a week apart and will take up to 60 minutes each session. You will be reimbursed for any reasonable travel expenses.

Your child's usual medication should be continued. Your child should not undertake the tests if they show any signs of infection and illness (high temperature, vomiting, and diarrhoea) on the planned day of testing or if they have shown these signs for more than 1 day in the week before the study or in between sessions.

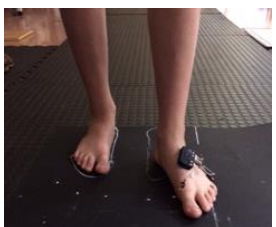
Week 1 (60 minutes)

The researcher will ask about your child's medical history, and measure their height and weight.

Next Step test- Your child will be asked to wear shorts, have bare feet and wear some electronic markers during this test. The test will be described and demonstrated by the researcher. The test involves standing and stepping to four different targets which light up. The signal from the markers is picked up by a camera in the room which allows the computer to create a 3-dimensional representation of your child's movement during stepping. After a demonstration of the test you will have time to consider whether you and your child wish to take part.

The electronic markers will be attached to your child by using soft elasticated Velcro belt and two small stick-on markers on their feet. Your child will be asked to take some practise steps to determine their usual step length. Your child's foot position will be drawn around using chalk to mark the floor to show the start position and the target position.

The photos below demonstrate the test.



Your child will be asked to follow a series of instructions, which will be said in a way that they can understand:

“A beep will signal the start of each test”

“A light will show on the floor in front of you”

“Please step forward onto the target footprint as accurately as you can”

“Bring the other foot forward to end with both feet together at the end of the test”

“The test finishes with a beep.”

“Now return to the start position”

“You can sit down and rest whenever you need to.”

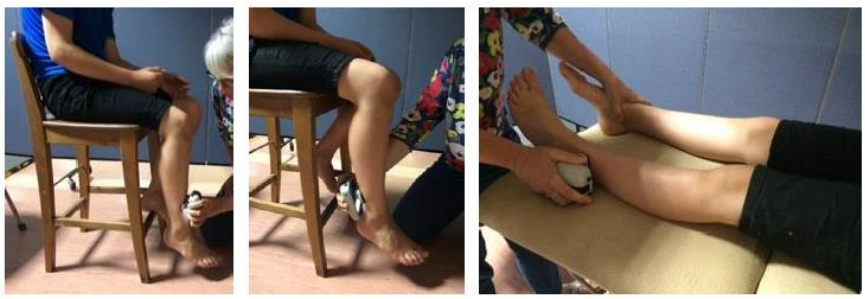
The test will be randomly generated to stepping to the four targets 15 times each, (60 times in total). It is anticipated that this part of the test will take up to 30-40 minutes with some time for rest.

Measuring leg movement and muscle strength

The researcher will measure leg movement at the hip knee and ankle and then test muscle power by asking your child to push against a hand held device which measures power.

Your child will be offered a small gift (e.g. lolly, pen, soft toy) to say ‘Thank you!’

The photos below show the researcher, Rachel Rapson, carrying out the tests:



Week 2 (60 minutes)

The Next Step-The 'Next Step' test performed in session 1, will be undertaken twice with two different people. Your child will be offered a rest break between each test.

Will any laboratory or genetic tests be done?

Apart from the tests described for Week 1 and 2, no other laboratory tests will be done. No genetic testing will be done.

Are there any side effects?

Your child may find the tests tiring to complete and they might experience muscle soreness after the strength testing.

What are the possible benefits of taking part?

The information we collect may help us to improve how we measure benefits from treatment in future. Taking part in this study will not directly benefit your child.

What happens when the research study stops?

We will collect all the information together and we will decide whether it is a useful test to use in research or for measuring response to treatments.

Will my child's taking part in the study be kept confidential?

Your child's name and address (which we need in order to contact you) will be stored on University premises and kept separate from the other information you supply during the project. Personal data will be stored for the duration of the study and 1 year after it has finished to allow us to contact you. We will not pass this information onto anyone else and will delete this information once the study has finished. All other information collected about your child during the course of this research will be kept strictly confidential. The film from the 3D camera will produce a computer model of movement (graphs) and will not be identifiable as your child. We will store your child's data using a unique code rather than their name. All information will be stored electronically on a University computer which is password protected and encrypted. All information will be handled in compliance with the General Data Protection Regulations (2018).

You can find out more about how we use your child/ward's information by contacting plymouth.sponsor@plymouth.ac.uk

What will happen if I don't want to carry on with the study?

You and your child can withdraw from the study at any time without giving a reason. Whatever decision you make will not affect the care your child receive in anyway.
Should you decide to withdraw your child from the study the measurements we have collected up to that point will be kept and used in analysis of the results unless you ask that they are also withdrawn.

Withdrawal from the project

You child's participation in the trial is entirely voluntary. You and your child are free to decline to enter or to withdraw from the study any time without having to give a reason. If you or your child choose not to enter the study, or to withdraw once entered, this will in no way affect your child's future medical care. All your information will be treated as strictly confidential. You can withdraw your child during the measurement session if they become upset or distressed or no longer want to participate for any reason.

What will happen to the results of the research study?

We aim to publish the results of this study in medical and other journals and to present at relevant national and international conferences. We will ask if you want to be sent a summary of the key findings or a copy of any publications at the time of the study.

Who has funded and reviewed the research?

This research has been funded by the National Institute of Health Research ICA-CDRF-2017-03-041 and it has been reviewed by independent experts external to Plymouth University.

Ethics approval has been gained for this study from the NHS South West Research Ethics Committee.

What should I do if we are interested in taking part?

If you and your child are interested in the study, please return the reply slip or contact Rachel Rapson whose contact details are given at the end of the sheet. She will then contact you to see if you have any further questions. If you and your child are happy to participate we will arrange an appointment to meet to carry out the tests.

What if there is a problem?

In the unlikely event your child is harmed by taking part in this study, there are no special compensation arrangements. However, neglectful harm will be covered by the insurance scheme of the University of Plymouth which is leading on this study. If your child is harmed due to someone's negligence, you may have grounds for a legal action but you may have to pay for it. Regardless of this, if you wish to complain, or have any concerns about this study, the normal National Health Service complaints mechanisms are available to you.

If you have a concern about any aspect of this study, you should speak to the research team who will do their best to answer your questions.

Rachel Rapson Chief Investigator and Physiotherapist 07870 501 834

Professor Jos Latour University of Plymouth 01752 586 578

Professor Jonathan Marsden University of Plymouth 01752 587 590

Professor Bernie Carter Edgehill University 01695 657 771

The Patient Advice and Liaison Service PALS are also there to help.

Patient Advice and Liaison Service

Freephone 0800 032 7657 or 01803 219700.
Lines are open between 8am – 6pm, Monday to Friday.

Contact for further information

If you would like any further information about this study, please contact:

Rachel Rapson

Faculty of Health, Education and Society

School of Health Professions

Plymouth University

Peninsula Allied Health Centre

Derriford Road

PL6 8BH

Tel 07870501834

Email rachel.rapson@nhs.net

You should be given a copy of this information sheet and a signed consent form to take home.

Thank you for reading this and considering whether to let your child take part in the project.

Contact reply form:

I would be interested in letting my child take part in this study.

I would be happy for the researcher to contact me:

Child/ward's name……

Parent/Guardian name……

Phone number …….

Email address……

Address……..

…….

Please return using the SAE to:

Rachel Rapson at PAHC, Plymouth University, Derriford Road, Plymouth, PL6 8BH

Or contact

rachel.rapson@plymouth.ac.uk

07870501834

Appendix 2- Participant Information Sheet- What constitutes 'usual Physiotherapy care' aimed at improving walking and balance for ambulant children with cerebral palsy? - A consensus study.

IRAS 254056
Participant Information Sheet

We would like to invite you to take part in our research study. Before you decide whether to take part, we would like you to understand why the research is being done and what it will involve.

One of our team will go through this information with you and answer any questions you may have. Talk to others about the study if you wish. Take time to decide whether or not you want to take part.

The main researcher is Rachel Rapson, who is a physiotherapist. She is doing this study as part of her PhD and is working with a supervisory team of Prof Jon Marsden and Prof Jos Latour (University of Plymouth) and Prof Bernie Carter (Edge Hill University).

What is the purpose of the study?

Children with cerebral palsy (CP) frequently have difficulties with walking and balance. However, the amount and type of physiotherapy they receive is resource dependent and can vary between practitioners and across the country.

In order to test a novel treatment against usual care, it is important to define what comprises usual care, both locally and nationwide. The study will involve a forum meeting to seek consensus on usual care in the South West and another in the South East of England. The results of both meetings will be pooled and finally all participants asked to reach consensus on usual care for this group of children.

This study will establish the 'usual care' control group for comparison to a novel intervention in a future feasibility Randomised Controlled Trial (RCT), to be carried out across Devon and Cornwall.

Why have I been chosen?

We are inviting you to take part as you are an experienced physiotherapist who works with children with cerebral palsy either in Devon and Cornwall or the South East of England.

Do I have to take part?

No. It is up to you to decide whether to join the study. We will explain the study and go through this information sheet with you. If you decide to take part, we will ask you to consent to participate in the study.

You will be have time to talk to a member of the research team by phone and at the forum meeting before deciding whether to consent to taking part. You will have a copy of the information sheet and consent form to keep for your records. You are free to withdraw at any time, without giving a reason.

What will I have to do if I choose to take part?

Survey

You will be sent an anonymous online survey, 2 weeks before the forum meeting it will ask you about your ideas on what usual care should be for children aged 8-18 years who have CP and who can walk. You will be asked to consider different aspects of care that a child may receive from a physiotherapist. Ideas and experience are likely to be different between the participants. There is no right answer. The ideas from all of the participants will be analysed by the research team, who will combine the ideas into a set of statements which best describe usual care for this group of children.

Discussion group

At the forum meeting the chief investigator, Rachel Rapson, will act as a facilitator. She will also have one or two people assisting her running the meeting.

At the start of the group, you will be asked to complete a questionnaire in which you score your level of agreement with the statements which have been written about usual care, using a 5 point scale. The group will have a break while all of the scores are analysed by the researchers. Your questionnaire will be returned to you so that you can compare your score with the group average score for each statement. The other participants will not know how you scored the statements.

The group will then have a facilitated discussion about the statements and whether the statements should be included, excluded or amended. Rachel will ensure that everyone has an equal say and chance to express their opinion. The statements on usual care will be rescored and discussed twice more to see if a consensus opinion can be reached within the group.

What are the possible benefits of taking part?

There will be no direct benefits to you to taking part. However, we hope that you will find it informative and enjoyable. You will be able to network with other physiotherapists and you may find that you can develop your professional practice by taking part in research. You will not be paid for your participation but you will be reimbursed any reasonable travel expenses and provided with refreshments at the forum meeting.

What are the possible risks of taking part?

We do not anticipate any risk in taking part. There will be some group rules to ensure confidentiality and to ensure that everyone in the group is treated equally and with respect.

What happens when the research study stops?

We will collect all the information together and we will decide whether consensus has been reached locally and between regions. This will then be used to describe the control group in a future RCT.

Will my taking part in the study be kept confidential?

The University of Plymouth is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study and will act as the data protection officer for this study. This means that we are responsible for looking after your information and using it properly. The University of Plymouth will keep identifiable information about you for 1 year after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

The University of Plymouth will use your name, and contact details to contact you about the research study, and make sure that relevant information about the study is recorded for your care, and to oversee the quality of the study. Individuals from the University of Plymouth and regulatory organisations may look at your research records to check the accuracy of the research study. The only people in the University of Plymouth who will have access to information that identifies you will be people who need to contact you to audit the data collection process. The people who analyse the information will not be able to identify you and will not be able to find out your name or contact details.

You can find out more about how we use your information at https://www.plymouth.ac.uk/your-university/governance/information-governance or by contacting the University Data Protection Officer at dpo@plymouth.ac.uk.

What will happen if I don't want to carry on with the study?

You can withdraw from the study at any time without giving a reason. Your employment or future interaction with University of Plymouth will not be affected.

Withdrawal from the project

Your participation in the study is entirely voluntary. You are free to decline to enter or to withdraw from the study any time without having to give a reason. All your information will be treated as strictly confidential.

What will happen to the results of the research study?

We aim to publish the results of the RCT, which will include this consensus work on usual care, in medical and other journals and to present at relevant national and international conferences. We will ask if you want to be sent a summary of the key findings.

Who has funded and reviewed the research?

This research has been funded by the National Institute of Health Research ICA-CDRF-2017-03-041 and it has been reviewed by independent experts external to University of Plymouth. Ethics approval has been gained for this study from the University of Plymouth Faculty of Health and Human Sciences Ethics Committee.

What should I do if we are interested in taking part?

If you are interested in the study, please contact Rachel Rapson whose contact details are given at the end of the sheet. She will then contact you to see if you have any further questions.

What if there is a problem?

In the unlikely event that you are harmed by taking part in this study, there are no special compensation arrangements. However, neglectful harm will be covered by the insurance scheme of the University of Plymouth which is leading on this study. If you are harmed due to someone's negligence, you may have grounds for a legal action but you may have to pay for it.

If you have a concern about any aspect of this study, you should speak to the research team who will do their best to answer your questions.

Rachel Rapson Chief Investigator and Physiotherapist 07971246592
Professor Jos Latour University of Plymouth 01752 586 578
Professor Jonathan Marsden University of Plymouth 01752 587 590
Professor Bernie Carter Edge Hill University 01695 657 771

Contact for further information

If you would like any further information about this study, please contact:

Rachel Rapson
Faculty of Health and Human Sciences
School of Health Professions
University of Plymouth
Peninsula Allied Health Centre
Derriford Road
PL6 8BH
Tel 07971246592
Email rachel.rapson@nhs.net

Appendix 3 -Checklist of usual Physiotherapy care aimed at improving walking and balance for ambulant children aged 4-18 years who have cerebral palsy.

Please enter an X in each box to indicate criteria are met

1	**Referral and discharge criteria**	
a	Children and young people with GMFCS III are managed on a long- term multi-disciplinary care pathway from initial referral to transition into adult services.	
b	Children with GMFCS levels I and II are offered episodes of care related to their functional needs and are discharged where there are no identifiable needs or their musculoskeletal condition is stable.	
c	Where children are discharged, information is given to them and their carers to help them identify key triggers/red flags for timely re-referral into the service.	
d	Ambulant children have ongoing access to orthotics as required.	

2	**Location of Physiotherapy**	
a	Children are offered virtual consultations and face-to-face appointments, as appropriate.	
b	Appointments take place in a Children's physiotherapy department or a child-friendly general outpatient clinic setting.	
c	Appointments are offered in school or at home when indicated due to environmental needs or co-morbidities.	

3	**Assessment** The following core areas should be included in assessment:	
a	Gait analysis (video/observation)	
b	Pain	
c	Leg length	
d	Spinal posture	
e	Muscle tone	
f	Muscle power	
g	Range of movement	
h	Functional task performance	
i	Balance	
j	Patterns of movement	
k	Gross motor function	
l	Psychosocial	

4	**goal setting**	
	Specific Measureable Achievable Realistic Timed (SMART) goal are set collaboratively with the child and their family.	

5	**Physiotherapy interventions**	
a	Functional, task-specific training is used to treat walking and balance difficulties. Adjuncts to task-specific training include using a treadmill with or without body weight support.	
b	Vibration training is used to improve balance and posture (where equipment exists).	
c	Physiotherapists encourage and facilitate children to develop active lifestyles including aerobic exercise, fitness training and modified sport.	
d	Strength-training using progressive resisted exercise is **not** employed as a treatment to improve walking and balance.	
e	Prolonged passive stretching is used to manage contractures using orthotics, serial casting and supported standing programmes.	
f	Postural management approaches are employed for children with GMFCS III, including mobility equipment and environmental adaptation.	
g	Exercise and functional activities that encourage full joint range are recommended where there is risk of contracture development.	

6	**Frequency and intensity of physiotherapy**	
a	The frequency of blocks and reviews is determined by clinical need.	
b	Blocks of 4-6 appointments are offered where there is a functional need.	
c	Children are reviewed every 3-12 months.	

7	**Outcome measurements** The following outcome measures are considered for the evaluation of interventions aimed at improving balance and walking:	
a	Passive range of movement	
b	Modified Ashworth scale	
c	Modified Tardieu scale	
d	MRC (Oxford) scale muscle strength	
e	Gross Motor Function Measure	
f	Observational Gait Scale	
g	10 metre walk test	
h	Instrumented gait analysis	
i	Patient reported outcome measures e.g. Goal attainment scale	
j	Therapy Outcome Measures	

8	**Advice and information**	
a	Support is given to the child, parents and school to understand the impact of diagnosis and prognosis of the child's condition.	
b	Signposting to local and national resources such as support groups, local offer, and charitable organisations.	
c	Training and education is offered for child, parents, school and other health and social care professionals.	
d	Timely input to SEND/EHCP process.	
e	Where the family or young person gives consent, information is shared with education, health and social care services through reports, therapy advice/programmes and clinic letters.	

Appendix 4 -Themes arising from interviews of parents, children and physiotherapist, with illustrative quotes.

Fitting therapy into normal life
Therapy at home: A bit of a battle at times
"usually when I am about to go to bed, I usually do it…usually daddy comes and helps me and tells me what move I have to do" (Isaac, aged 11). " … with our life being fairly chaotic and having another child with additional needs in the house. It can be really quite hard because [helping her to do physiotherapy exercise] will quite often set her sister off. In terms of the attention … so then that becomes really tricky to manage" (Gabby, aged 7). "…getting 3 kids to bed situation, with baths….ready for school, whatever the next day…Trying to fit everything in. And I guess sometimes, you know, as is with everything, you sometimes give up on that bit don't you? There's only so much, there's only so many times you can tell someone…" (Isaac's Father). "… when we put him to bed, we do little bridges and stuff like that. And when I put his shoes and socks songs, I give his ankle a little flex trying to do a little bit. When the opportunity arises, because it's so hard with him to get him to sit and do a whole session of physio" (Harry's Mother).
Therapy at school: Time out versus feeling singled out
"He does about 95% it will be at school. And we just do little bits here and there when we can fit him in" (Harry's Mother). " I was more or less happy as I got to slip school" (Isaac, aged 11). "The trouble is … he'd be missing some type of schooling to have to do it. And This is why right from when he was young, …we do it all at home because then it's not putting anything on the school. So, so to speak. You know school is school and then physio is at home" (Ethan's Mother). "We didn't do a lot of it, a lot of it was done at school. But they didn't seem to say that they had any problems with it. But we've definitely seen an improvement from him doing it" (Harry's Mother). "I'm very grateful that she gets a lot of good work done at school with her TA's and is very much more receptive to them and the situation. And they get to go to a quiet room and focus properly" (Gabby's Mother).
Motivation to exercise
Motivation and engagement with usual therapy programmes

“They are boring and they’re repetitive and things” (Daisy’s Father).

“ the usual physio is like the same thing as like exercising every time” (Ethan, aged 13).

“…either bribery or trying to make it as fun as possible, but it is always a challenge” (Gabby’s Mother)

“…maybe a bar of chocolate at the end of it sometimes” (Ethan’s Mother).

“At school, ….sing with TA” (Bella, aged 16).

“ I like doing my exercises… I like everything about my exercises…They make me feel happy… [I like] playing with the ball” (Freddie, aged 7).

“well she likes that, she giggles when she does physio… so I think that was good” (Bella’s Mother).

“we were doing something achievable and manageable and a routine type thing first doing this exercise and this one and then this one [usual care]. With the autism, routine it’s very important, to keep it to a routine, because then he knows what’s coming next, he knows, and that’s how he likes his life really” (Freddie’s Father).

“When he brings his [exercise] sheet home, he is very keen to take it off me and show me what he does” (Harry’s Mother).

“I thought it would be much better, to be honest…coz , it didn’t really work. Cus it only made one improvement, which is putting my jeans on” (Alfie, aged 10).

“I kept my foot down a little more” (Ethan, aged 13).

“ Yeah, he seems to be able to run around more. He's not falling over quite so much. He does seem to be a bit more steady. He's still got a little way to go. But there's definitely some improvement there. I think longer term he could still make some more improvement” (Harry’s Mother).

“ I’ve made a lot [of progress] on my scooter but not on the swing ball. At the beginning of the summer. I couldn't really balance [on my scooter], I it's just like I couldn’t push on it. And now I know how to go down hill with it” (Gabby, aged 7).

“[his] balance,[improved], like how much longer he was able to last being stood on like the one leg or the other” (Ethan’s Mother).

The impact of gaming on motivation and engagement

“From my point of view, it’s a great thing. You know, if you can get something like that entertains, you know from the point we're trying to get Daisy to do her exercises, especially when she was younger. They are boring and they’re repetitive and things. So if you've got something that will encourage them to do what they should be doing, then it's got to be a good thing” (Daisy’s Father).

“when you're doing a stretches, if you talk to him and you take his, you his mind away from what you're doing…there’s a little bit more give because he's ...not fighting against you, and I think that's the same with using this machine because he's focused on the game” (Ethan’s Mother).

“He really loves playing computer games and kids do, and speaking for him, if he could do some sort of gamified physio, it would really support him it would be really positive . I think kids, especially now all kids have games so if they can incorporate physio into games…using the same muscle groups and the same physio exercise is a really good idea” (Freddie’s Father).

“if I did have the machine, because it has games it would have helped me. Because I do like to play games and I feel like I would do it more” (Isaac, aged 11).

“I think because if I was enjoying it a bit more amount of time, I would do a bit more because, I was entertained it same time” (Daisy, aged 14).

““the fact that you might get some kind of interactive thing. From our point of view, I think it would have helped us to try and keep him doing his exercises” (Isaac’s Father).

“..They got on really well with the Happy Rehab because the child is really motivated and wanted to use it. But he won't do any [of his usual] physio with his parents at home” (Physio 3).

“I would say the difference is, she comes home every day… she talks about the [Happy Rehab] machine. If she had the chance to do more physio on that machine, she would love it for longer sessions” (Bella’s Mother).

“…they lasted a little bit longer. They tolerated more time doing physio [with the Happy Rehab]” (Physio 2).

“It will probably improve their compliance with it. [They are] more likely to **want** to do it and have that encouragement to do it … because they know they are going to have fun. It makes it less of a therapy program then it makes it more of an activity” Physio 2).

The opportunity to try something new

The opportunity for fun and games

“It was nice to do something a little bit different and interactive, and I think as it was a new piece of equipment the child enjoyed using it and the staff are really engaged as well because I did it as part of school sessions” (Physio 2).

“I think it's a nice modality to have as something as something different, and that's a bit ‘computery’ for our boys that like that” (Physio 1)

“I think the theory of it is really great. Anything that gets them moving or exercising in some form or they enjoy. It's a … plus 'cause we need more interventions like that I think” (Physio 3).

“Ethan's very competitive. So he was very wanted to beat the previous score, but if he had a bad round on the first couple of games that was it, then he would feel deflated. He would know already that he wasn't gonna beat his score” (Ethan’s Mother).

“Once I got more than 9 points” (Caleb, aged 7).

“Yeah. [I liked] Everything” (Bella, aged 16).

“ [I liked] the games on it. I liked making the things (characters) pop up… but not the duck one” (Ethan, aged 13).

“So it was really fun… I liked the hockey game. I want to keep it [the Happy Rehab]” (Caleb, aged 7).

“I think it could do with a few more instructions about what the games [work on] ….’cause Alfie would pick a few games and then you'd go into it [and realise] that's gonna be actually too hard for him” (Physio 1).

“It was just the way you have to strap him all in, he didn't like it…these sort of restricted him. He tried to get out himself once and [to Alfie] you hurt yourself...” (Alfie’s Mother).

“I thought it [Happy Rehab] has a nice selection of games and activities or the different age ranges and different cognitive levels as well. There were some nice basic ones that didn't take a lot of working out, which I think was quite good for my child because she's got a learning disability” (Physio 2).

An opportunity realised

“I think it's the concept of it. I think it's great... the machine does stretches that you I don't think as a person could physically do not when people were so tight” (Ethan’s Mother).

“I thought Bella got more range [of movement] than she would do… I think the machine encouraged her to do it a little bit more” (Physio 2).

“I can actually remember the stretch being quite hard for her, and then the machine just made it so fun for her” (Bella’s Mother).

“I think that worked really nicely to improve that weight [bearing] to the side because those exercises are really specific and were brilliant. Those games are really good encouraging that movement” (Physio 2).

“he was having more sort of compensatory movements than I hoped he would have….There was a lot of like ‘I'm moving my shoulders and I moving everything’ but the bit that he needed to do. So I found that to start with, he's still needed, sort of hands-on” (Physio 1).

“But there was no power. There were definite times where there was no power going to it. 'cause you would plug it in and the footplates normally move a bit… there was none of that” (Physio 1).

“Hard to concentrate on blob game. Machine playing up again after 5 minutes so stopped early. Couldn’t control the game with the left pedal properly” (Bella’s teaching assistant).

“ It kept stopping, and it was really hard. All the games except the space invaders. [it would be better] if it had easier games and the machine worked better” (Alfie, aged 10).

“…the games were really hard. Except for space invaders and the machine as well –it messed up- kept messing up, so I think it put him off them” (Alfie’s Mother).

Opportunity knocks, but does it fit in?

“[the Happy Rehab] takes up a lot of space. No, actually that was alright, yeah” (Caleb’s Mother).

Yes I mean- its quite a big piece of kit. But in our situation we would be able to accommodate that. We have a reasonable size house. So having it in the house wouldn’t be an issue “ (Isaac’s Father).

“…for 10 weeks now. I mean, yeah, you would have taken up a fair bit of room, but we can get over that. You can work around something over a shortish period of time, you know. We would’ve found room in a bedroom or in the lounge or somewhere for it to go where we could just put into a corner” (Daisy’s Father).

“And it was nice that it was at home, so it was something that he could do that fitted in with his routine, that he could do it for as long or as little as he wanted” (Physio 1).

“I would like [it again in] six months from now. I would like to give it another go…it would vary it a bit , because like it gets repetitive to do the same [physio exercises] all the time” (Ethan, aged 13).

“I think because of his family circumstances; being unable to support the child so well to use it every day because his mobility … he needs that bit more support and time getting in getting it set up…. and also they had it downstairs” (Physio 3).

“transporting it and getting it into front doors … the larger one’s quite wide and in the area where we live there's a lot of small inaccessible cottages with smaller doors and lots of steps” (Physio 2).

“there’s wires everywhere, where all the wires come out. It is hard when [children have] got cerebral palsy because they trip over everything” (Alfie’s Mother).

It would be the kind of thing that during the school holidays would be really helpful” (Gabby’s Mother).

“It feels like it still needs that Physio support… Have one here [CDC] … so they could come in and set it up here and one at home for six weeks post op or post Botox” (Physio 1).

“I think in a perfect world I'd quite like to see it like here [CDC] and you could book it out almost like a clinic where you could maybe have an assistant running it and they could just come in, set it up, wipe it down, go on it and you'd get a bit more use of it” (Physio 1).

“ [using HR in CDC] It wouldn't be something we would regularly be able to access. So if you had travelled somewhere to go on it three times a week, it's quite a……You know, it's quite a….trek” (Ethan’s Mother).

“It's just not practical for us, both working to then be able to get back in time. You know on a regular basis. If it been once in a while, then fine, but to do every week, … it would eat into my day. So three days a week, you know would be half a day [of work] really” (Daisy’s Father).

“-they [Happy Rehab devices] take up a lot of space! We did effectively lose a treatment space while we have them all” (Physio 1).

Physiotherapists out of their comfort zone

Unsettling practice

“… I think [the training package] was good. I think that covered kind of the FAQ [frequently asked questions] and like the setting up and glitches. So

covering those kind of things, this happens, look at these connections and things was really useful" (Physio 2).

"I wasn't as confident as using and setting up the happy rehab, so in that instance it took more time and probably took more resources. They took an assistant with me to help me set up because I didn't feel that confident" (Physio 3).

"I did have a quite a long training. I didn't … feel completely satisfied that I felt 100% confident to set it up… It probably would have been better if I'd then had a child straight away following the training … but I think then it was like a couple two months later…. I'd, you know, forgotten" (Physio 2).

"I think I'd really like to have a video of how to set it up…..so I'd be able to watch a video exactly…. and pause it. So for example, setting the range of movement, setting the strength, which games did which?" (Physio 3).

"Alfie had a lot of issues at the start didn't he? With it not working and that, and I wasn't confident at all in it...it was just working out how to use it. I think I think once that technical side of it ...if I was a bit more familiar with it, I think I'd be so much more happy" (Physio 1).

"…it's been quite a difficult time for a team point of view … so I think it's been quite difficult. [Participating in the trial] has been another thing that we've had to think about or that I've had to try and think about in the back of my mind" (Physio 2).

"But we have found that quite difficult to stick to the timelines of the follow ups…. sometimes Covid, if children have been unwell or isolating … also work commitments, family commitments" (Physio 3).

Unsettling device

So the first size didn't fit him. So there's quite a lot of time like faffing around getting the right size. He would have fitted in what their manufacturer's guidance was for the top end of 1… his feet were like hanging off the footplates" (Physio 3).

"Problem solving that's that took longer that meant extra time that I haven't sort of banked on as being part of it. And luckily, Alfie's mum is really understanding and is really nice, but a different parent might not have been quite so understanding of you know it's gonna be a couple of weeks so I can come out or let's try and work it out" (Physio 1).

"So I think that he [research assistant] tried to get in touch with them [the distributer], but it was we didn't really have any success in getting them" (Physio 1).

Altruism and the challenges of participating

An altruistic decision

“You do to sort of get something back, if you like. Daisy has had a lot of help along the way. Yeah, it was nice to try and give something back to obviously then feed into the future” (Daisy’s Father).

“ we were keen to do anything we can to try to improve his situation, and if that improves things for other people going forward, then it can only be a good thing can’t it?” (Isaac’s Father).

“Felt good to gets to kind of, I don't know in someway to be contributing some sort of progress, you know, need developments and stuff in the in the field” (Gabby’s Mother).

“Yeah, everything we needed to know and what we were letting ourselves in for… Ha , ha!. They gave us the option not to keep on, but we chose to. So it cant be that bad” (Daisy’s Father).

“…we knew what you were going to be doing and testing … the equipment and tests that you ran as well” (Isaac, aged 11)
“You explained about the machine exercises that they were going to do what she had to do?” (Bella’s Mother)

“I fully understood what was gonna happen.” (Caleb’s Mother).

““I found that [the information pack] was pretty easy and comprehensive to understand it wasn’t very technical… I mean it was written in a good simple way that someone like myself could understand” (Freddie’s Father).

“Yes fine, it was quite easy to understand and I could explain it to my husband” (Harry’s Mother).

“you expect to be randomised in some way. I mean that’s life, you know. What he gets picked for is what he gets picked for…” (Freddie’s Father).

“I mean it's the luck of the draw whatever doesn't matter. It's just the fact that somewhere along the line you've got to have a differential, haven’t you?” (Daisy’s Father).

“I think most people assume they're gonna get randomized into the intervention group. I think as long as it's clear from the start that you know there is actually a 50:50 chance” (Physio 1).

“I was quite happy. Just wanted to see how it goes” (Daisy, aged 14).

“[randomisation] was all right, because….but I haven’t given it much thought” (Isaac, aged 11)

“yes I’m pleased that he got in the [intervention] group” (Alfie’s Mother).

Some consequences of altruism

“A bit disappointed because the robot physio sounded fun. Because there’s games and stuff that sound interesting” (Gabby, aged 7).

“Well, [the online diary] stopped working at the beginning of the year, so I ended up not using it, so I've had to log it on my phone” (Caleb’s Mother).

“I think we made an assumption that an online diary would be easy, but … it would be easier just to have like a little booklet just to fill out at the time that you're doing it. “…our life is really quite chaotic- I did struggle to keep up with it at some points” (Gabby’s Mother).

“[The diary] should be like a weekly one… you should have a little diary- like ‘my child has done it 3 times per week, and this is how he reacted’” (Alfie’s Mother)

“Yeah, I didn't understand the [online diary] questions you would ask me at the end. Like, was this a practice like a rest day or something or an exercise day?” (Caleb’s Mother)

“[the information leaflet] had everything on there, on the pictures of the machine as well, which was good, so he knew… even before arrived what it was gonna be like,. “ (Ethan’s Mother)

“…because its all been done at school, there is nothing to put into them. Was school getting them as well? I guess they were filling them in at school. But the emails were very informative and all that” (Harry’s Mother).

“So, yeah, they were very easy, it was a good system with the drop down menus and just picking from those, yeah it was really good actually” (Freddie’s Father).

“ [the reminders] was it once per week or five days? …which was fine cos there are a couple of days where you forget and it was helpful to have a reminder” (Freddie’s Father).

“I filled it in for a couple of days, but I kind of forgot about it after that. kind of went over my head” (Daisy, aged 14)

The challenge of assessment

“she found [Next Step] more tricky. I could tell because of her body and the way she was trying to move and how you know her body didn't look natural until she got into the position. Yeah, so that was a wee bit more difficult for her, I would say” (Bella’s Mother).

" [interviewer speaking] what did you think about the lights on your legs. Did you like it? (Harry-shaking his head) You are shaking your head. You didn't like it" (Harry, aged 9).

"Well, it's kind of felt weird and It hurt when they took all this stuff off… it's on the skin and they peeled it off …" (Gabby, aged 7).

" [speaking about gait analysis] She had a winter school uniform on the stuff was stuck to her clothes a lot more than her skin and some of it is probably makes it less accurate. But I did notice that it was a bit less owchy" (Gabby's Mother).

"if we had had a bit more knowledge about the fact he was going to have the motion captures stuff, we might have been able kind of prime him …maybe even a video-'we are going to do this'" (Freddie's Father).

"[more information would be helpful] for the parents so that they could … know how much to encourage the child. For example 'we are going to do the stepping 4 more times'" (Harry's Mother).

" I don't like the thing you do going fast…I just like the slow stretches" (Freddie, aged 7).

"It did make my legs a bit sore, because it made my leg go a bit higher" (Gabby, aged 7).

"The bit of the strength test where I was pushing hard, it sort of hurt my shin…it's alright it just hurts (laughing)" (Isaac, aged 11).

Enjoying the challenge

"for some of the children, perhaps some of the assessment the pre and post assessments were quite long and for some that was really tough for the children to get through" (Physio 3).

"You did give her plenty of opportunities to say… I don't want to do anymore or I'm too tired or whatever. And she quite happily chose to carry on herself" (Gabby's Mother).

"Its not that bad. I mean its, its no different to any other physio sessions where [my child] is not very compliant" (Freddie's Father).

"I really enjoyed it and I thought that Harry coped really well, because they were quite long and for him, it was quite mentally taxing" (Harry's Mother).

Well they [assessment sessions] are ok. Obviously he gets a bit tired after a while… they are a bit long winded" (Caleb's Mother).

“we are never going to get his 100% of the assessment done with him whatever you do, because he doesn’t have that level of behaviour and understanding, because of his autism” (Harry’s Mother).

“I don't mind [how long it took]…because it's a study and when you do a study you have to like cover every part, don't you?” (Bella’s Mother).

“it's taken us about an hour most times on average, doesn't it lasts about an hour. So yeah, I don't think it's not an issue from my point of view” (Daisy’s Father).

“That was OK” (Ethan, aged 13).

“I enjoyed watching, with all the little things on [gait analysis] and seeing how it worked. It was quite good” (Harry’s Mother).

“… it was quite cool, having all the equipment on” (Isaac, aged 11).

“[It was] good! The lights on the thing [Next Step] came on! The lights that I wanted came on” (Caleb, aged 7).

“[it was] fun. Uh, the dance mat game thing [Next Step]…It made me concentrate” (Daisy, aged 14).

“it was a bit easy” (Gabby, aged 7).

“ I think it was all good” (Ethan, aged 13).

“It was quite fun to be honest. I really liked it when I got to the strength part and the bit when I was walking up and down” (Isaac, aged 11).

“it's kind of a bit of a little bit of a morale boost in a way I think… to get lots of … positive validation about how powerful you are” (Gabby’s Mother).

Appendix 5- **Participation information sheet.**

Study Title: The ACCEPT Study: Does using a novel interactive trainer improve walking ability and quality of life for children with cerebral palsy?

IRAS 269948

Parent/Guardian Information Sheet for Child to Participate

We would like to invite your child to take part in our research study. Before you decide whether to let them take part, we would like you to understand why the research is being done and what it will involve.

One of our team will go through this information with you and answer any questions you may have. Take time to decide whether or not you want your child to take part and talk to other people about the study if you wish.

The main researcher is Rachel Rapson. Rachel is a physiotherapist who is highly experienced in working with children. She is doing this study as part of her PhD and her supervisory team is Prof Jon Marsden and Prof Jos Latour (University of Plymouth) and Prof Bernie Carter (Edge Hill University).

What is the purpose of the study?

Children with cerebral palsy often need to do exercises to keep their muscles flexible and strong and to help maintain their walking and balance skills. In order for the exercises to work, children need to do them several times per week, and this can be a burden for them and their family. Exercises are more effective when done in a functional position, such as standing. This may be difficult to do without adult help. Sometimes children don't do their exercises because they find them boring or dislike having adults helping them to exercise.

A new interactive trainer has been developed to help address these problems. The trainer supports the child in an upright position in order for them to do a series of exercises prescribed by their physiotherapist. The child plays a series of computer games, controlling the games with their leg movement. The trainer has motors in the footplates and knee pads which assist or resist the child's movements (Figure 1).

Figure 1: The new interactive trainer

We do not know if the trainer can help children improve their balance and walking. To answer this question, we need to compare this new trainer with usual physiotherapy care, in a large study (a full randomised controlled trial). Before we can do this we need to do a small study to see if it is feasible to conduct a full randomised controlled trial.

Why has my child been chosen?

Your child has been chosen as they are aged between 4-18 years old and have cerebral palsy but we will need to ask you some questions about your child to see if they are eligible. Some children who have CP wont be able to take part if they are not be able to control a computer game with a joystick.

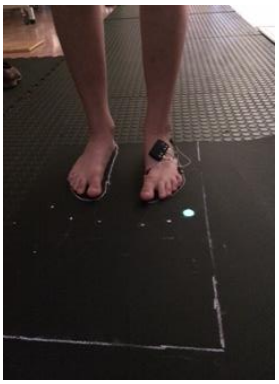
Does my child have to take part?

No. It is up to you and your child (wherever possible) to decide whether or not to take part in the study. Before you decide, a member of the research team will explain the study and go through this information sheet with you. If you and your child decide to take part, we will ask you to sign a consent form allowing them to participate. If your child is able to understand the research and is happy to take part and can write their name, they will be asked to sign an assent form, if they want to.

You will be given a copy of the information sheet and the signed consent/ assent forms to keep for your records. You and your child are free to withdraw at any time, without giving a reason. This will not affect the standard of care your child receives in the future.

What will my child have to do if we choose to take part?

If you decide to take part the researcher will arrange a convenient time to meet you and your child at your local Child Development Centre (CDC) for an initial assessment. During this initial assessment, the researcher will measure your child's walking and balance. You will both be asked to fill in a questionnaire (you can help your child complete this). Your child will then be randomly allocated to receive either the new intervention or their usual physiotherapy care. Your child has an equal chance of being allocated to the new intervention or to their usual care. Both treatments will be delivered by your local physiotherapy team. Your child's physiotherapist will meet you to set up the programme. Your child will be asked to train(new intervention or usual physiotherapy) for 10 weeks and then the researcher will book follow up assessments to re-measure your child's walking and balance after the 10 weeks and again at 20 weeks. You will be reimbursed for any reasonable travel expenses during the study.



Your child's usual medication should be continued. Your child should not undertake any assessment sessions if they show any signs of infection and illness (high temperature, vomiting, and diarrhoea) on the planned day of assessments.

As part of the study, we would also like to interview a small group of children and families about their experience of being in the study. You and your child will be asked if you would like to take part in the interviews. You don't have to take part in the interviews even if you want to take part in the rest of the study.

Week 1- Assessment with the researcher

The researcher will ask about your child's medical history, and measure their height and weight. The researcher will help your child set your goals for treatment using the Canadian Occupational Performance Measure. This tool will help us your child to think about the functional tasks which they would like to improve and asks them to rate their current performance and satisfaction with the way they do it. You or your child (depending on their age) will also be given the (CHU-9D) quality of life questionnaire to fill in.

Next Step test (30 minutes)- Your child will be asked to wear shorts, have bare feet and wear some electronic markers during this test. The test will be

described and demonstrated by the researcher. The test involves standing and stepping to four different targets which light up. The signal from the markers is picked up by a camera in the room which allows the computer to create a 3-dimensional representation of your child's movement during stepping. Following the demonstration, you and your child will have time to consider whether you wish to carry on and participate.

If you wish to take part, electronic markers will be attached to your child by using soft elasticated Velcro belt and four small stick-on markers on their feet. Your child will be asked to take some practise steps to determine their usual step length. Your child's foot position will be drawn around using chalk to mark the floor to show the start position and the target position. These photos demonstrate the test.

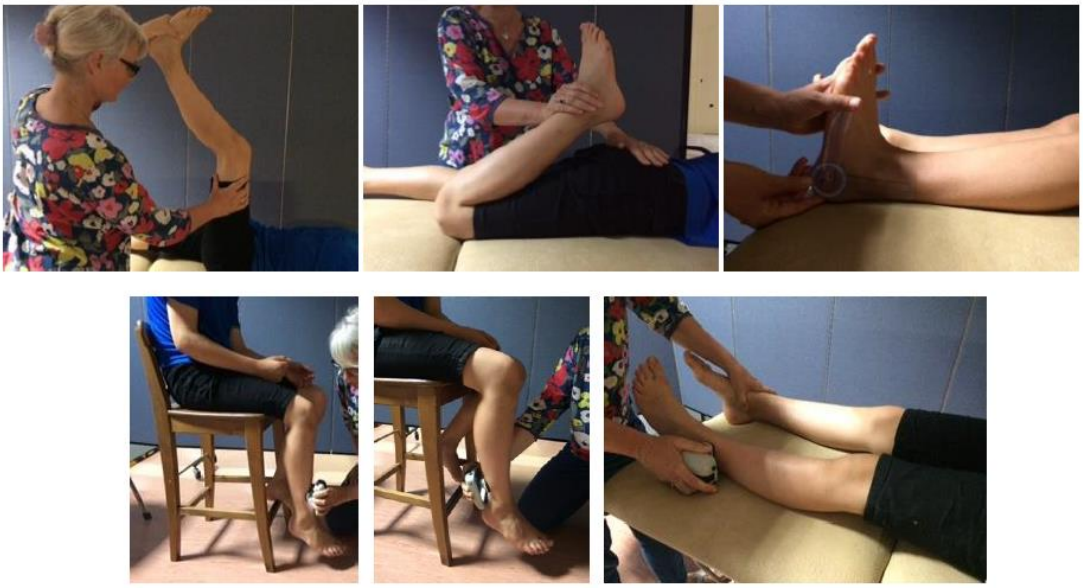
Your child will be asked to follow a series of instructions, which will be said in a way that they can understand. Your child will be asked to step 60 times onto the four randomly lit targets. Your child can rest whenever they wish.

Measuring walking (10 minutes)- The researcher will record how your child walks using with the markers which will still be in place. Your child will be asked to walk backwards and forwards in front of the camera six times.

Measuring balance (10 minutes)- The researcher will ask your child to try a series of simple balance measures such as standing on one leg.

Measuring leg movement and muscle strength (20 minutes)

The researcher will measure leg movement at the hip knee and ankle and then test muscle power by asking your child to push against a hand held device which measures power. In the photos below you can see the researcher, Rachel Rapson, carrying out the tests:



Randomisation- This is a randomised controlled trial. This means that we use a computer to randomly allocate your child to either treatment group. Your child has an equal chance of receiving treatment with the new training device or the usual physiotherapy treatment.

For children allocated to the Interactive trainer group: Your child will be asked to use the training device in place of their usual treatment for 10 weeks. This may take place in either your home, school or CDC. The location will depend on your local facilities and will be arranged between you and your physiotherapist. Your physiotherapist make an appointment to set up several simple balance tasks device to fit your child, tailoring it so that your child can operate the games using their leg movement. The computer games will include a warm up, balance games and strength training games using the resistance of the knee pads and footplates.

Your child will be asked to train for 20 minutes, three times per week. You or your child will be asked to keep a diary to record their training sessions. Your physiotherapist will review the treatment on week 5 and progress the exercise games as appropriate. Your child will then be encouraged to train for 30 minutes three times per week. Your child will be asked to discontinue their usual physiotherapy exercises during the training period.

For children allocated to the usual care group: Your physiotherapist will arrange an appointment to set goals and establish your child's physiotherapy

care plan. This will include a series of exercises targeted to meet your child's needs. Your child will be asked to train for 20-30 minutes, 3 times per week. You and your child will be asked to record their training in a diary, which will be provided.

Re-measuring the outcomes of the treatments at week 10 and week 20.

The researcher will arrange an appointment in your local CDC to re-measure your child's balance and walking using the same tests as week 1. The researcher will also ask you and your child to re-score the COPM measures of performance and the quality of life questionnaire at week 10 only.

Will any laboratory or genetic tests be done?

Apart from the assessments described, no other laboratory tests will be done. No genetic testing will be done.

Are there any side effects?

Your child may find the tests tiring to complete. It would be usual to feel some fatigue or muscle soreness following strength training.

What are the possible benefits of taking part?

Your child may benefit from treatments in either group. We are unable to pay you and your child for taking part.

What happens when the research study stops?

We will collect information about the number of children taking part and the number who did not respond or who declined to take part. We will report information such as how many measurements we were able to complete and whether the randomisation to the groups was acceptable to the people taking part. This will allow us to decide whether a full trial should go ahead.

The Happy rehab equipment is available for private purchase following the trial.

How will we use information about you and your child?

We will need to use information from you and your child's medical records for this research project. This information will include you and your child's initials/ name/ contact details. The film from the 3D camera will produce a computer model of movement (graphs) and will not be identifiable as your child. People will use this information to do the research or to check your records to make

sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead. We will keep all information about you safe and secure. Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

What are your choices about how your information is used?

You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have. We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

Where can you find out more about how your information is used?

You can find out more about how we use your information

at www.hra.nhs.uk/information-about-patients/

by asking one of the research team

at https://www.plymouth.ac.uk/your-university/governance/information-governance

by contacting the University Data Protection Officer at dpo@plymouth.ac.uk.

The University of Plymouth privacy notices can be assessed at https://www.plymouth.ac.uk/your-university/governance/information-governance/privacy-notices

What will happen if we don't want to carry on with the study?

You and your child can withdraw from the study at any time without giving a reason. Whatever decision you make will not affect the care your child receives in anyway. Should you decide to withdraw your child from the study, the measurements we have collected up to that point will be kept and used in analysis of the results unless you ask that they are also withdrawn. You can withdraw your child during the measurement session if they become upset or distressed or no longer want to participate for any reason.

What will happen to the results of the research study?

We aim to publish the results of this study in medical and other journals and to present at relevant national and international conferences. We will ask if you want to be sent a summary of the key findings or a copy of any publications at the time of the study.

Who has funded and reviewed the research?

This research has been funded by the National Institute of Health Research ICA-CDRF-2017-03-041 and it has been reviewed by independent experts external to Plymouth University. Ethics approval has been gained for this study from the North of Scotland Research Ethics Committee.

What should I do if we are interested in taking part?

If you and your child are interested in the study, please return the reply slip or contact Rachel Rapson whose contact details are given at the end of the sheet. She will then contact you to see if you have any further questions. If you and your child are happy to participate we will arrange an appointment to meet to carry out the tests.

What if there is a problem?

In the unlikely event your child is harmed by taking part in this study, there are no special compensation arrangements. However, neglectful harm will be covered by the insurance scheme of the University of Plymouth which is leading on this study. If your child is harmed due to someone's negligence, you may have grounds for a legal action but you may have to pay for it. Regardless of this, if you wish to complain, or have any concerns about this study, the normal National Health Service complaints mechanisms are available to you.

If you have a concern about any aspect of this study, you should speak to the research team who will do their best to answer your questions.

- **Rachel Rapson** Chief Investigator and Physiotherapist 07971 246 592
- **Professor Jos Latour** University of Plymouth 01752 586 578
- **Professor Jonathan Marsden** University of Plymouth 01752 587 590
- **Professor Bernie Carter** Edge Hill University 01695 657 771

The Patient Advice and Liaison Service PALS are also there to help.

Patient Advice and Liaison Service

Freephone 0800 032 7657 or 01803 219700.

Lines are open between 8am – 6pm, Monday to Friday.

Contact for further information

If you would like any further information about this study, please contact:

Rachel Rapson
Faculty of Health, Education and Society, School of Health Professions
University of Plymouth, Peninsula Allied Health Centre
Derriford Road, PL6 8BH
Email rachel.rapson@plymouth.ac.uk

You should be given a copy of this information sheet and a signed consent form to take home.

Thank you for reading this and considering whether to let your child take part in the project.

Contact reply form:

I would be interested in letting my child/ward take part in this study.

I would be happy for the researcher to contact me:

Child's name……

Parent/Guardian name……

Phone number ……

Email address……

Address……..

……

Please return using the SAE to:

Rachel Rapson at PAHC, GF15, University of Plymouth, Derriford Road, Plymouth, PL6 8BH

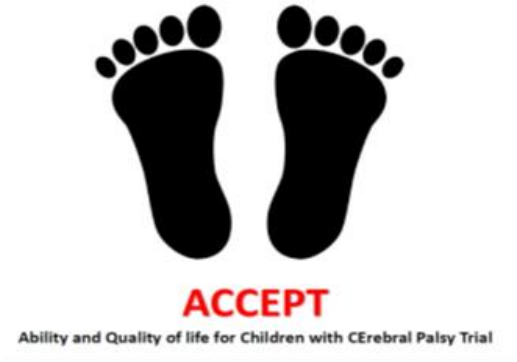
Or contact

rachel.rapson@plymouth.ac.uk

07971 246

Appendix 6- **Statistical Analysis Plan**

A Multi-Centre Feasibility Randomised Control Trial of a Physiotherapy Programme using an Interactive Exercise Equipment to Improve Balance in Ambulant Children with Cerebral Palsy



Ability and quality of life for Children with CErebral Palsy Trial (ACCEPT study)

Statistical Analysis Plan version 1

TABLE OF CONTENTS

LIST OF ABBREVIATIONS

AE Adverse Event

CDC Child Development Centre

CI Chief Investigator

COPM Canadian Occupational Performance Measure

CP Cerebral Palsy

CRF Case Report Form

CTU Clinical Trials Unit

DMC Data Monitoring Committee

GCP Good Clinical Practice

GMFCS Gross Motor Function Classification System

ICF Informed Consent Form

ISRCTN International Standard Randomised Controlled Trials Number

NHS R&D National Health Service Research & Development

PBS Pediatric Balance Scale

PI Principal Investigator

PPI Patient and Public Involvement

PIC Participant Identification Centre

PIS Participant Information Sheet

QA Quality Assurance

RCT Randomised Control Trial

REC Research Ethics Committee

SAE Serious Adverse Event

SDV Source Data Verification

SOP Standard Operating Procedure

SSI Site Specific Information

TMF Trial Master File

TMG Trial Management Group

TSC Trial Steering Committee

TRIAL IDENTIFIERS

SAP version	Version 1
SAP Version date	13.06.2022
REC Number	HCRW 20/NS/0018
ISRCTN Number	80878394
IRAS number	269948
Trial Sponsor	University of Plymouth
Sponsors number	19/20-1168
Funder	NIHR
Funders number	ref. ICA-CDRF-2017-03-041
Trial Chief Investigator	Rachel Rapson
Trial Statistician	Victoria Allgar
SAP Authors	Rachel Rapson and Victoria Allgar
SAP Revisions	Any revisions to the SAP will be documented here, including a brief justification and timing of revision in relation to unblinding of data

KEY SIGNATORIES

	Name	Signature	Date
Statistical Analysis Plan Authored by:	Chief Investigator Rachel Rapson	RRapson	13.6.22
	CTU Statistician Victoria Allgar		
Approved by	TSC Chair Stuart Logan		
	Independent Statistician Trish Hepburn		

TRIAL SUMMARY

Title	A Multi-Centre Feasibility Randomised Control Trial of a Physiotherapy Programme using an Interactive Exercise Equipment to Improve Balance in Ambulant Children with Cerebral Palsy.	
Internal ref. (or short title)	ACCEPT	
Clinical Phase	Feasibility	
Trial Design	Mixed methods RCT	
Trial Participants	Children with cerebral palsy aged 4-18 years	
Planned Sample Size	20	
Treatment duration	10 weeks	
Follow up duration	20 weeks	
Planned Trial Period	1/5/2020 until 1/8/2022	
	Objectives	Outcome Measures
1	To assess the feasibility of conducting an RCT evaluating the effect of interactive exercise equipment on walking for children with cerebral palsy	Feasibility Outcomes Descriptive statistics of planned outcome measures
2	To assess the feasibility of the intervention	Adherence, cost and safety of the intervention. Descriptive statistics
3	Investigate the participants' views of participating in the study	Thematic analysis of semi structured interviews

BACKGROUND AND RATIONALE

1 BACKGROUND

Cerebral palsy (CP) is a group of permanent disorders affecting the development of movement and posture that occurs in two to four per 1000 children [1]. Difficulties with walking and balance are common and can limit participation in schooling and functional activities [2-5]. Children with CP, for example, only spend 3.4 hours per week engaging in physical activity, nearly half that seen in typically developing children [6].

Walking ability can be classified using the Gross Motor Function Classification system (GMFCS) [7]. Children with GMFCS classification I-II are able to walk functionally outdoors, while children with grade III GMFCS require walking aids. Children with GMFCS I-III, the focus of the proposed study, comprise around 67% of the population (about 23,400) of children with CP in the UK [8].

There are multiple causes of walking difficulties in children with CP, including muscle weakness, contracture or bony deformity. Spasticity and weakness affect 80% of ambulant children with CP [9]. Secondary musculoskeletal problems develop throughout childhood due to the effect of spasticity on muscle length. Muscle growth does not keep pace with bone growth and this leads to deformity of the developing skeleton [10]. Children with CP often have poor balance, which further impacts on walking ability and everyday function [11].

Children with CP frequently undertake daily exercise programmes aimed at maintaining range of movement, strengthening weak muscles and developing balance skills. In Patient and Public Involvement (PPI) consultations, some children have reported that they do not want to do their therapy at home, feeling that it is boring or that it limits their participation in other activities. Parents reported that having to act as the therapist, facilitating their child to do stretches or training, conflicts with the parental comforting and protector role. Therefore, it is desirable to find exercise activities that are both fun and therapeutic which the child can do as independently as possible.

Current usual care consists of factors such as stretching, progressive strengthening exercises and functional task-related training. In many cases, children with CP find it hard to undertake exercises in functional positions such as standing, without support from a carer. The Happy Rehab™ (Innovaid, Denmark) interactive gaming trainer was developed (see figure 1) and marketed to help children exercise independently in a functional supported standing position. The novel interactive trainer provides support around the hips and additional assistance via servomotors aligned to the ankle and knees. This allows the child to exercise muscles functionally in novel ranges, e.g. strengthening the thigh muscles with the hip and knee in a straighter position. Children play a series of tailored exercise games controlled by the child's leg movement. The games-based exercises increase motivation and require the child to control the games by moving their weight side-to-side, forward and backward. It is proposed that this may improve balance during dynamic tasks such as walking.

Figure 1-The interactive exercise equipment

Whilst the interactive trainer is used more readily in Scandinavian countries and there is growing interest in the UK, there is limited evidence as to its effectiveness. A small-scale study of the interactive trainer that found marked improvements in walking, had a number of limitations in terms of outcome measures used, lack of follow up or control group [12]. Therefore, evidence is still required to establish the efficacy of the equipment.

2 RATIONALE

Before a full trial can be conducted to establish the effectiveness of the Happy Rehab™ interactive gaming trainer, there are still some questions with regards to the feasibility of the trial, intervention and conduct of the trial which need to be resolved.

In PPI consultations, children and families have expressed a desire to use the trainer at home in place of their usual care. Elsewhere, stakeholders have expressed the view that the interactive trainer would be more efficiently used in a clinical base to enable multiple users to train on the equipment. Physiotherapists have described the ability to support intensive training in a clinic as potentially challenging to staff. However, there are some clinical bases in special schools, which may allow the ease of access to multiple users. Therefore, as part of the feasibility study, the location of the trainer may be clinic, home or school based in order to compare the intensity of training in different settings, clinicians, parents and children's views on training, as well as treatment costs and the durability of the equipment or ease of arranging repairs.

In order to measure dynamic balance, we have devised and established the validity and reliability of a simple stepping test in children with CP and typically developing children (Rapson, Marsden, Pitsouni un-published work 2019). However, as this is a new measurement the necessary information to produce a power calculation is not currently available.

This study aims to establish whether it is feasible to conduct an RCT to assess the effectiveness of using an interactive trainer in children with CP. Children will be randomised to either a ten-week programme of intensive training with the Happy Rehab™ device in either a CDC, school or home setting, or to the control group of usual physiotherapy care. It will explore the feasibility and acceptability of the intervention, participants' views on randomisation, likely recruitment and retention rates and the frequency of any adverse events. The study will assess the feasibility of the proposed outcome measures, in terms of user satisfaction, percentage completed and ability to detect. Standard deviation confidence intervals, together with previous literature, will inform power calculations for the main RCT.

OBJECTIVES AND OUTCOMES

STUDY OBJECTIVES

The objectives of this study are as follows:

1) Determine the feasibility of a definitive trial by assessing:

Recruitment rate

Retention rates

Effectiveness and acceptability of randomisation

Change in clinical outcome measures

Effectiveness of concealment of allocation up to week 10

Concurrence with other surgical and medical interventions

Fidelity to treatment protocol

Appropriateness of clinical outcome measures

Sample size estimate for definitive trial

2) Determine the acceptability of the intervention by assessing:

Adherence to treatment

Safety of intervention

Cost of intervention and support needed to use it

3) Explore the views of a sub group of the study participants

By interviewing children, parents and physiotherapists about their experiences of participating in this feasibility RCT to assess the acceptability of the trial and the intervention.

Table 1 ACCEPT Feasibility Outcomes

To achieve the trial objectives the following outcome measures in Table 1 will be obtained.

Objective	Outcome
Feasibility of Definitive Trial	
Acceptability of the trial and intervention	Interviews of staff, parents and children
Can we recruit and retain participants?	Number of participants eligible Number recruited and randomised, date of recruitment recorded on study database Recruitment source Number of withdrawals. Number of participants lost to follow-up.
Effectiveness and acceptability of randomisation	Comparison of participant characteristics: severity, distribution of motor impairment, associated impairments at baseline Interviews
Effectiveness of concealment of allocation up to week 10	Number of times CI correctly guessed treatment allocation
Concurrence with other surgical and medical interventions	Number of operations or procedures that target balance and walking during the intervention and follow up period.
Change in clinical outcome measures	Change in assessment scores of outcome measures

Assess appropriateness of outcome measures	Number and percentage of outcome measures completed at each time point Interviews
Feasibility of Intervention	
Adherence to treatment	Diary data frequency and duration of training
Acceptability of treatment intervention	Incidence of breakdown of equipment Number of times participants were unable to access equipment Participant view on acceptability of interventions by Interview
Cost of intervention and support needed to use it	Local physiotherapist record of staff time and grade used to support intervention. Travel costs of staff and families. Number and cost of repairs
Safety of intervention	Number and type of SAE and AE
Investigate the participants' views of participating in the study	
Semi structured interviews	Thematic analysis of interviews and photos

POTENTIAL OUTCOME MEASURES

The following assessments will be carried out at week 0, 10 and those indicated with * at 20 weeks follow up. The physical assessments will take 70 minutes followed by up to 30 minutes goal setting using the COPM.

Potential Primary outcome measures:

Next Step test of dynamic balance* [30, 175]. This involves the calculation of peak medio-lateral (ML) , anterio-posterior (AP) motion of the COM estimate

Pediatric Balance scale* [77]

Potential secondary outcome measures:

Walking kinematics* involving the calculation of the knee angle at midstance relative to standing, peak-to-peak knee range during swing phase, ankle angle at initial contact and midstance relative to standing.

Muscle strength of quadriceps, tibialis anterior, and gastrocnemius and hip abductors using a hand-held dynamometer (three measurements)*.

Passive range of movement and modified Tardieu scale [176] of quadriceps, hamstrings, gastrocnemius and hip adductors using goniometer (three measurements)*.

COPM- Canadian Occupational Performance Measure [177]

CHU-9D- Paediatric Quality of Life measure [178]

TRIAL DESIGN

The trial is a single-blinded; mixed-methods feasibility randomised controlled trial. Participants will be randomly allocated 1:1 to either usual care or Happy Rehab™ interactive trainer. A sub group of participants will be interviewed about to find their experiences of taking part. The research question can be framed in the following way:

P Population – Children with cerebral palsy aged 4-18 years

I Intervention – A programme of physiotherapy using the interactive training equipment

C Comparison group – Usual care

O Outcome of interest – Feasibility of the trial and intervention

T Time – Three times per week for 10 weeks training plus 10 weeks follow up

PARTICIPANT ELIGIBILITY CRITERIA

Inclusion criteria

Diagnosis of CP GMFCS I-III.

Aged 4-18 years.

Leg weakness (≤4/5 on the MRC muscle strength rating scale) in at least 1 muscle group

Leg hypertonia (≥1 on the Tardieu scale fast stretch) in at least 1 muscle group

Ability to interact with a computer game using a mouse or joystick.

The age range reflects the recommended age range for the interactive exercise. Other inclusion criteria reflect the need to show an impairment in leg strength and tone, core features of a spastic Cerebral Palsy presentation.

Exclusion criteria

Selective dorsal rhizotomy or Multi level orthopaedic surgery within the last 12 months

Soft tissue surgery in lower limbs in last 6 months.

Anti-spasticity botulinum toxin injections within previous 3 months.

Training with the Happy Rehab™ in the last 4 months.

The exclusion criteria include interventions that could still produce a clinical effect during the trial training period. Children will not be excluded if, after recruitment into the trial, they undertake operative procedures and/or receive botulinum injection.

SAMPLE SIZE CALCULATION

Target recruitment of n=20

PLANNED RECRUITMENT RATE

3-4 participants per month over 6-month period

RANDOMISATION

at a ratio of 1:1 using minimisation criteria:

age 9 or below Vs 10 years or above;

GMFCS level I or II vs level III.

STATISTICAL METHODS

All analyses and data summaries will be conducted on the intention-to-treat (ITT) population which is defined as all participants randomised regardless of non-adherence with the protocol or withdrawal from the study (Table 9).

Participants will be analysed according to the intervention they were allocated to.

There is no interim analysis planned. The TSC will receive a quarterly report of all AEs and SAEs. If safety concerns arise, the chair of the TSC will contact the trial coordinator to review this.

The plan takes into consideration CONSORT guidance for reporting feasibility and pilot trials [252], CONSORT Patient-Reported Outcome (PRO) [253] and CONSORT statement for randomised trials of non-pharmacological treatments [254]. The results will be presented using the blank flowchart in Figure 1 (Appendix).

Participants will not be able to be identified from presented data. Analysis will be done by the CI with guidance from the Trial statistician.

STATISTICAL ANALYSIS

Feasibility Analysis

Data from screening, recruitment rate and source, withdrawals will be displayed in Figure 1 Consort diagram. This data will be used to generate realistic estimates for the full trial. Time from screening to consent will be described as mean (SD) number of days. Where data is non-parametric, median and inter-quartile range will be reported.

The baseline characteristics will be descriptively compared to ensure balance between the two treatment groups and also of those lost to follow up in order to identify any potential bias.

Potential primary and secondary outcome analysis

The planned primary and secondary outcome measures will be reported at each time point using descriptive statistics Mean (SD) for parametric data and Median(IQR) for non-parametric data. As this is a feasibility trial, it is not appropriate to perform a hypothesis test between-group treatment effects [179]. Instead, the baseline values and difference between allocated groups of the

follow-up minus baseline score will be estimated with confidence intervals. Blank Tables in the Appendix will be used to display the results.

Participant Population

Participant progression through the trial will be reported via a CONSORT diagram (Figure 1). Patient demographics will be described using the following variables(Table 2):

Age

GMFCS level

Impairment

Gender

Assessment of baseline variables

The following baseline variables will be descriptively compared using Mean (SD) between the two treatment groups (Tables 2 and 4):

GMFCS level

Age

Medical and surgical history

Height, weight

Frequency and location of usual physiotherapy

Other sports and social activities

Functional mobility

Assessment of recruitment rates (at baseline)

We will assess recruitment rates by calculating the proportion of participants (%) who were recruited to the study out of those eligible for each site (Table 1).

The time taken to recruit participants at each site will be described. We will calculate the mean and standard deviation or medians and inter-quartile ranges as appropriate of the time taken to recruit participants at each site.

Reasons for non-participation will be recorded

Descriptive list of factors affecting recruitment will be recorded

Assessment of retention rates

A comparison of outcomes by treatment group by site (Table 7a and b) and overall for:

Number and proportion of participants who attend at 10 and 20 weeks for trial procedures.

Number and proportion of participants who had no protocol deviations

Reasons for non-attendance

Assessment of clinical outcome measures

Assessment of the suitability of the primary outcome measures will be evaluated by a comparison of outcomes by change in scores with 95% CIs of treatment group for:

Number and percentage of participants with primary and secondary clinical outcome measures recorded at with baseline and follow up (Table 7a and b).

Number of participants reaching ceiling scores in PBS.

Calculation of mean and SD; median and IQR of clinical outcome measures at baseline, 10 and 20 weeks. (table 6a,b)

Assessment of differences between groups

Descriptive comparison of control and intervention group characteristics and clinical measures at baseline (Table 4a-e)

Assessment of fidelity to intervention

Comparison of frequency and amount of exercise undertaken as recorded in the diaries, % completion of diaries (Table 9). Measurement of adherence to the

intervention as the % of the prescribe dosed of the intervention e.g. 20 minutes , 3 times per week.

Harm data

Harm will be evaluated by a comparison of outcomes by treatment group for:

Number and percentage of Adverse events (AE) and relatedness to intervention

ii. Number and percentage of Serious Adverse Events (SAE) and relatedness to intervention (Table 8)

Definitive Trial Sample Size Estimation

A sample size estimate for a definitive trial will be undertaken for the proposed primary outcome. Choice of the primary outcome measure will be based on the feasibility of collecting the data and also the MCID. Estimation of the standard deviation, correlation between baseline and follow-up measures and a clinically meaningful difference will be used in the power calculation.

Missing Data

Missing outcome data will be recorded at each time point (Table 7). In particular:

Participant recorded diary – Mean (SD) number of days of diary data will be reported, including a percentage of the total number of days (Table 9)

Blinding

Effectiveness of concealment of allocation up to week 10 will be reported by listing the number of instances and circumstances of unblinding (Table 3).

Triangulation of data and progression criteria

Results from all aspects of the qualitative data will be triangulated with the results of the qualitative aspects of the study. This will be done by presenting the qualitative data alongside themes and quotes which detail the lived experience of participants. The triangulated data will determine the suitability of the protocol for incorporation into the main RCT.

Green progression criteria, determined in advance of recruitment and in consultation with the TMG will include minimum monthly recruitment and

retention rates (~70%) and a 90% completion rate of our outcome measures. Amber criteria would be 90% completion rate of the selected primary outcome measure and resetting an acceptable and achievable recruitment and retention rate , acceptable to the TSC. Red criteria: Failure to achieve the Amber criteria will indicate that a full trial is not feasible unless our qualitative study indicates clear means by which the rates may be improved. A recommendation list will be generated to enable refinement of the subsequent RCT protocol.

QUALITATIVE ANALYSIS

Anonymised transcripts of interviews, photographs, diary entries and field notes taken during the fieldwork will be imported into a qualitative data analysis computer software package, NVivo, to enable the organisation and analysis of the data. Photographs will be anonymised as needed. The qualitative data will be analysed in three separate groups:

1) Child data (diaries and photo-elicitation interviews)

2) Parent data (including those who withdrew or declined)

3) Physiotherapist data.

The textual data will be interrogated using Thematic Analysis methods [255].

The first analysis step will involve familiarisation of the text, and then the researcher will code the text by allocating the text fragments to codes. These codes may be revised during the process of reading the transcripts and subthemes developed. For the purpose of rigour, a small sample of data will be analysed by a second person. After this, the codes will be reviewed and themes will be formulated. Finally, meaningful text fragments, sub-themes and themes will be determined related to the study objectives.

Analysis of the photographs will follow the same thematic analysis strategy as described above where the researcher is coding for factors such as type of photograph, setting, people.

Themes arising from the diaries, interviews and photographs will be triangulated in order to check the consistency of the analysis and to generate a deeper understanding of the experience of the participants in order to draft recommendations for the main RCT.

Rigour and Credibility

Preliminary themes arising from the individual data will be feedback to the participants so they can judge whether the analytic interpretations reflect their experiences. This will take place within 2 weeks of the interview.

Checking of the quality of data will take place after every third interview data has been analysed. This quality assessment will be done by sharing the initial codes and themes with the PPI Advisory Group to seek their opinions/perspectives as well as during supervision sessions. Any data shared for this purpose will be anonymous. This will facilitate refinement of future analysis and topic guides.

The position of the researcher as an experienced paediatric physiotherapist could potentially be a limitation and introduce bias to both interviewing and the thematic analysis. For this reason, supervisors will act as a “reflecting team”.

Bias could be introduced, as the research fellow will be aware of which group the child and parents will have been allocated to at week 10. Although this is clearly a limitation, it was felt that as this is a feasibility study and the study is being run by one person (a doctoral student) that this limitation was acceptable as the priority should be to gain data to inform a successful full RCT.

Qualitative data presentation

Demographic data items will be presented using descriptive statistics. This will be for participant interviews, podiatrist interviews and journal entries.

Meaningful text fragments will be determined, as will codes (sub-themes) and themes related to the trial objectives. Data extracts will be accompanied by narrative to elaborate why the extract is analytically interesting.

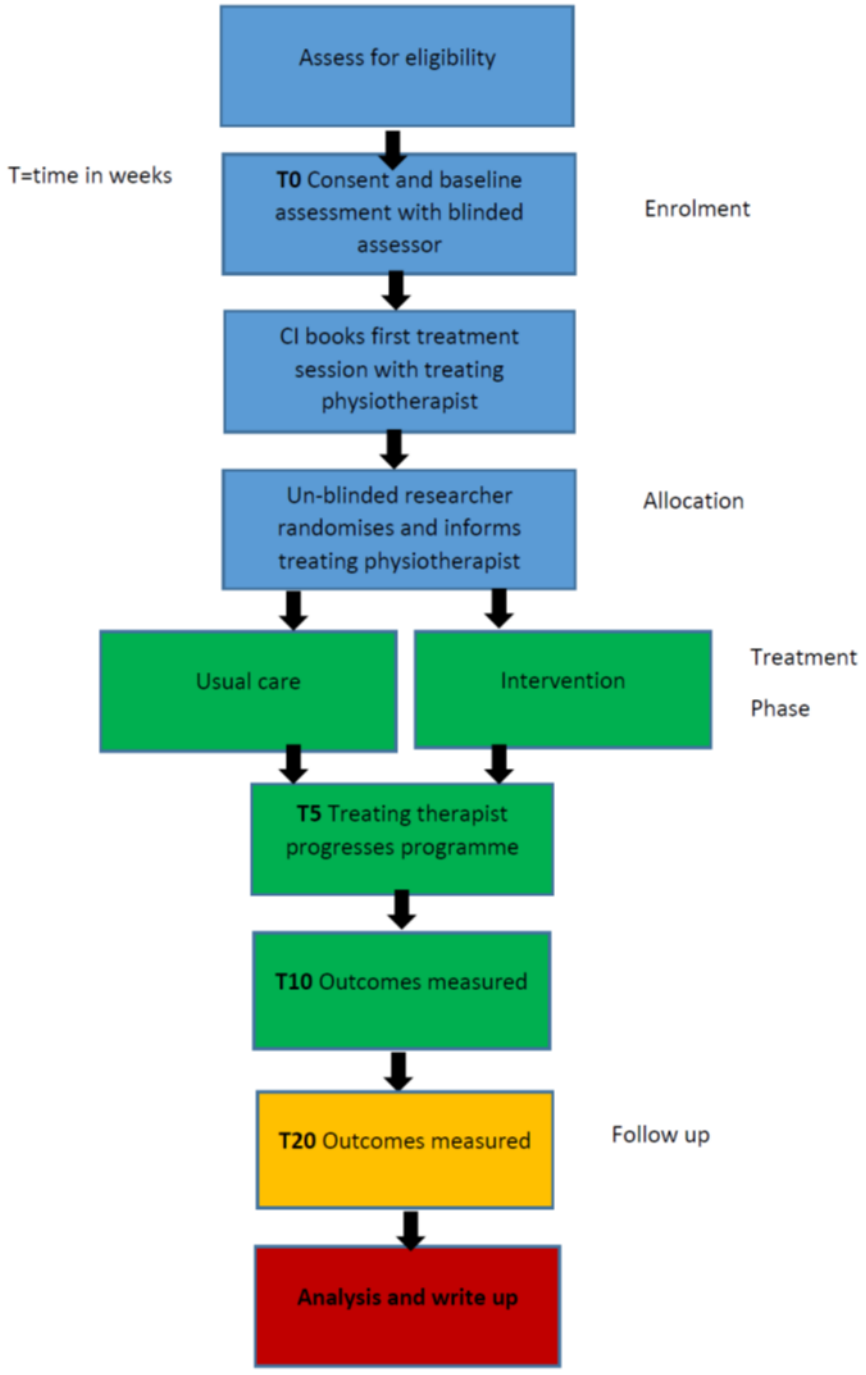
iii. The findings of the analysis will be presented in themes. These will be made meaningful to refine and support recommendations for the main RCT.

REFERENCES

Figure 1 Recruitment and retention flow diagram

Tables Blank Tables 1-9

Figure 2- Trial Flow Diagram



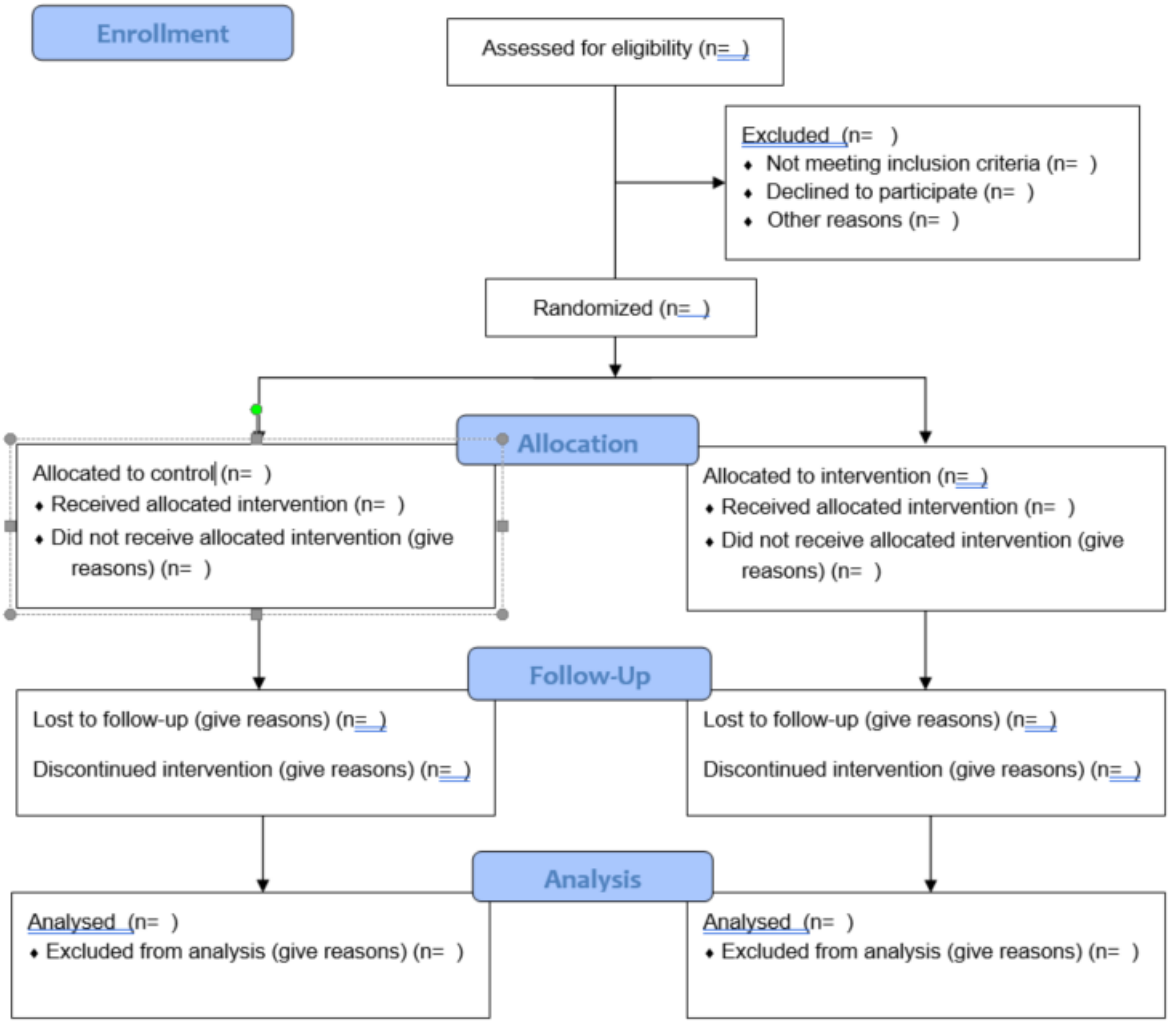

Enrollment
Assessed for eligibility (n=)
Excluded (n=)
Not meeting inclusion criteria (n=)
Declined to participate (n=)
Other reasons (n=)
Randomized (n=)
Allocation
Allocated to control (n=)
Received allocated intervention (n=)
Did not receive allocated intervention (give reasons) (n=)
Allocated to intervention (n=)
Received allocated intervention (n=)
Did not receive allocated intervention (give reasons) (n=)
Follow-Up
Lost to follow-up (give reasons) (n=)
Discontinued intervention (give reasons) (n=)
Lost to follow-up (give reasons) (n=)
Discontinued intervention (give reasons) (n=)
Analysis
Analysed (n=)
Excluded from analysis (give reasons) (n=)
Analysed (n=)
Excluded from analysis (give reasons) (n=)

Table 1 Number and percentage of the total children recruited and retained by site

	TSDFT		UHPT		Total	
	n	%	n	%	n	%
Assessed for eligibility						
PIS packs given out						
Did not meet criteria during caseload screening						
Declined to participate						
Consented but not eligible						
Participants randomised						
Participants allocated to control						
Participants allocated to intervention						
Participants who received the allocated intervention						
Participants who did not receive the allocated to intervention						
Participants lost to follow up						
Participants who discontinued the intervention						
	Mean (SD)		Mean (SD)		Mean (SD)	
Mean (SD) time taken from identification to consent (days)						

Table 2 Participant Demographics and group allocation

Patient characteristics	Total N=xx	Interactive Exercise Trainer N=xx	Usual Care N=xx	Withdrawn	Lost to follow up
Age (years) Mean (SD)					
GMFCS level Median(Q1,Q3)					
Distribution of Impairment n= Bilateral: right hemi: left hemi					
Gender m:f (n=)					
Height (cm) Mean (SD)					
Weight (kg) Mean (SD)					
Hip migration % right: left					
Functional Mobility Scale 5m (0-6) median(Q1,Q3)					
Functional Mobility Scale 50m (0-6) median(Q1,Q3)					
Functional Mobility Scale 500m (0-6) median(Q1,Q3)					

Table 3 Effectiveness of concealment of allocation

Patient characteristics	Total n=	Interactive Exercise Trainer n=	Usual Care n=
Allocation revealed before T10			
Cause of unblinding			

Table 4a Patient characteristics – Physical activity and physiotherapy and 4b medical history at baseline 4c surgical history 4d Medications 4e Orthotics and walking aids

Participant Characteristic	All Participants N=xx	Interactive Exercise Trainer N=xx	Usual Care N=xx
Usual amount of sport or physical activity per week (hours) (Mean(SD))			
Amount Physio contacts per year(Mean(SD))			
Time spent carrying out physio programme per week (mins) (Mean(SD))			

Number of medical conditions	All Participants	Interactive Exercise Trainer N=	Usual Care N=
Fatigue			
Congenital /genetic condition			
Epilepsy			
Learning Disability			
ADHD			
Visual Impairment			
Communication needs			
Asthma			
Constipation			
Other			

Number of previous surgical interventions	All Participants	Interactive Exercise Trainer N=	Usual Care N=
Selective Dorsal Rhizotomy			
Previous Botulinum toxin gastrocnemius			
Previous Botulinum toxin hamstrings			
Previous Botulinum toxin hip adductors			
Previous Botulinum toxin tibialis posterior			

Femoral derotation osteotomy			
Tibial derotation osteotomy			
Gastrocnemius lengthening			
Hamstring lengthening			
Adductor lengthening			
Other			
Deep Brain Stimulator			

Number of medication or class of drug	All Participants	Interactive Exercise Trainer N=	Usual Care N=
Constipation medication			
Bronchodilators			
Anti-epileptics			
Epilepsy rescue medication			
Baclofen			
Melatonin			
Anti-spasmodic			
Other			
Other			
Other			

Number and type of orthotics and walking aids	All Participants	Interactive Exercise Trainer N=	Usual Care N=
Insoles			
AFOs			
DAFOS			
GRAFOs			
DEFOs			
FES			
Wheeled walker			
1 crutch/stick			
2 crutches/sticks			
Manual wheelchair			
Powered wheelchair			

Table 5 Number of concurrent interventions or potential confounding factors during intervention and follow up that may affect balance or walking

Number of Concurrent Intervention	All Participants	Interactive Exercise Trainer N=	Usual Care N=	Withdrawn	Lost to follow up
Bony Surgery					
Soft tissue surgery					
Botulinum toxin					
Change in spasticity medication					

Change in postural management					
New diagnosis					
Change in eligibility					
Other					

Table 6a Comparison of values of gait, balance and Next Step assessment scores and 6b changes from baseline scores 6c differences measures of impairment between groups and 6d changes of these measures from baseline.

Outcome	Baseline		10 weeks		20 weeks	
	Interactive Exercise Trainer N=	Usual Care N=	Interactive Exercise Trainer N=	Usual Care N=	Interactive Exercise Trainer N=	Usual Care N=
Knee angle Midstance (°)Mean (SD)						
Knee angle trough to peak (°)Mean (SD)						
Ankle angle at initial contact (°)Mean (SD)						

Ankle angle at midstance (°)Mean (SD)						
Pediatric balance scale Median (Q1,Q3)						
Peak ML COM Paretic leg(mm) Mean (SD)						
Peak ML COM non-paretic leg(mm) Mean (SD)						
Peak AP COM Paretic leg(mm) Mean (SD)						
Peak AP COM non-paretic leg(mm)						

Mean (SD)						
Velocity ML COM Paretic leg(mm) Mean (SD)						
Velocity ML COM non-paretic leg(mm) Mean (SD)						
Velocity AP COM Paretic leg(mm) Mean (SD)						
Velocity AP COM non-paretic leg(mm) Mean (SD)						
stepping error (mm) Paretic leg Mean (SD)						

stepping error (mm)non-paretic leg Mean (SD)						

6b Change from baseline scores

Outcome	Baseline		Change at 10 weeks		Change at 20 weeks	
	Interactive Exercise Trainer N=	Usual Care N=	Interactive Exercise Trainer N=	Usual Care N=	Interactive Exercise Trainer N=	Usual Care N=
Knee angle Midstance (°)Mean (SD)						
Knee angle trough to peak (°)Mean (SD)						
Ankle angle at initial contact (°)Mean (SD)						
Ankle angle at midstance						

(°)Mean (SD)						
Pediatric balance scale Median (Q1,Q3)						
Peak ML COM Paretic leg(mm) Mean (SD)						
Peak ML COM non-paretic leg(mm) Mean (SD)						
Peak AP COM Paretic leg(mm) Mean (SD)						
Peak AP COM non-paretic leg(mm) Mean (SD)						

Velocity ML COM Paretic leg(mm) Mean (SD)						
Velocity ML COM non-paretic leg(mm) Mean (SD)						
Velocity AP COM Paretic leg(mm) Mean (SD)						
Velocity AP COM non-paretic leg(mm) Mean (SD)						
stepping error (mm) Paretic leg Mean (SD)						
stepping error						

(mm)non-paretic leg Mean (SD)						

6c differences measures of impairment between groups

Outcome	Baseline		10 weeks		20 weeks	
	Interactive Exercise Trainer N=	Usual Care N=	Interactive Exercise Trainer N=	Usual Care N=	Interactive Exercise Trainer N=	Usual Care N=
Right Hamstrings(°) R1 Mean (SD)						
Right Hamstrings(°) R2 Mean (SD)						
Left Hamstrings(°) R1 Mean (SD)						
Left Hamstrings(°) R2 Mean (SD)						
Right Hip Adductors (°) R1 Mean (SD)						
Right Hip Adductors (°) R2 Mean (SD)						
Left Right Hip Adductors (°) R1 Mean (SD)						

Left Right Hip Adductors (°) R2 Mean (SD)						
Right Gastrocnemius (°) R1 Mean (SD)						
Right Gastrocnemius Hip Adductors (°) R2 Mean (SD)						
Left Right Gastrocnemius Hip Adductors (°) R1 Mean (SD)						
Left Right Gastrocnemius (°) R2 Mean (SD)						
Right Duncan Ely (°) R1 Mean (SD)						
Right Duncan Ely (°) R2 Mean (SD)						
Left Right Duncan Ely (°) R1 Mean (SD)						
Left Right Duncan Ely (°) R2 Mean (SD)						
Muscle strength right hip abductors (kg) Mean (SD)						
Muscle strength left hip abductors (kg) Mean (SD)						

Muscle strength right hip adductors (kg) Mean (SD)						
Muscle strength left hip adductors (kg) Mean (SD)						
Muscle strength right ankle dorsiflexors (kg) Mean (SD)						
Muscle strength left ankle dorsiflexors(kg) Mean (SD)						
Muscle strength right quadriceps (kg) Mean (SD)						
Muscle strength quadriceps(kg) Mean (SD)						

6d Changes from baseline measures of impairment between groups at 10 and 20 weeks

Outcome	Baseline		10 weeks		20 weeks	
	Interactive Exercise Trainer N=	Usual Care N=	Interactive Exercise Trainer N=	Usual Care N=	Interactive Exercise Trainer N=	Usual Care N=
Right Hamstrings(°) R1 Mean (SD)						

Right Hamstrings(°) R2 Mean (SD)						
Left Hamstrings(°) R1 Mean (SD)						
Left Hamstrings(°) R2 Mean (SD)						
Right Hip Adductors (°) R1 Mean (SD)						
Right Hip Adductors (°) R2 Mean (SD)						
Left Right Hip Adductors (°) R1 Mean (SD)						
Left Right Hip Adductors (°) R2 Mean (SD)						
Right Gastrocnemius (°) R1 Mean (SD)						
Right Gastrocnemius Hip Adductors (°) R2 Mean (SD)						
Left Right Gastrocnemius Hip Adductors (°) R1 Mean (SD)						

Left Right Gastrocnemius (°) R2 Mean (SD)						
Right Duncan Ely (°) R1 Mean (SD)						
Right Duncan Ely (°) R2 Mean (SD)						
Left Right Duncan Ely (°) R1 Mean (SD)						
Left Right Duncan Ely (°) R2 Mean (SD)						
Muscle strength right hip abductors (kg) Mean (SD)						
Muscle strength left hip abductors (kg) Mean (SD)						
Muscle strength right hip adductors (kg) Mean (SD)						
Muscle strength left hip adductors (kg) Mean (SD)						
Muscle strength right ankle dorsiflexors (kg) Mean (SD)						
Muscle strength left ankle						

dorsiflexors(kg) Mean (SD)						
Muscle strength right quadriceps (kg) Mean (SD)						
Muscle strength quadriceps(kg) Mean (SD)						

Table 7 -7a Completeness of data- Number of outcomes collected at each timepoint and 7b Number, proportion and reasons for non-attendance

Outcome	Baseline		10 weeks		20 weeks	
	n	%	n	%	n	%
Knee angle Midstance (°)Mean (SD)						
Knee angle trough to peak (°)Mean (SD)						
Ankle angle at initial contact (°)Mean (SD)						
Ankle angle at midstance (°)Mean (SD)						
Pediatric balance scale Median (Q1,Q3)						
Peak ML COM Paretic leg(mm) Mean (SD)						
Peak ML COM non-paretic leg(mm) Mean (SD)						
Peak AP COM Paretic leg(mm) Mean (SD)						
Peak AP COM non-paretic leg(mm) Mean (SD)						
Velocity ML COM Paretic leg(mm) Mean (SD)						

Velocity ML COM non-paretic leg(mm) Mean (SD)						
Velocity AP COM Paretic leg(mm) Mean (SD)						
Velocity AP COM non-paretic leg(mm) Mean (SD)						
Stepping error (mm) Paretic leg Mean (SD)						
Stepping error (mm)non-paretic leg Mean (SD)						
Right Hamstrings(°) R1 Mean (SD)						
Right Hamstrings(°) R2 Mean (SD)						
Left Hamstrings(°) R1 Mean (SD)						
Left Hamstrings(°) R2 Mean (SD)						
Right Hip Adductors (°) R1 Mean (SD)						
Right Hip Adductors (°) R2 Mean (SD)						
Left Hip Adductors (°) R1 Mean (SD)						
Left Hip Adductors (°) R2 Mean (SD)						
Right Gastrocnemius (°) R1 Mean (SD)						
Right Gastrocnemius Hip Adductors (°) R2 Mean (SD)						
Left Gastrocnemius Hip Adductors (°) R1 Mean (SD)						
Left Gastrocnemius (°) R2 Mean (SD)						

Right Duncan Ely (°) R1 Mean (SD)						
Right Duncan Ely (°) R2 Mean (SD)						
Left Duncan Ely (°) R1 Mean (SD)						
Left Duncan Ely (°) R2 Mean (SD)						
Muscle strength right hip abductors (kg) Mean (SD)						
Muscle strength left hip abductors (kg) Mean (SD)						
Muscle strength right hip adductors (kg) Mean (SD)						
Muscle strength left hip adductors (kg) Mean (SD)						
Muscle strength right ankle dorsiflexors (kg) Mean (SD)						
Muscle strength left ankle dorsiflexors(kg) Mean (SD)						
Muscle strength right quadriceps (kg) Mean (SD)						
Muscle strength left quadriceps(kg) Mean (SD)						

Outcome	Baseline		10 weeks		20 weeks		Reasons for non-attendance
	n	%	n	%	n	%	
Intervention Participants missed assessments							
Control Participants missed assessments							

Table 8 Safety of the intervention showing number and type of adverse events, and relatedness to the intervention

	Number	Type	Number Related to Intervention
Adverse Events			
Serious adverse events			

Table 9 Completion of diaries and adherence to intervention

	Intervention
Number of diary entries Mean (SD)	
% completed diary returns	
% adherence to prescribed does of intervention	
Number of participants unable to access the intervention	
Number of photos submitted Mean (SD)	
Number of exercise sessions per week Mean (SD)	
Number of minutes of exercise per week Mean (SD)	
Number of times unable to train due to unavailability of equipment Mean (SD)	
Amount of NHS staff hours per week needed to support intervention Mean (SD)	
Amount travel time (mins) needed access intervention Mean (SD)	
Cost of travel to access intervention Mean (SD)	

Appendix 7- **Pediatric Balance Scale**

PEDIATRIC BALANCE SCALE

Name: _____ Date: _____

Location: _____ Examiner: _____

Item Description		Score *0 - 4*	Seconds *optional*
1.	Sitting to standing	_____	
2.	Standing to sitting	_____	
3.	Transfers	_____	
4.	Standing unsupported	_____	_____
5.	Sitting unsupported	_____	_____
6.	Standing with eyes closed	_____	_____
7.	Standing with feet together	_____	_____
8.	Standing with one foot in front	_____	_____
9.	Standing on one foot	_____	_____
10.	Turning 360 degrees	_____	_____
11.	Turning to look behind	_____	
12.	Retrieving object from floor	_____	
13.	Placing alternate foot on stool	_____	_____
14.	Reaching forward with outstretched arm	_____	
	Total Test Score	_____	