# Bike skills training for children with cerebral palsy

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# DEDICATION

For my son, Ernest George Toovey Milsom.

# ABSTRACT

Training targeted towards goals that are meaningful to children with cerebral palsy (CP) and their families is needed to improve function and support participation in physical activities in this population. Riding a two-wheel bike is a common goal for ambulant children with CP, yet little specific evidence exists to guide clinicians and families. This thesis developed and tested a task-specific approach to training bike skills in this population through three studies: 1) a systematic review, 2) a practice survey, and 3) a randomised controlled trial (RCT). Each study used the International Classification of Functioning, Health and Disability (ICF) as a framework.

While strong evidence exists for task-specific training (TST) for improved upper limb (UL) function in this population, prior to this thesis the literature regarding TST for gross motor skills, including bike riding, in ambulant children with CP had not been systematically appraised. Thus, Study 1 aimed to systematically evaluate the effectiveness of task-specific gross motor skills training for improving activity and participation outcomes in ambulant school-aged children with CP. This review involved 13 studies of low-to-moderate overall quality and found effects of TST were positive for participation-related outcomes, and mixed for specific skill performance and functional skills, while little or negative effects were found for general gross motor skills. This study identified the need for higher quality studies and reporting that enables evidence synthesis.

Given the importance of understanding current practice when designing effectiveness studies, Study 2 involved a survey of 95 physiotherapists (PTs) and occupational therapist (OTs) in Australia about their practices when training two-wheel bike skills in children with CP. This study found that while functional approaches to training and goal-based assessment and evaluation were predominant, overall practices appear highly variable. Moreover, the need to develop and test bike-specific measures and interventions in this population was highlighted.

The findings from Studies 1 and 2 informed the design of Study 3. This multi-site assessor-blind RCT aimed to determine if a task-specific approach was more effective than a parent-led home program for attaining individualised two-wheel bike riding goals

in ambulant children with CP. Sixty-two children were randomly allocated to either the task-specific approach (n=31) or home program (n=31). The primary finding was that the task-specific program was more effective than the home program for goal attainment at one week post-intervention. Greater odds of goal attainment were retained at three months and evidence of better outcomes following the task-specific program were found for some outcomes related to participation in bike riding, physical activity and self-perception. In addition, there was evidence of mixed effects for functional skills, and little difference in bike skills and health-related quality of life.

While each of these studies provides an original contribution to the literature, together they form a significant foundation for evidence on training bike skills in ambulant children with CP. Use of the ICF across the thesis meant findings could be synthesised and enhanced the clinical relevance of the research. Given that an effective approach for attaining two-wheel bike riding goals in this population now exists, training for clinicians to optimise knowledge translation should be developed. Future research should seek to understand relationships between bike skills training and a broader range of ICF domains and levels of function in CP, tailor interventions to individuals and determine longer-term outcomes.

# DECLARATION

This is to certify that:

(i) This thesis comprises only my original work towards the Doctor of Philosophy;

(ii) Due acknowledgement has been made in the text to all other materials used;

(iii) The thesis is fewer than 100,000 words in length, exclusive of tables, maps, bibliographies and appendices.

_____

# PREFACE

This thesis with publication is comprised of seven chapters, four of which report studies carried out in collaboration with others. Chapters 3, 4 and 5 are published manuscripts and Chapter 6 is a manuscript prepared for submission. No part of this thesis has been submitted for other qualifications or was carried out prior to enrolment in the PhD. No third-party editorial assistance was provided in preparation of the thesis. The publication status, contributions of each author and funding sources regarding each of these chapters are described below.

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This manuscript reports the primary and secondary outcomes from the RCT detailed within the protocol publication above.

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# PUBLICATIONS AND PRESENTATIONS DURING CANDIDATURE

## Publications arising from this thesis

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**Toovey R**, Spittle AJ, Nicolaou A, McGinley JL, Harvey AR (2019). Training two-wheel bike skills in children with cerebral palsy: A practice survey of therapists in Australia. *Physical & Occupational Therapy in Pediatrics*. DOI 10.1080/019242638.2019.1585404 [Chapter 4]

**Toovey R,** Harvey AR, McGinley JL, Lee KJ, Shih STF, Spittle AJ (2018). Bike skills training for children with cerebral palsy: Protocol for a randomised controlled trial. *BMJ Open*. 8: e019898. DOI: 10.1136/bmjopen-2017-019898 [Chapter 5]

## Related publications

**Toovey, R.,** Reid, S., Rawicki, B., Harvey, A., & Watt, K. (2017). Ability of independently ambulant children with cerebral palsy to ride a two-wheel bicycle: A case-control study. *Developmental Medicine & Child Neurology*, *59*(4). https://doi.org/10.1111/dmcn.13340

## Conference presentations

**Toovey R**, Harvey AR, McGinley JL, Lee KJ, Shih STF, Spittle AJ. A randomised controlled trial of a novel task-specific approach for attaining two-wheel bike riding goals in children with cerebral palsy. Accepted for presentation at the *American Academy of Cerebral Palsy and Developmental Medicine Conference* in Anaheim, September 2019.

**Toovey R**, Harvey AR, McGinley JL, Lee KJ, Shih STF, Spittle AJ. Effectiveness of a novel task-specific approach for attaining two-wheel bicycle relared goals in ambulant children with cerebral palsy: A randomised controlled trial. Oral presentation at the *European Academy of Childhood Disability Meeting* in Paris, May 2019.

**Toovey R**, Harvey AR, McGinley JL, Spittle AJ. Training two-wheel bicycle skills in children with cerebral palsy in Australia: A current practice survey of physiotherapists and occupational therapists. Poster presentation at the *Australasian Academy of Cerebral Palsy and Developmental Medicine* conference in Auckland, March 2018.

**Toovey R,** Bernie C, Harvey AR, McGinley JL, Spittle AJ. Task-specific gross motor skills training for ambulant school-aged children with cerebral palsy: A systematic review. Oral presentation at the *Australian Physiotherapy Association conference* in Sydney, October 2017.

**Toovey R**, Harvey AR, McGinley JM, Spittle AJ. Current practices of training bike riding skills in children with cerebral palsy in Australia. Poster presentation at the *Australian Physiotherapy Association conference* in Sydney, October 2017.

**Toovey R**, Harvey AR, McGinley JM, Lee KJ, Shih STF, Spittle AJ. Bike skills training for children with cerebral palsy: Protocol for a randomized controlled trial. Oral presentation at the *Participation: Motives, Methods and Measures symposium*, Melbourne, March 2017.

**Invited presentations**

**Toovey R**, Edwards L, Spencer T, Daniels L. Sports Participation Pathways: Therapy, Community, Competition. Invited lecture to the *Australian Physiotherapy Association Victorian Paediatric and Disability Groups*, Melbourne, September 2019.

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MCRI Postgraduate Health Research Top-Up Scholarship (2015 – 2019)

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# GLOSSARY OF ABBREVIATIONS

| | |
|---|---|
| 6MWT | Six-minute walk test |
| AACPDM | American Academy of Cerebral Palsy and Developmental Medicine |
| ASD | Autism spectrum disorder |
| BoNT-A | Botulinum toxin-A |
| COPM | Canadian Occupational Performance Measure |
| CRE-CP | Centre of Research Excellence in Cerebral Palsy |
| CP | Cerebral palsy |
| CHU-9D | Child Health Utility - Nine Domains |
| Comp | Comparison program |
| CI | Confidence interval |
| CONSORT | Consolidated Standard of Reporting Trials |
| GAS | Goal attainment scale |
| GRADE | Grading of Recommendations, Assessment, Development and Evaluations |
| GMFCS | Gross Motor Function Classification System |
| GMFM | Gross Motor Function Measure |
| HABIT-ILE | Hand-arm bimanual intensive therapy including lower extremities |
| HRQoL | Health-related quality of life |
| ID | Identification number |
| IQ | Intelligence quotient |
| IQR | Interquartile range |
| ITT | Intention-to-treat |
| ICF | International Classification of Functioning, Disability and Health |
| LL | Lower limb |
| MACS | Manual Abilities Classification System |
| MD | Mean difference |
| Monash | Monash Children's Hospital |
| MI | Multiple imputation |
| MCRI | Murdoch Children's Research Institute |
| NHMRC | National Health and Medical Research Council |
| NDT | Neurodevelopmental Therapy |

| | |
|---|---|
| OR | Odds ratio |
| OT | Occupational therapist |
| PICF | Participation information and consent form |
| PEDI | Pediatric Evaluation of Disability Inventory |
| PEDI-CAT | Pediatric Evaluation of Disability Inventory - Computer Adapted Test |
| PEDI-NL | Pediatric Evaluation of Disability Inventory - Dutch Version |
| PAQ-C | Physical Activity Questionnaire for Children |
| PhD | Doctor of Philosophy |
| PT | Physiotherapist |
| PROSPERO | International Prospective Register of Systematic Reviews |
| PRISMA | Preferred Reporting Items for Systematic Reviews and Meta-Analysis |
| RCT | Randomised controlled trial |
| REDCap | Research Electronic Data Capture |
| RCH | Royal Children's Hospital |
| SPP | Self-Perception Profile |
| SD | Standard deviation |
| SMD | Standardised mean difference |
| TIDiER | Template for intervention description and replication |
| TSP | Task-specific program |
| TST | Task-specific training |
| TD | Typically developing |
| UL | Upper limb |
| US | United States |
| VCPR | Victorian Cerebral Palsy Register |
| VPRS | Victorian Paediatric Rehabilitation Service |

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# CHAPTER 1: INTRODUCTION

*Overview of the clinical issue to address, thesis outline and significance*

## 1.1. An overview of the clinical issue to address

Cerebral palsy (CP) is the most common cause of childhood physical disability with a prevalence of 1.4 per 1000 live births (Australian Cerebral Palsy Register, 2018). It is a group of disorders of the development of movement and posture, causing activity limitations attributed to non-progressive disturbances that occurred in the developing fetal or infant brain (Bax et al., 2005). The most widely used means of classifying the motor function of children with CP is the Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997). The GMFCS uses five levels (I-V) to classify children by motor function with GMFCS I being the least impaired and GMFCS V being the most impaired. Children classified as levels I-II are independently ambulant with or without hand-held devices.

Children with CP at all GMFCS levels, including levels I and II, participate less in physical and recreational activities than their typically developing (TD) peers (Bjornson, Belza, Kartin, Logsdon, & McLaughlin, 2007). Low levels of physical activity in childhood are associated with chronic disease in adulthood (Fernandes & Sansecso, 2010) and participation in meaningful activities is key to all children's development (Miller, Ziviani, Ware, & Boyd, 2015). Effective means of engaging children with CP in interventions are required to improve functional outcomes including motor performance, physical activity, and participation. To do this, clinicians need to provide evidence-based training targeted towards goals of the child and their family (Novak et al., 2013).

Bike riding is a functional activity that involves the performance of a skill and participation in physical activity. Learning to ride a two-wheel bike is considered a common goal of childhood (Munro & Arnold, 2015). For TD children, this is often done in home or community environments and is led by parents (Toovey, Reid, Rawicki, Harvey, & Watt, 2017). The positive effects of cycling as a peer, family and community activity in the

general population are well known (Raje & Saffrey, 2016). These include personal benefits such as improved participation in physical and social activities, as well as economic and environmental benefits.

Two-wheel bike riding may be an effective means for involving children with CP in a physical activity that is meaningful to them. While it is possible for this population to ride a two-wheel bike, currently fewer children with CP aged 6-15 years and classified GMFCS I-II have learnt to ride than their TD peers and those who do ride, learned later (Toovey, Reid, et al., 2017). The paucity of specific evidence available to guide clinicians and families may contribute to this gap between ability and performance – no studies training dynamic bike skills for bike-riding related outcomes in this population have been reported. The literature in CP focuses on stationary cycling (Armstrong, Spencer, et al., 2019) or on outcomes not directly related to bike riding (Mohanty, Meshram, & Pattnaik, 2015), while low quality evidence exists for training bike skills in children with disabilities more broadly (Dunford, Rathmell, & Bannigan, 2016; Halayko, Magill-Evans, Smith, & Polatajko, 2016; Macarthur, Parkin, Sidky, & Wallace, 1998; MacDonald et al., 2010; Ulrich, Burghardt, Lloyd, Tiernan, & Hornyak, 2011). These findings support the need to develop and test training approaches when working with ambulant children with CP with two-wheel bike riding goals.

Task-specific training focuses on training of context-specific functional tasks through goal-directed practice and repetition (Hubbard, Neilson, & Carey, 2009). This approach is informed by the principles of motor learning and dynamic systems theory (Thelen & Smith, 2007; Wolpert, Ghahramani, & Flanagan, 2001). Systematic meta-analyses have found positive functional outcomes from UL training involving task-specificity in this population (Sakzewski, Gordon, & Eliasson, 2014; Sakzewski, Ziviani, & Boyd, 2014) and from task-specific locomotor training in adults post-stroke (French et al., 2007). This approach may be effective for training bike skills in children with CP. Yet, to my knowledge, until this thesis no systematic evaluation of TST for gross motor skills, such as bike riding, had been conducted.

In addition to a strong theoretical basis and relevant evidence base, it is important to understand current clinical practice when developing an intervention for evaluation, particularly by RCT (Craig et al., 2013). However, current two-wheel bike skills training practices in children with CP are not known. When determining the effectiveness of an intervention, it is also pertinent to select appropriate outcomes to judge this upon. These outcomes need to be clinically relevant, able to be captured by measurement tools with strong psychometric properties and meaningful to the child and family (Coulter, 2017; Oeffinger et al., 2008).

## 1.2. Aims of the thesis

The overall aim of this thesis is to develop and test the effectiveness of a task-specific approach to training two-wheel bike skills in children with CP. The specific aims are to:

i. Evaluate and synthesise the evidence for the effectiveness of task-specific gross motor skills training in ambulant school-aged children with CP;
ii. Examine the current practices of Australian physiotherapists and occupational therapists when training two-wheel bike skills in children with CP. This includes determination of eligibility, initial assessment, training characteristics, and outcome measurement;
iii. Determine if a task-specific approach to training two-wheel bike skills is more effective than a parent-led home program in children with CP for
    a. Primary outcome: attaining individualised two-wheel bike-specific goals at one week post-intervention
    b. Secondary outcomes: acquiring and retaining two-wheel bike skills, improving functional skills, self-perception, health-related quality of life and participation in two-wheel bike riding at one week and three months post-intervention; and goal attainment and physical activity at three months post-intervention.

## 1.3. Thesis outline

This thesis consists of seven chapters including three studies. Chapters 3 to 5 (Studies 1, 2 and Methods for Study 3) are included as published manuscripts and Chapter 6 (Study 3) is included as a manuscript prepared for submission. The structure of the thesis is shown in Figure 1.1. The map illustrates that the overall literature review identifies gaps in the evidence and informs the specific systematic review of task-specific gross motor skills training and the survey of current practice. These studies guide the design of the primary study of the thesis, an RCT. Each study is then considered in the Grand Discussion chapter.

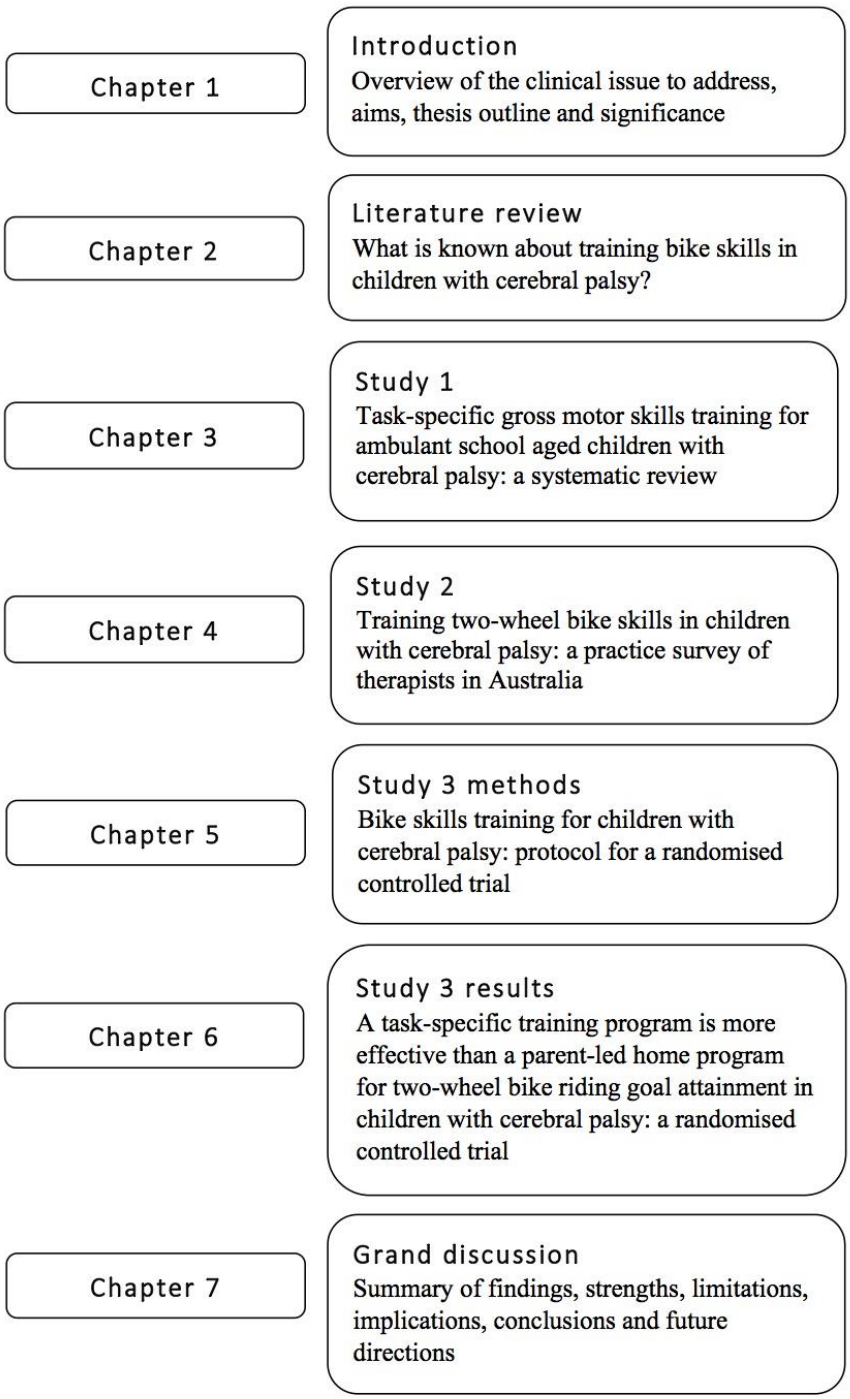



**Figure 1.1** Thesis outline

**Chapter 2** is a **Literature Review** in response to the research question *What is known about training two-wheel bike skills in children with CP?* The International Classification of Functioning, Disability, and Health (ICF) (World Health Organisation, 2001), a well-recognised conceptual framework for understanding health and disability is introduced. Motor impairments, activity limitations and participation restrictions related to CP are described and bike riding as a motor skill, physical activity, and means of participation is explored. This chapter identifies gaps in studies of two-wheel bike skills training in children with CP. Thus, the literature on bike skills training in other paediatric populations is considered and approaches to training motor skills in CP are reviewed with a focus on TST. Finally, frameworks and tools for measuring outcomes following two-wheel bike skills training this population are reviewed.

**Chapter 3** addresses the first aim of this thesis by evaluating and synthesising the effectiveness of task-specific gross motor skills training in ambulant school-aged children with CP through a **systematic review (Study 1)**. Standardised tools are used to determine the level of evidence and risk of bias within included studies and the quality of the evidence across activity and participation outcomes. Motor learning strategies reported within TST are also identified and any relationship to outcome is assessed.

**Chapter 4** responds to the second aim of this thesis by describing current practices when training two-wheel bike skills in children with CP **(Study 2)** through a **survey** of physiotherapists and occupational therapists in Australia. Practices relating to considerations for eligibility, initial assessment, training characteristics and evaluation of training effectiveness are reported within an ICF framework.

**Chapters 5** and **6** relate to the main study **(Study 3)** within the thesis: an **RCT** of a task-specific approach to training two-wheel bike skills compared to a parent-led home program in ambulant children with CP. Chapter 5 describes the methods for Study 3 in the form of the trial protocol. Chapter 6 addresses the third aim of this thesis. The primary outcome, individualised bike-specific goal attainment, and secondary outcomes; two-wheel bike skills, functional skills, self-perception, physical activity and participation in bike riding, of the trial are reported in this chapter.

**Chapter 7**, the **Grand Discussion**, brings together the results of the three studies within the thesis. Main findings are synthesised using the ICF as a framework, and strengths and limitations of the thesis are identified. The significance of the PhD in the context of previous work is considered and implications for practice and policy are discussed. Finally, future research directions are proposed, and conclusions of the thesis are made.

## 1.4. Significance of this research

While CP is a childhood-onset disability, its impact extends across the lifespan of the individual, into families and communities and across multiple sectors including health, disability, sport and education (Australian Institute of Health and Welfare, 2006). It is well documented that children with CP currently have poorer health and participation outcomes in adulthood compared to their TD peers (Bottos, Feliciangeli, Sciuto, Gericke, & Vianello, 2001; Imms & Adair, 2016; Peterson, Ryan, Hurvitz, & Mahmoudi, 2015), and that experiences in childhood influence these outcomes (Moore, Arefadib, Deery, Keyes, & West, 2017). Of relevance is the association between greater physical activity in childhood and both higher levels of physical activity and reduced risk of chronic disease in adulthood (Fernandes & Sansecso, 2010).

Increasing children's physical activity is a national health priority (Australian Health Policy Collaboration, 2018). The need for action is magnified for children with CP due to these documented poorer health outcomes. To achieve this, effective means of engaging this population in functional interventions with meaningful outcomes are needed (Damiano, 2006; Vargus-Adams & Martin, 2009). This has been recognised in the current roll-out of the National Disability Insurance Scheme in Australia where participants' goals directly influence the services and supports funded (National Disability Insurance Agency, 2016). Riding a two-wheel bike is a common childhood goal and a potential pathway to increased participation in physical and recreational activity (Raje & Saffrey, 2016). However, it can be difficult for children with movement impairments to learn this skill (Toovey, Reid, et al., 2017) and there is a paucity of evidence to guide clinicians and families when working towards bike riding goals.

The three studies included in this thesis contribute new knowledge and can synergise to form a significant body of work. The systematic review (Study 1) is the first study to evaluate and synthesise the evidence for TST for gross motor skills in ambulant school-aged children with CP. It also provides guidance for researchers around strengthening the evidence so that motor interventions tailored to individual goals and lifelong physical activity can be developed. The survey of therapists (Study 2) fills the gap in knowledge around current bike skills training and highlights the variability in approaches in Australia. Both studies inform Study 3 in which this thesis culminates – the first RCT of two-wheel bike skills training in ambulant children with CP. Guidance from the systematic review and the survey informed the rigorous yet pragmatic design of this study. This trial has provided high-quality and clinically relevant evidence supporting a task-specific approach for the attainment of two-wheel bike-specific goals. The reporting of both the methods and results of the trial enhances the potential for the approach to be replicated globally and in other populations. Guidance for clinicians when working with children with CP who have bike riding goals now exists, potentially providing more children with the opportunity to learn to ride and participate in physical activity. Moreover, the functional nature of the intervention and focus of outcomes measured may be meaningful for children and their families and encourage engagement in bike riding.

# CHAPTER 2: LITERATURE REVIEW

*What is known about two-wheel bike skills training in children with cerebral palsy?*

## 2.1. Introduction

The thesis seeks to develop and test a novel approach to training two-wheel bike skills for children with CP. This chapter addresses the key first step in achieving this by considering, "*What is known about training bike skills in this population*?" First, a framework for understanding disability and health is introduced and is used to describe CP and bike riding, and guide the review of literature pertaining to this research question. The evidence specific to bike skills training in CP is reviewed including knowledge of bike riding ability, intervention studies targeting bike skills and understanding of current therapy practices. In the absence of extensive research on this specific topic, gaps in the literature are identified and a wider net is cast. The relevant evidence regarding bike skills training in other developmental disabilities and motor skills training in CP more broadly is considered and further directions for future research emerge. Finally, goal-based tools and a model for understanding participation in childhood disability are presented for consideration in designing a study that measures meaningful activity and participation outcomes.

## 2.2. The International Classification of Functioning, Disability and Health

The World Health Organisation's ICF provides a framework for describing, understanding and studying disability and health, and related outcomes and determinants (see Figure 2.1) (World Health Organisation, 2001). It integrates the medical and social models of disability and health into a biopsychosocial model. It conceptualises functioning as a dynamic interaction between the health condition, and the contextual – environmental and personal factors. Within the ICF framework, functioning involves the body structures and functions, activity and participation domains. Body structures and functions involve physiological functions and anatomical parts of the body, activities are defined as the execution of a task whilst participation refers to involvement in a life situation. Environmental factors involve the external influences on functioning such as social attitudes, policy and the built

environment while personal factors are the considerations that influence how individuals experience their disability or health condition including age, behaviour and social circumstances.

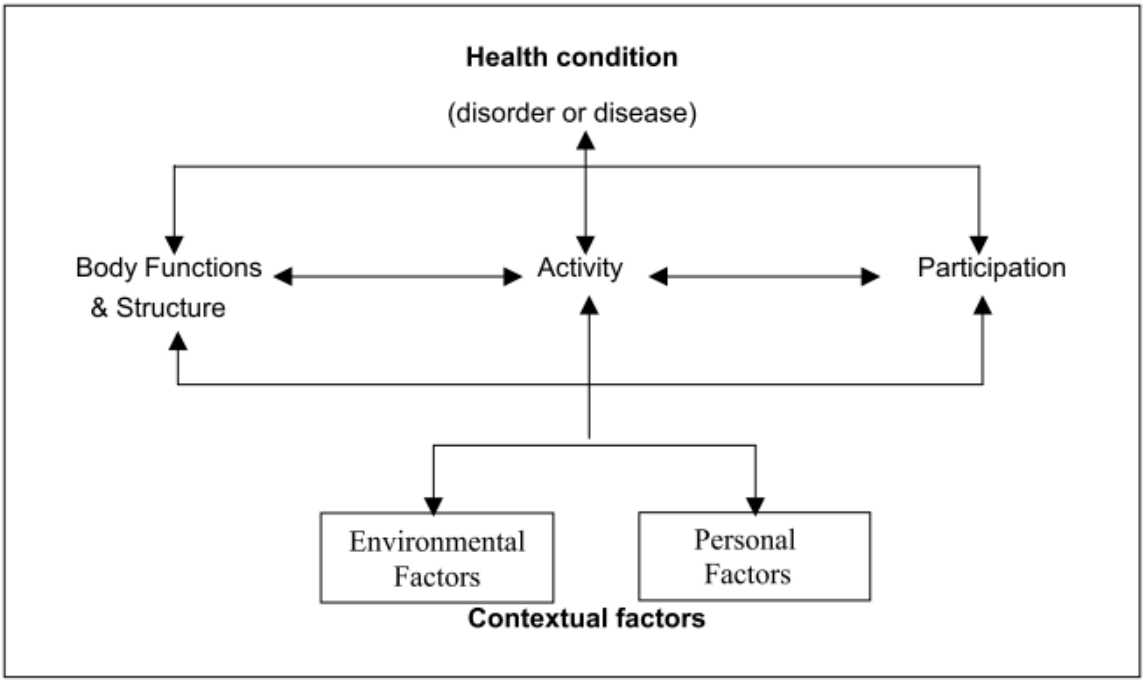



**Figure 2.1:** Diagrammatic representation of the International Classification of Functioning, Disability and Health (World Health Organisation, 2002, p9). Permission to reproduce not required by the World Health Organisation.

The ICF can be used at individual, institutional and social levels to better understand health and disability. Since its publication, this framework has become the common language for clinicians and families in determining the goals and management of children with disabilities (Rosenbaum, 2003). It is also increasingly being used to inform, analyse and report research interventions and outcomes (World Health Organisation, 2002). This has signalled a shift in focus in clinical and research practice towards interventions for children with disabilities that target activity and participation outcomes rather than outcomes in the

body structures and functions domain (Novak et al., 2013; Rosenbaum & Gorter, 2012). Further, use of the framework across research studies enables findings to be synthesised in a consistent and meaningful way.

## 2.3. Cerebral palsy

Cerebral palsy is the most common cause of childhood physical disability in Australia affecting 1.4 births in every thousand (Australian Cerebral Palsy Register, 2018). Cerebral palsy is an umbrella term for a group of permanent but not unchanging disorders of movement and posture causing activity limitations, and attributed to non-progressive disturbances that occurred in the developing fetal or infant brain (Bax et al., 2005). CP impacts more males than females, and almost half of CP births are preterm (Australian Cerebral Palsy Register, 2018). Individuals with CP can experience limitations across all domains of the ICF.

*Body structure and function impairments* in CP include changes in muscle tone and posture leading to disorders in movement including spasticity, dyskinesia, ataxia and mixed motor types (Graham et al., 2016). These movement disorders can have bilateral (quadriplegic, triplegic, diplegic) or unilateral (hemiplegic, monoplegic) topography and contribute to musculoskeletal impairments in range of motion, muscle strength and postural deformity. Vision, hearing, speech and intellectual impairments, as well as epilepsy, are also associated with CP (Australian Cerebral Palsy Register, 2018).

*Activity limitations* in CP include, but are not limited to, challenges with gross motor function and manual ability. The most widely used means of classifying the gross motor function of children with CP is the GMFCS. (Palisano et al., 1997). The GMFCS uses five levels (I-V) to classify motor function. Children classified as level I are independently ambulant without mobility devices across all settings and while they can typically run and jump, they can experience challenges with speed, coordination and balance. Children classified as level II are also independently ambulant but may use hand-held devices over longer distances and their ability to run and jump is limited. As described in the introduction, as GMFCS level increases, impairment and functional restrictions become greater - children classified as level V are transported in a manual wheelchair in all settings.

The Manual Ability Classification System (MACS) also uses five levels to classify manual ability with a similar grading system used to describe severity of limitations (Eliasson et al., 2006). Children classified MACS I-II handle objects easily or with somewhat reduced quality and speed while those classified MACS V have no or very limited hand function. Most children with CP in Australia are independently ambulant, with 62% classified GMFCS I or II (Australian Cerebral Palsy Register, 2018). Similarly, MACS levels I-II have been reported to represent the manual abilities of most of the CP population in developed settings (Carnahan, Arner, & Hägglund, 2007).

*Restricted participation* including low levels of diversity and intensity in participation; and high levels of non-participation in preferred physical and skill-based activities specifically, have been reported in studies of children with CP across all GMFCS and MACS levels (Bjornson et al., 2007; Imms & Adair, 2016; Imms et al., 2017; Imms, Reilly, Carlin, & Dodd, 2008; Majnemer, Shikako-Thomas, Schmitz, Shevell, & Lach, 2015).

*Environmental and personal factors* can influence body structures and functions and activities, however, recent literature in CP has focused on the interaction of these contextual factors with participation. For example, commonly reported facilitators to participation in this population include mastery motivation, positive self-perception, access to rehabilitation and parental support (Majnemer et al., 2008; Verschuren, Wiart, Hermans, & Ketelaar, 2012), while negative social attitudes, inaccessible physical environments and parental stress can be barriers (Imms, 2008; Majnemer et al., 2008).

## 2.4. Two-wheel bike riding

Cycling is a popular activity for Australian children (Australian Bureau of Statistics, 2012; Munro & Arnold, 2015). There are various forms of cycling including riding a two-wheel bicycle / bike, a balance bike (also known as a walker bike), bike with training wheels, tricycle / trike, recumbent bike or hand-cycle, as well as, riding stationary bicycles (Munro & Arnold, 2015). The focus of this PhD is on the most common form of cycling, two-wheel bike riding in outdoor environments. The terms two-wheel bike riding and bike riding will be used interchangeably to describe this activity.

Bike riding is a specialised gross motor task involving all four limbs, with greater demand on the lower limbs (LL) (Ducheyne, De Bourdeaudhuij, Lenoir, Spittaels, & Cardon, 2013). Independent performance of this skill involves interaction between several physiological systems including motor, cognitive, visual, vestibular and somatosensory systems (Wierda & Brookhuis, 1991; Wozniak Timmer, 1991). It requires components of these systems including, but not limited to, joint range of motion, muscle coordination and power, executive function, visual acuity and balance and proprioception (Raje & Saffrey, 2016). Bike riding involves a continuous closed kinetic system but can be considered an open perceptual-motor skill; there is a dynamic interaction with environmental factors such as surface, gradient and other riders. (Wierda & Brookhuis, 1991). Performance can also be influenced by internal task-related factors including seat height, pedal position, force generation and pedal rate (Fonda, Sarabon, & Li, 2014; Wozniak Timmer, 1991). Outdoor bike riding involves several safety considerations, largely due to the unpredictability of external environments and speed of movement. Thus, helmets are required by law to be worn in Australia (VicRoads: Victorian Government, 2018).

Learning to ride a two-wheel bike is a common childhood milestone (Munro & Arnold, 2015). For TD children in Australia, this is often done in home or community environments between ages four and six and is generally led by parents (Toovey, Reid, et al., 2017). The positive effects of two-wheel bike riding as a peer, family and community activity and form of active travel in the general population are well known. These include personal benefits such as improved cardiovascular fitness, independence, confidence and participation in physical and recreational activities; as well as economic and environmental benefits (Raje & Saffrey, 2016). Bike education programs outside of the family environment do exist. In Australia, these programs are largely run by National and State level cycling organisations and are generally delivered through schools (Cycling Australia, 2018; VicRoads: Victorian Government, 2018). However, bike riding education is not compulsory and the focus of these programs is on safety and higher-level skills, rather than on early motor learning such as riding a two-wheel bike for the first time. Moreover, program materials and activities tend to be targeted towards the TD population and do not address children with motor dysfunction.

## 2.5. Two-wheel bike riding and children with CP

Two-wheel bike riding may be an effective means for involving children with CP, especially those classified within GMFCS I-II, in a physical activity that is meaningful to them with potential benefits at the activity and participation levels. Prior to developing a novel intervention that targets this activity it is important to understand bike riding ability, previous bike skills training research and current clinical practices specific to bike skills in this population.

### 2.5.1 Bike riding ability of children with CP

The early motor development of children with CP is increasingly being investigated (Barbosa, Campbell, Sheftel, Singh, & Beligere, 2003; Lebeer & Rijke, 2003; Morgan et al., 2016). Knowledge of delayed milestones and differing abilities for sitting, crawling and walking has been established, largely through the widespread use of the GMFCS and outcome measures including the Gross Motor Function Measure (GMFM) (Rosenbaum et al., 2002; Russell et al., 1989). However, knowledge around development of complex movement skills beyond walking in this population - those that are common childhood activities, such as bike riding – is less well developed.

Riding a bike is a common therapeutic goal for children with CP (Reedman, Boyd, Trost, Elliott, & Sakzewski, 2019). However, until recently there was no research evidence documenting the ability of ambulant children with CP to ride a two-wheel bike. As part of my Masters-level research, I led a case control study in which parents of 114 children with CP and 87 TD children aged 6-15 years were surveyed regarding their child's two-wheel bike riding ability and age at skill acquisition, if at all (Toovey, Reid, et al., 2017).

Results of this study suggested a substantially lower proportion of children with CP (GMFCS I-II) can ride a two-wheel bike at any given age compared with their TD peers. Overall, 37% of the children in the CP group were reported to ride independently in the community compared with 87% of TD children. Further, while most of those children with CP who did learn to ride also learned at home with their parents, they learned later. The median age at skill acquisition in the CP group was estimated to be 10.9 years, compared

with 5.0 years for the TD group. Factors found to be associated with ability to ride a two-wheel bike included GMFCS level I, higher parent-rated importance of riding a two-wheel bike and increasing age. These factors related to the activity, environmental and personal domains of the ICF.

This study documented the size of the gap in bike skill acquisition between children who are TD and children with CP, even those at the highest levels of function. However, it also highlighted that it is possible for ambulant children with CP to learn to ride. Overall, these findings supported the need for research into training approaches for ambulant children with CP who are not yet riding and have goals to do so.

### 2.5.2 Bike skills training in children with CP

There is sparse evidence to guide best practice training of two-wheel bike riding skills specific to children with CP. To my knowledge, no studies have specifically focused on two-wheel bike riding in this population. A case series study involving a small subset (n=5) of children with CP found a task-oriented two-wheel bike skills intervention resulted in positive skill acquisition, but results were not reported separately for this population (Dunford et al., 2016). Community cycling goals have been documented in studies testing cognitive and participation approaches in children with CP with positive effects on goal attainment (Gimeno, Brown, Lin, Cornelius, & Polatajko, 2019; Reedman et al., 2019). However, in these two studies bike riding goals were related to individual participants only or it was unclear what form of cycling was being targeted.

RCTs involving cycling interventions in children with CP do exist but have been conducted on stationary bikes and focus on outcomes at the body structures and functions domain (Armstrong, Spencer, et al., 2019). Positive effects on strength and endurance (Fowler et al., 2010), muscle co-activation (Johnston, Lauer, & Lee, 2008), and bone mineral density have been documented (Chen et al., 2013, 2012), while improved gross motor function and quality of life in cycling groups compared to controls have also been found (Bryant, Pountney, Williams, & Edelman, 2013; Demuth, Knutson, Fowler, & et al, 2012). An RCT protocol testing a combined functional electrical stimulation cycling and adapted community cycling approach on activity and participation outcomes has recently been

published but results are not yet available (Armstrong, Boyd, Kentish, Carty, & Horan, 2019).

Lower level evidence for cycling interventions specific to this population also exists. A mixed methods study found that adapted dynamic cycling was enjoyable, while a pre-post study involving 30 children compared stationary cycling to dynamic riding with training wheels and found better overall gross motor function in the dynamic cycling group (Mohanty et al., 2015; Pickering, Horrocks, Visser, & Todd, 2013). In any case, there is little evidence to suggest findings from studies involving stationary cycles, adapted cycles or bikes with training wheels translate to riding a two-wheel bike in the community.

### 2.5.3 Current practices for training bike skills in children with CP

Prior to researching the effectiveness of novel interventions, it is important to understand current practice (Craig et al., 2013). Knowledge of current practice helps to inform the comparison intervention, shapes the interpretation of findings and enhances the potential translation of novel interventions. In Australia, physiotherapists and occupational therapists often deliver training targeted towards motor goals of children with CP. However, prior to this thesis, the current practices of these disciplines when training bike skills in children with CP, were not well understood – no published studies existed documenting current bike skills training practices for children with CP, nor any other disabilities, in an Australian-context nor any other settings globally. An examination of current practice for training bike skills is required to inform components of the design of an effectiveness study, provide a framework for interpreting the findings and address the lack of evidence for training bike-specific skills in children with CP.

## 2.6. Two-wheel bike skills training in other developmental disabilities

Given the dearth of studies in training bike skills specific to CP, reviewing the literature from a broader childhood disability population may provide insight. A small number of low-quality studies exist for training two-wheel bike riding skills in other developmental disorders including young people with Trisomy 21 and autism spectrum disorder (ASD) (Halayko et al., 2016; MacDonald et al., 2010; Mishin, Hunt, Decker, Coley, & Nelson,

2015; Ulrich et al., 2011). The intervention approaches described in these studies are generally task-specific, involving training on a two-wheel bike with (Halayko et al., 2016) or without (MacDonald et al., 2010) a cognitive focus, or task-oriented whereby training progresses from a modified bike to a two-wheel bike (Ulrich et al., 2011). While training setting was variable, total dose was similar, involving 6 - 8 hours with two studies taking a high-density approach delivering the training over five consecutive days (MacDonald et al., 2010; Ulrich et al., 2011). Positive findings of these studies include learning to ride a bike and improved longer-term physical activity behaviour, although overall case series design and high risk of bias across means these results should be interpreted with caution. Further, it is important to consider that children with CP who experience problems primarily related to movement may face different challenges when learning new complex motor skills compared to those populations where intellectual impairment is a main feature.

## 2.7. Motor skills training in CP

In the absence of evidence specific to training bike skills in children with CP, broadening the focus of reviewing the literature around motor training in CP is necessary. A comprehensive systematic review of interventions in this population found strong evidence for improved motor activities following goal-directed training, bimanual training and constraint-induced movement therapy (Novak et al., 2013). Each of these approaches involve a common factor – task-specific training.

### 2.7.1 Task-specific training

Task-specific training refers to the active and repetitive practice of context-specific tasks (Bayona, Bitensky, Salter, & Teasell, 2005). It is informed by principles of motor learning and is underpinned by the dynamic systems theory which proposes that achievement of a motor skill in a task-specific context involves a dynamic interaction between systems including the task, the child and the environment (Hubbard et al., 2009; Thelen & Smith, 2007). Ideally, TST should be directed by the goals of the individual, focus on whole-task practice and take place in contexts that promote transfer to the individual's everyday life.

Two recent systematic reviews of training involving task-specificity in children with CP reported modest to strong effects on UL function including fine motor skills (Sakzewski, Gordon, et al., 2014; Sakzewski, Ziviani, et al., 2014). However, bike riding involves all four limbs making it a gross motor task. The available evidence for task-specific gross motor skill training in ambulant school-aged children with CP includes eight RCTs reporting positive outcomes for overall gross motor function (Bar-Haim et al., 2010; Grecco, Zanon, Sampaio, & Oliveira, 2013; Ketelaar, Vermeer, Hart, van Petegem-van Beek, & Helders, 2001; Swe, Sendhilnnathan, van Den Berg, & Barr, 2015), skill performance (Declerck, Verhuel, Daly, & Sanders, 2016; Hemayattalab, Arabameri, Pourazar, Ardakani, & Kashefi, 2013; Hemayattalab & Rostami, 2010), broader functional skills including self-care and mobility (Bleyenheuft, Arnould, Brandao, Bleyenheuft, & Gordon, 2015; Grecco et al., 2013; Ketelaar et al., 2001; Swe et al., 2015) and participation-related outcomes (Bleyenheuft et al., 2015). However, prior to this thesis, there had been no comprehensive evaluation of the evidence regarding this approach to training gross motor skills for children with CP. A systematic review of this literature is needed to examine the quality of this evidence, synthesise results and potentially identify active ingredients within the TST. Moreover, this type of study will inform the development of a task-specific approach to training bike skills.

### 2.7.2 Pilot work testing task-specific bike skills training in CP

I have evaluated a pilot intensive task-specific group-based bike riding training program led by physiotherapists at a metropolitan paediatric rehabilitation centre in Victoria, Australia through a small case-series. This study has been published as a conference abstract (Toovey, Reid, Harvey, Rawicki, & Watt, 2016). Each of the five children with CP (GMFCS I-II) who participated demonstrated a clinically meaningful improvement in goal performance and satisfaction following the training. While these findings are promising, high quality studies are required to test the effectiveness of this approach in this population.

## 2.8. Study design considerations

This chapter has identified that further research on task-specific gross motor skills training and current practices of therapists when training bike skills is required to inform the design

of the interventions within an effectiveness study. However, if the effectiveness of a task-specific approach for training bike skills in children with CP is to be tested, it is also important to consider which outcomes are measured and how.

### 2.8.1 Measuring a meaningful primary outcome

The skill of riding a two-wheel bike inherently sits within the activity domain of the ICF; however, arguably the ultimate goal of training bike skills is participation in community-based bike riding. Thus, in measuring effectiveness it is important to be able to capture both activity and participation constructs.

Whilst skill acquisition is a key activity-level outcome for determining the effect of training, there is no measure of bike-specific skills that has been validated in children with CP. Further, measures of participation that are validated in CP tend to have a broad focus and may not be responsive to changes in one specific activity. Moreover, the heterogeneity of the CP population in terms of motor type, topography and functional abilities limits the specificity and responsiveness to change in many generic standardised outcome measures (Vargus-Adams & Martin, 2009).

Therapy tailored to the goals of the family and child has become standard in the delivery of paediatric rehabilitation (Oien, Fallang, & Ostensjo, 2010). Measuring children's progress towards individualised goals is increasingly important and allows for both activity and participation goals to be set. Moreover, within an effectiveness study that aims to determine the effects of training towards bike riding goals, it is logical to measure attainment of individualised bike-specific goals as the primary outcome.

The Goal Attainment Scale (GAS) and Canadian Occupational Performance Measure (COPM) are the most common tools in the CP literature used to capture the goal attainment construct (Cusick, McIntyre, Novak, Lannin, & Lowe, 2006). The COPM is a measure that involves a conversation between the clinician, the child and family to identify issues within each domain of self-care, productivity or leisure, define their importance and detect changes in self-perception and satisfaction over time (Law et al., 1990). The GAS is a criterion-referenced tool for individualised and collaborative goal setting between the child,

family and clinician (Kiresuk & Sherman, 1968; Steenbeek, Ketelaar, Galama, & Gorter, 2007). The GAS is generally a five to seven-point scale from -3/-2 to +2/+3 with a score of zero representing an expected level of attainment, a negative score indicating less than expected progress and a positive score, more than expected progress. Both measures have been validated for use in interventions involving children with CP (Cusick et al., 2006; Palisano, 1993; Steenbeek, Ketelaar, Lindeman, Galama, & Gorter, 2010), however, the GAS offers more adaptability for setting and measuring goals related to specific skills, whereas there is little evidence to support use of a modified, more specific, version of the COPM. Overall, valid measurement of the attainment of individualised bike-specific goals can be achieved using the GAS.

### 2.8.2 Selection of secondary outcomes

Measuring secondary outcomes may capture additional benefits of training bike skills, aside from goal attainment. As previously acknowledged, bike riding primarily relates to the activity and participation domains of the ICF. While activity can be measured relatively simply through assessing the child's competence to perform a task at varying levels of ability, in similar, and different environments, participation has proven harder to define in the literature around children with disabilities.

A recent systematic review provided guidance in this area by proposing that participation consists of two key elements: attendance (being there) and involvement (the experience of being there) (Imms, Adair, et al., 2016a). Further to this review, a Family of Participation-Related constructs model has been proposed (Imms, Granlund, et al., 2016). As shown in Figure 2.2, this model proposes that there is a dynamic relationship between an individual's activity competence, sense of self, and preferences, and the key elements within participation (Figure 2a). This family of constructs then sits within the environment and broader context and again a dynamic relationship exists between the individual and these external factors (Figure 2b).

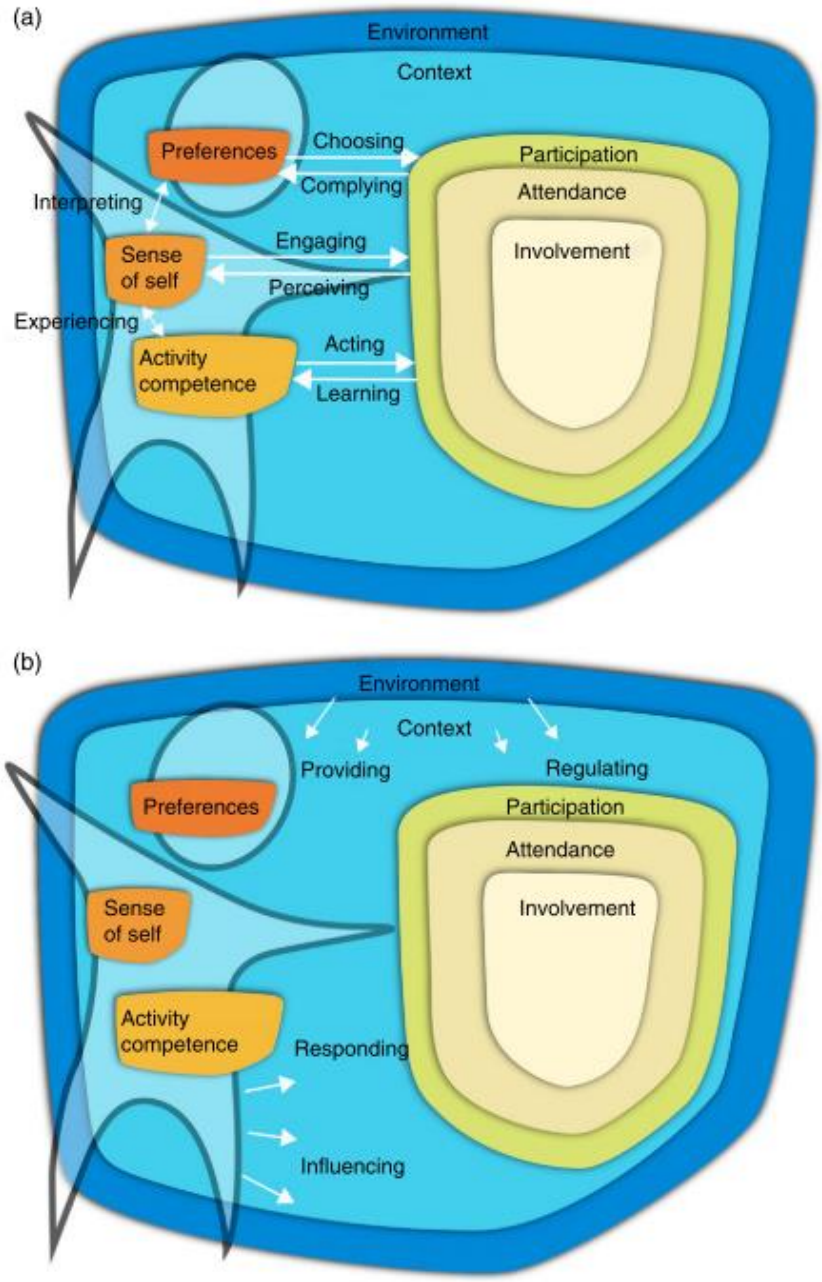



**Figure 2.2:** Family of Participation-Related Constructs: (a) internal factors, (b) external factors (Imms, Granlund, et al., 2016). Reproduced with permission from John Wiley & Sons (Appendix I)

This model can inform the selection of secondary outcomes of training bike skills to capture both activity *and* the participation construct. For example, important outcomes such as bike skills and self-perception, which map to the activity competence and sense of self constructs respectively, could be measured alongside participation in bike riding to identify any changes in these intrinsic factors that influence, and are influenced by, participation (Imms, Granlund, et al., 2016; Shields, Loy, Murdoch, Taylor, & Dodd, 2007).

## 2.9. Conclusions

Overall, this chapter found that minimal research evidence exists for training two-wheel bike skills in children with CP. However, reviewing the literature on bike riding ability in CP, bike skills training in other populations and motor skills training more broadly in CP, was useful in identifying gaps for future research and a potential intervention approach. This review indicated that investigating the effectiveness of a TST approach to training bike skills in children with CP is warranted. Yet, it is necessary to first evaluate the evidence for task-specific gross motor skills training and understand current practices of clinicians when working with children with CP who have bike riding goals. A systematic review of task-specific gross motor skills training and survey of current bike skills training practices in this population will inform intervention design within an effectiveness study and provide greater context for interpreting findings. Further, using the ICF as a framework within each study would enable common terminology and synthesis of results across the PhD. This chapter also identified that the GAS may be a useful tool for measuring effectiveness in that it has good psychometric properties for use in children with CP and can capture individualised bike-specific goals. Moreover, given the dearth of research in bike skills training in this population, the three studies proposed in this thesis would contribute significantly to the knowledge base in this area.

# CHAPTER 3: TASK-SPECIFIC GROSS MOTOR SKILLS TRAINING FOR AMBULANT SCHOOL-AGED CHILDREN WITH CEREBRAL PALSY: A SYSTEMATIC REVIEW

## 3.1. Preamble

Based on the paucity of evidence specific to task-specific bike skills training for ambulant children with CP identified in the literature review (Chapter 2), a systematic review of the literature regarding TST of gross motor skills training in this population was undertaken. The knowledge gained from this review informs the design of the experimental group in the RCT, the major study in this thesis (Chapters 5 and 6).

This chapter contains the manuscript of this systematic review reprinted from *BMJ Paediatrics Open.* Details of the author contributions and funding sources are listed in the preface to the thesis. The manuscript is reported using Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines and Vancouver style referencing as required by the journal, with references at the end of the chapter. As an example of the search strategy used in this study, the search strategy for Ovid databases is included as in section 3.2 as a supplementary appendix following the manuscript. The Grading of Recommendations Assessment, Development and Evaluation (GRADE) table is also included as a supplementary appendix (Table 4) in section 3.3. Co-author authorisation forms are included at the end of this thesis (Appendix II). As the author of this article, no third-party copyright permission is required (Appendix III).

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BMJ Paediatrics Open

# Task-specific gross motor skills training for ambulant school-aged children with cerebral palsy: a systematic review

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## ABSTRACT

**Objectives** The primary objective is to systematically evaluate the evidence for the effectiveness of task-specific training (TST) of gross motor skills for improving activity and/or participation outcomes in ambulant school-aged children with cerebral palsy (CP). The secondary objective is to identify motor learning strategies reported within TST and assess relationship to outcome.

**Design** Systematic review.

**Method** Relevant databases were searched for studies including: children with CP (mean age >4 years and >60% of the sample ambulant); TST targeting gross motor skills and activity (skill performance, gross motor function and functional skills) and/or participation-related outcomes. Quality of included studies was assessed using standardised tools for risk of bias, study design and quality of evidence across outcomes. Continuous data were summarised for each study using standardised mean difference (SMD) and 95% CIs.

**Results** Thirteen studies met inclusion criteria: eight randomised controlled trials (RCTs), three comparative studies, one repeated-measures study and one single-subject design study. Risk of bias was moderate across studies. Components of TST varied and were often poorly reported. Within-group effects of TST were positive across all outcomes of interest in 11 studies. In RCTs, between-group effects were conflicting for skill performance and functional skills, positive for participation-related outcomes (one study: Life-HABITS performance SMD=1.19, 95% CI 0.3 to 2.07, p<0.001; Life-HABITS satisfaction SMD=1.29, 95% CI 0.40 to 2.18, p=0.001), while no difference or negative effects were found for gross motor function. The quality of evidence was low-to-moderate overall. Variability and poor reporting of motor learning strategies limited assessment of relationship to outcome.

**Conclusions** Limited evidence for TST for gross motor skills in ambulant children with CP exists for improving activity and participation-related outcomes and recommendations for use over other interventions are limited by poor study methodology and heterogeneous interventions.

**Registration** PROSPERO ID42016036727

### What is already known on this topic?

- Strong evidence for motor interventions involving task specificity for functional mobility in adults poststroke and for upper limb function in children with cerebral palsy exists.
- The effectiveness of task-specific gross motor skills training in ambulant school-aged children with cerebral palsy has not been systematically evaluated or synthesised.

### What this study hopes to add?

- A low-to-moderate overall quality of evidence was found for task-specific gross motor skills training for ambulant school-aged children with cerebral palsy.
- Limited evidence for task-specific training to improve specific skills performance, functional skills and participation-related outcomes exists.
- While clear recommendations for use of task-specific training over other interventions are limited, ways to strengthen the evidence in future studies are identified.

## INTRODUCTION

Cerebral palsy (CP) is an umbrella term used to describe a group of disorders of movement which cause varying degrees of activity limitations.[1] The most widely used means for classifying gross motor function in children with CP is the Gross Motor Function Classification System (GMFCS).[2] Children classified GMFCS I are able to walk and run but have limitations with speed, balance and coordination while children classified GMFCS V are transported in a wheelchair in all settings. Although the focus of the GMFCS is on functional mobility, the realm of gross motor activities, that is skills involving movement of the large muscles of the limbs or whole body, undertaken by children is much broader.[3] Development of gross motor skills underpins functional, play and social activities across childhood and complex movement skills required for sports in older children.[3] In children with CP, limitations in gross motor function increase as GMFCS level increases, however, children at all GMFCS levels (I–V)

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participate, on average, less in physical activities than their typically developing peers.[2] This is an issue because of the known poor health outcomes in adulthood due to inactivity in childhood.[4] Effective interventions tailored to GMFCS levels and developmental stages are required to improve these outcomes in this population.

The International Classification of Functioning, Disability and Health (ICF) has become the common language for clinicians, researchers and families for understanding the effect of CP on the individual and for targeting interventions.[5 6] Where interventions previously focused on remediating limitations in body structures and functions, there has been a more recent acknowledgement of the importance of the effect of interventions within the activity and participation domains.[7 8] Clinicians working with children with CP need guidance from evidence synthesis to implement effective means of improving physical skills *and* improve the uptake of these skills in the child's daily life. The historical bias towards impairment-focused motor interventions yielded few effective treatments,[6 9 10] thus more functional approaches have emerged.

Task-specific training (TST) involves practice of context-specific tasks where the intervention focuses on the skills needed for a task(s)[11]—there is similarity between the training task and the goal of the intervention. Although level I evidence exists for TST to improve gross motor activities in adults after stroke,[12] the majority of high-level evidence for interventions involving task specificity in children with CP relates to training of upper limb or fine motor activities[6 13] with limited evidence for gross motor skills training. TST inherently involves principles of motor learning with components including context, practice and dosage.[11] Other motor learning strategies, such as feedback and task modification, have the potential to optimise TST, however, this has not been systematically studied.[14] TST should involve varied components depending on the requirements of the skill, the environment and the function of the child.[15] Moreover, training for a child of higher-level motor function (eg, GMFCS I–III) should be targeted towards different skills compared with training with a child of lower-level motor function (eg, GMFCS IV–V). Similarly, children of different ages and developmental stages have varying learning capabilities, and physical demands placed on them by their context.[16]

Previous systematic reviews of motor interventions in children with CP have been broad in terms of ages (including infants and children) and motor function (all GMFCS levels), and included interventions have been heterogeneous.[17 18] TST may be a promising approach for ambulant children who have specific gross motor skills goals, however, there has been no systematic review to examine the effectiveness of this approach in this population. The primary aim of this study is to evaluate and synthesise the evidence for the effectiveness of task-specific gross motor skills training in ambulant children aged 4–18 years with CP for activity and participation outcomes. The secondary aim of this study is to identify motor learning strategies reported within TST and assess relationship to outcome.

## METHOD

### Eligibility criteria

Published studies were included if they met all of the following criteria:

1. Level of evidence: all group design studies categorised as level II–IV using the American Academy of Cerebral Palsy and Developmental Medicine (AACPDM) levels of evidence[19] and studies classified as level I single-subject designs involving over 10 participants.
2. Population: the majority (ie, >60%) of the participants were ambulant children with CP (GMFCS I–III), and the mean sample age was 4–18 years.
3. Interventions: TST of gross motor skills where there was similarity between the training task and the goal of the intervention, including those interventions described as involving motor learning strategies/coaching, goal-directed training, activity focused training and/or functional skills training. Any duration or intensity of TST.
4. Comparison: studies comparing TST with another intervention, another type of TST or no intervention.
5. Outcomes: activity outcomes including gross motor skill performance (specific to the task being trained or other gross motor task to assess for transferability), gross motor function and functional skills; and participation-related outcomes. Only studies reporting outcomes separately for children with CP.

Exclusion criteria: TST was applied within a combined intervention approach and the influence of TST could not be isolated (eg, botulinum toxin-A, virtual reality, treadmill training, orthoses or robotics), >10% of the intervention was passive or the article was not in English.

### Search strategy

Relevant articles were identified from Medline, EMBASE, CINAHL, PsycINFO, SPORTDiscus and PubMed with all searches limited to articles published in peer-reviewed journals in English. A search was conducted in June 2016 and search terms were tailored for each database. Reference lists of included studies and related reviews were also searched. See online supplementary appendix 1 for full search strategy for Ovid databases (Medline, EMBASE and PubMed).

### Study selection, data extraction and quality assessment

Study selection, data extraction and quality assessment was completed by two authors (RT and CB) independently, with a third author (AH, JM or AS) to resolve any disagreements. If inclusion was uncertain from abstract, the full-text was retrieved.

For included studies, data were extracted using a customised form based on the Cochrane recommendations.[20] Data extracted included: study details (author,

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year, country, funding), study design, AACPDM level of evidence,[19] study sample characteristics, detailed TST characteristics (including motor learning strategies), comparison intervention characteristics, outcomes measured and associated ICF domain, effects of the intervention and conclusions. Reported motor learning strategies in all TST, including when studies involved a comparison of two or more TST approaches, were identified using predefined motor learning strategy codes based on current literature.[21 22] The authors were contacted for complete data extraction when needed.

Risk of bias for individual studies was assessed using the Cochrane Risk of Bias Tool.[20] Overall risk of bias was determined by predominant level of bias (at least four of the seven domains) across the domains of the Risk of Bias Tool. Intervention replicability was assessed using relevant components of the Template for Intervention Descriptions and Replication (TIDieR) Checklist.[23] The Grading of Recommendations Assessment Development and Evaluation (GRADE) system was used to assess the quality of the evidence across outcomes.[24]

### Narrative synthesis and data analysis

Study design and AACPDM level of evidence were used for narrative synthesis. Higher-level group designs (AACPDM levels II–III) were reported together, while lower-level group designs (IV) and single-subject design studies were reported separately. Within the level II–III group designs, studies were considered as either 'TST versus a comparison' or 'TST versus TST'. Finally, within each grouping, activity outcomes were organised into three constructs: gross motor skill performance, functional skills and gross motor function while participation-related outcomes were grouped together. Gross motor skill performance outcomes included those measuring a specific gross motor skill (eg, walking or throwing), functional skills outcomes included those measuring a broader range of skills in domains related to function (eg, self-care or social skills), while gross motor function outcomes included those measuring a range of gross motor-related skills.

Analyses were conducted using Cochrane Review Manager Software RevMan V.5.3. Only data from level II studies comparing TST with comparison interventions were included in quantitative analysis to ensure comparison was between studies of similar design. Continuous data were summarised for each study within outcomes of interest using standardised mean difference (SMD) and 95% CIs. An SMD of 0.2 was considered small, 0.4–0.6 moderate and 0.8 a large effect size.[20] Given the heterogeneity between studies in tasks and characteristics of the TST and comparison interventions, meta-analysis was not undertaken.

## RESULTS

Following removal of duplicates, the abstracts of 1247 studies were screened and 145 full-text articles were retrieved for full appraisal (figure 1). Thirteen studies involving 405 participants met inclusion criteria and underwent narrative synthesis with six of these studies involving 237 individuals included in data analysis. Characteristics of included studies are summarised in table 1.

The 13 studies included 12 group designs and one single-subject design (level I). The group designs involved eight randomised controlled trials (RCTs) (level II), three comparative studies with concurrent controls (level III) and one repeated measures study (level IV). Eleven studies did not report adverse events as an outcome and two studies reported no adverse events.[25 26]

### Risk of bias assessment

Risk of bias was low in four studies,[14 27–30] unclear in seven studies[14 25 26 31–34] and high in two studies[34–36] leading to an overall moderate risk of bias across the studies (table 2). Major sources of bias included: studies without

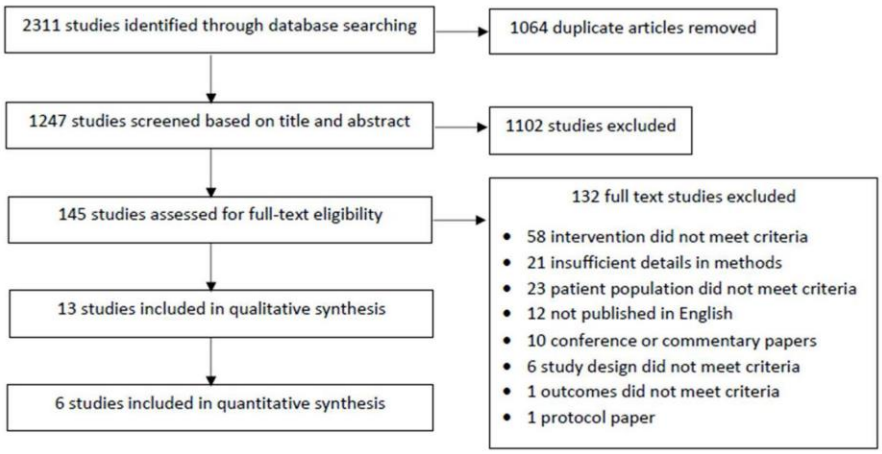



**Figure 1** Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow chart of included and excluded studies.

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Table 1 Characteristics of included studies

| Study | Study design | AACPDM LoE | CP n | Age range, mean and SD in years | GMFCS levels/ proportions | Task-specific training (T) | Comparison (C) or other task-specific training | Activity outcomes: Gross motor function — Outcome measured used | Activity outcomes: Functional skills — Outcome measured used | Activity outcomes: Gross motor skill performance — Outcome measured used | Participation related outcomes (measure) | Other outcomes (ICF domain) |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| **1. Group research designs** | | | | | | | | | | | | |
| **a. Level I–III studies (task-specific vs comparison)** | | | | | | | | | | | | |
| Bar-Haim *et al*[27] | RCT | II | Total=78<br>T=39<br>C=39 | Range=5.5–12.2<br>Total=8.9± 1.7<br>T=8.8 ± 1.7<br>C=8.9 ± 1.7 | T<br>II=36%, III=64%<br>C<br>II=44% III=56% | Motor learning coaching | Routine physiotherapy—NDT | GMFM | Parent survey | | | Mechanical efficiency (BSF) |
| Bleyenheuft *et al*[25] | Crossover RCT | II | Total=24*<br>T=12<br>C=12 | T=8.9± 1.7 C=8.5± 1.7 | T<br>I=50% II=50%<br>C<br>I=42% II=58% | Hand arm bimanual intensive training including lower extremity (HABIT-ILE) | Routine physiotherapy (in general had NDT while waiting for delayed HABIT-ILE) | | PEDI (self-care domain only) | 6MWT<br>ABILOCO Kids | Social participation (Life-H Performan-ce and Satisfaction) | Body weight distribution (BSF)<br>Mean step length (BSF)<br>UL function (Act)<br>Adverse events |
| Declerck *et al*[26] | RCT | II | Total=14<br>T=7<br>C=7 | Range=7–17<br>T=8.7± 3.4<br>C=11.8± 3.5 | T<br>I=14% II=86%<br>C<br>I=29% II=57% III=14% | Swimming skills programme | Routine physiotherapy | | | WOTA-2†<br>1MWT | Adherence/ enjoyment | Pain (BSF)<br>Fatigue (BSF)<br>Adverse events |
| Grecco *et al*[28] | RCT | II | Total=36*<br>T=17<br>C=18 | T=6.8± 2.6<br>C=6.0± 1.5 | T<br>I=47% II=41% III=12%<br>C<br>I=31% II=50% GMFCS III=19% | Overground walking | Treadmill training | GMFM | PEDI | 6MWT†<br>TUG | | Balance (Act) |
| Ketelaar *et al*[33] | RCT | II | Total=55<br>T=28<br>C=27 | Range (total)=2.0–7.25<br>T=4.5± 1.7<br>C=4.7± 1.7 | T mild=79%, mod=21%<br>C mild=77%, mod=23% | Functional therapy | Routine physiotherapy—generally NDT or Vojta method | GMFM | PEDI | | | |
| Kumban *et al*[29] | RCT | II | Total=21<br>T=10<br>C=11 | T=12.3± 2.6<br>C=12.4± 2.4 | T<br>I–II=60%, III=40%<br>C<br>I–II=55% III=45% | Regular PT + sit-to-stand task training | Routine physiotherapy | | | FTSST† | | Balance ability (Act)<br>Functional balance and lower limb strength (Act) |
| Swe *et al*[30] | RCT | II | Total=30*<br>T=15<br>C=15 | Total=13.2 +/- 3.4<br>T=13.0 +/- 3.6<br>C=13.4 +/- 3.3 | T<br>II=53% III=47%<br>C<br>II=67%, III=33% | Overground walking | Partial weight support treadmill training | GMFM (standing and walking domains only) | | 6MWT†<br>10MWT† | | |
| Capio *et al*[31] | Comparative study with concurrent controls | III | Total=24<br>T=12<br>C=12 | T=6.92 +/- 3.04<br>C=7.98 +/- 1.74 | T<br>I=33% II=58% III=8%<br>C<br>I=25% II=58% III=17% | Functional motor skills training | Routine physiotherapy or physical education | TGMD- 2 | | | Physical activity (accelerome-ter) | |
| **b. Level II–III (task-specific vs task-specific)** | | | | | | | | | | | | |
| Hemayattalab *et al*[32] | RCT | II | Total=20<br>T1=10<br>T2=10 | Total=11.6 +/- 1.5<br>T1=11.9 +/- 1.6<br>T2=11.3 +/- 1.4 | I–III<br>Proportions not stated | Throwing task + self-control feedback | Throwing task + yoked feedback | | | Throwing accuracy† | | |
| Hemayattalab and Rostami[34] | Comparative study with concurrent controls | III | Total=24<br>T1=8<br>T2=8<br>T3=8 | Range (total)=7–15 | I=100% | T1 = dart throwing task+50% KR<br>T2 = dart throwing task+100% KR | T3=dart throwing task+0% KR | | | Throwing accuracy† | | |
| Löwing *et al*[35] | Comparative study with concurrent controls | III | Total=44<br>T=22<br>C=22 | Total=4.1 +/- 1.4<br>T=4.3 +/- 0.7<br>C=3.8 +/- 1.3 | T<br>I=45% II=23%, III=14% IV=18%<br>C<br>I=41% II=23%, III=22% IV=14% | Activity focused individual therapy | Goal-directed functional group therapy | GMFM | PEDI | | Goal attainment (GAS) in GDT group only | |
| **c. Level IV** | | | | | | | | | | | | |
| Sorsdahl *et al*[36] | Repeated measures design | IV | T=22* | Range=2.8–9.25<br>Average age=5.5 | T<br>I=36% II=23%, III=27% IV–V=14% | Functional family-centred goal directed therapy | | GMFM<br>GMPM | | | Goal attainment (GAS) | Quality of fine motor movements (Act) |

Continued

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Table 1 Continued

| Study | Study design | AACPDM LoE | CP n | Age range, mean and SD in years | GMFCS levels/ proportions | Task-specific training (T) | Comparison (C) or other task-specific training | Activity outcomes: Outcome measured used: Gross motor function | Activity outcomes: Outcome measured used: Functional skills | Activity outcomes: Outcome measured used: Gross motor skill performance | Participation related outcomes (measure) | Other outcomes (ICF domain) |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| **2. Single-subject research designs** | | | | | | | | | | | | |
| Thorpe and Valvano[44] | 13 x n=1 studies with random sampling to protocols (A, B or C) | I | N(total)=13 | Range=6.0–12.7 Age=8.6 +/- 1.89 | III=100% | All protocols = task specific<br>A=2x no AI 1 x KP and 5 x KP+CS | B=3x no AI, 2 x KP and 3 x KP+CS<br>C=4 x no AI, 3 x KP and 1 x KP+CS | | | Change in backward displacement† | | |

*Sample size justified with statistical consideration.
†Skill performance outcome is specific to skill being trained.
ABILOCO, measure of locomotion ability; Act, activity; AI, augmented information; BSF; body structures and functions; C, comparison; CS, cognitive strategies; GAS; Goal Attainment Scale, GMPM, Gross Motor Performance Measure; ICF, International Classification of Functioning, Disability and Health; GMFCS, Gross Motor Function Classification System; GMFM, Gross Motor Function Measure (-88 or -66); KP, knowledge of performance; KR, knowledge of results; LoE, level of evidence; Life-H, assessment of life habits; NDT, neurodevelopmental therapy; PEDI, Paediatric Evaluation of Disability Inventory; T, task-specific intervention; TGMD-2, Test of Gross Motor Development -2nd edition; WOTA-2, Water Orientation Test Alyn-2nd edition; 1MWT, 1 min walk test, 10MWT, 10 metre walk test; 6MWT, 6 min walk test.

randomisation or allocation concealment, a lack of participant/personnel blinding and incomplete outcome data. No study reported the use of intention-to-treat analysis and reporting of outcome assessor blinding was inconsistent. Poor reporting of study conduct limited assessment of risk of bias in the seven studies with unclear risk of bias.

## Level II–III group designs

### Task-specific training versus comparison

#### *Interventions*

Seven RCTs and one comparative study[31] (table 1) compared TST with another intervention. The target of TST varied; four studies involved training a variety of tasks or movement skills,[21 25 31 33] one focused on swimming[26] and another trained sit to stand.[29] Six studies compared TST with 'routine physiotherapy'.[25–27 29 31 33] This was generally poorly defined; described as neurodevelopmental therapy (NDT)[25 27 33] in three studies, physical education in one study[31] or not reported.[26 29] The remaining two studies involved overground walking compared with treadmill training; with overground walking deemed as the TST, as treadmill training was considered not specific to the gross motor aim of improving overground walking.

Two of the eight studies were goal-directed while four studies reported the TST was driven by motor learning principles[25 27 31 33] (table 3). Repetitive practice was the most commonly reported motor learning strategy with feedback and task modification in four[25 27 28 33] and five[25 27 29 31 33] studies, respectively. Physiotherapists generally conducted interventions but characteristics varied widely. Overall intervention time ranged from 3[31] to 90 hours[25] over periods from 10 days[25] to 6 months.[33] Reported settings were largely ecological while format was group-based in two studies,[25 26] otherwise was not stated.

#### *Outcomes and effects*

All eight studies reported outcomes at the activity level, three reported participation-related outcomes and three involved body structure and function outcomes (table 1). Outcomes were measured at one to three time points. In general, T1 was immediately following the intervention, while T2 (4 weeks–6 months)[25–30 33] and T3 (2–12 months)[26 33] were varied.

#### *Activity outcomes*

Gross motor skill performance was measured in five of the eight studies[25 26 28–30] with four measuring skill performance specific to the training tasks[26 28–30] (table 1). Overall, skills and measures were varied. Four studies measured walking performance[25 26 28 30]: using the 6 min walk test (6MWT)[25 28 30] or the timed up and go, 1 min walk test, 10 metre walk test and the ABILOCO Kids questionnaire.[25 26 28 30] Between-group effects of TST on walking performance were mixed (figure 2A-B). Large effects favouring TST on walking performance at T1 were

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Table 2 Risk of bias within included studies

| | Study | Selection bias | | Performance bias | Detection bias | Attrition bias | Reporting bias | Other bias | Overall bias |
|---|---|---|---|---|---|---|---|---|---|
| | | Random sequence generation | Allocation concealment | Participant and personnel blinding | Outcome assessor blinding | Incomplete outcome data | Selective outcome reporting | | |
| **Group research designs** | | | | | | | | | |
| Level II and III studies | Bar-Haim *et al*[27] | Low | Low | High | Low | High | Low | High | Low |
| | Bleyenheuft *et al*[25] | Low | Low | High | Unclear | High | Unclear | Unclear | Unclear |
| | Declerck *et al*[26] | Low | Low | High | Unclear | Unclear | Low | High | Unclear |
| | Grecco *et al*[28] | Low | Low | High | Low | Unclear | Low | Unclear | Low |
| | Ketelaar *et al*[33] | Low | Unclear | High | Low | Unclear | Unclear | Unclear | Unclear |
| | Kumban *et al*[33] | Low | Low | High | Low | High | Low | Unclear | Low |
| | Swe *et al*[30] | Low | Low | High | Low | Low | Low | Unclear | Low |
| | Capio *et al*[31] | High | High | High | Unclear | Unclear | Unclear | Unclear | Unclear |
| | Hemayattalab *et al*[32] | High | Unclear | High | Unclear | Unclear | Unclear | Unclear | Unclear |
| | Hemayattalab and Rostami[34] | Unclear | High | High | Unclear | Low | Low | Unclear | Unclear |
| | Löwing *et al*[35] | High | High | High | High | Low | Low | High | High |
| IV | Sorsdahl *et al*[36] | High | High | High | Low | High | Low | Unclear | High |
| **Single-subject design** | | | | | | | | | |
| I | Thorpe and Valvano[14] | Low | Unclear | High | High | Low | Low | Unclear | Unclear |

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**Table 3** Interventions: components of task-specific training for included studies

| Study | Task-specific intervention/s | Key characteristics of task-specific training | | | | | | | | | Components of the TIDIER checklist | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | | Overall approach | | Motor learning strategies reported | | | | | | | | | | | Adherence | Fidelity |
| | | Goal-directed | Motor learning driven | Feedback | Cogn. strat. | Repet. practice | Task mod. | Environ. mod | Obs. learning | Dual task learning | Dosage (total hours) | Setting | Format | Providers | Reported? | |
| **Group designs** | | | | | | | | | | | | | | | | |
| AACPDM level II and III task-specific versus comparison | Bar-Haim *et al*[27] | Motor learning coaching | ✓ | ✓ | ✓ | | ✓ | ✓ | ✓ | ✓ | 1 hour, 3 days/week for 3 months (36 hours) | School | NS | Trained PT | | |
| | Bleyenheuft *et al*[25] | HABIT-ILE | | ✓ | ✓ | | ✓ | ✓ | ✓ | ✓ | 9 hours/day for 10 consecutive days (90 hours) | Recreation camp | Group | Trained PTs, OTs, and PT/OT students | ✓ | |
| | Declerck *et al*[26] | Swimming skills programme | | | | | ✓ | | | | 40–50 mins, 2 days/week for 10 weeks (15 hours) | Swimming pool | Group | PT and PT students | ✓ | |
| | Grecco *et al*[28] | Overground walking | | | ✓ | | ✓ | ✓ | | | 30 mins, 2 days/week for 7 weeks (7 hours) | NS | NS | Therapist | | |
| | Ketelaar *et al*[33] | Functional therapy | ✓ | ✓ | ✓ | | ✓ | ✓ | ✓ | | 6 months – intensity or hours NS (NS) | Clinic and home | NS | Trained PTs and parents | | |
| | Kumban *et al*[29] | Sit-to-stand task training | | | | | ✓ | ✓ | | | 20 min, 3 days/week for 6 weeks (6 hours) | School | NS | Child's own PT | | |
| | Swe *et al*[30] | Overground walking | | | | | ✓ | ✓ | | | 30 mins, 2 days/week for 8 weeks (8 hours) | School | NS | PTs | ✓ | |
| | Capio *et al*[31] | Functional motor skills training | | ✓ | | | ✓ | ✓ | | | 45 mins, 1 day/week for 4 weeks (3 hours) | NS | NS | Each child's regular PT | ✓ | |
| Level I and III task-specific vs task-specific | Hemayattalab *et al*[32] | Throwing task with different feedback conditions | | ✓ | ✓ | | ✓ | | ✓ | | Two sessions in 2 days (NS) | NS | NS | NS | | |
| | Hemayattalab *and Rostami*[34] | Throwing task with different feedback conditions | | ✓ | ✓ | | ✓ | | ✓ | | Eight sessions Intensity or hours NS (NS) | NS | NS | Specialist trainers | | |
| | Löwing *et al*[35] | Activity focused individual therapy vs goal-directed therapy | ✓ (goal group only) | | | | ✓ inc. home programme | | | | Three sessions/ fortnight for 12 weeks (NS) | Clinic and home/ preschool | Group and individual | PT, OT, SP, teacher and parents | | |
| Level IV | Sorsdahl *et al*[36] | Functional family centred goal-directed therapy | ✓ | | ✓ | | ✓ inc. home programme | ✓ | ✓ | | 3 hours, 5 days/week for 3 weeks (45 hours) | Clinic | Group | Trained PTs and parents | ✓ | |
| **Single-subject design** | | | | | | | | | | | | | | | | |
| Level I | Thorpe and Valvano[14] | Novel motor task learning with different conditions | | ✓ | ✓ | ✓ | ✓ | | | | Three sessions over 5 days (NS) | School | Individual | PT (also the PI) | | |

Cogn, cognitive; environ, environmental; inc, including; NS, not stated; HABIT-ILE, Hand Arm Bimanual Intensive Training Including Lower Extremity; mod, modification; PI, principal investigator; Obs, observational; OT, occupational therapist; PT, physiotherapist; SP, speech therapist; Strat, strategies; TIDIER, Template for Intervention Descriptions and Replication; trained, trained in specific intervention protocol.

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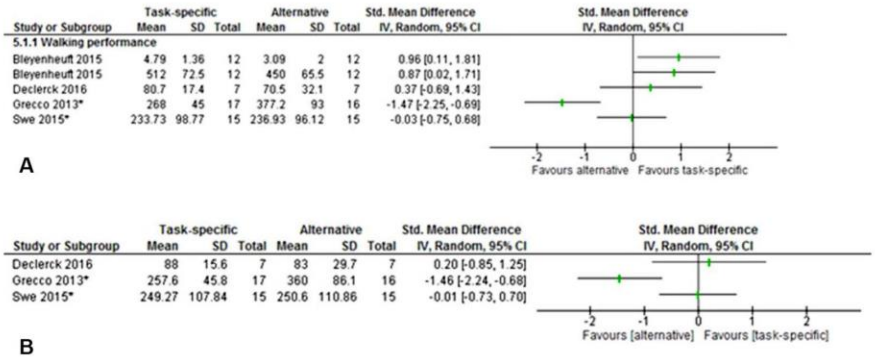



**Figure 2** (A) Gross motor skill performance—task specific vs alternative: walking performance at T1 (NB Grecco *et al* TUG scores not included). *Outcome is specific to task being trained. (B) Gross motor skill performance—task specific vs alternative: walking performance at T2 (NB Grecco *et al* TUG scores not included). *Outcome is specific to task being trained.

found (ABILOCO Kids SMD=0.96, 95% CI=0.11 to 1.81; 6MWT SMD=0.87, 95% CI 0.02 to 1.71) by Bleyenheuft *et al* (Hand Arm Bimanual Intensive Training Including Lower Extremity vs routine physiotherapy).[25] However, large negative effects at both T1 (6MWT SMD=−1.47, 95% CI −2.25 to −0.69) and at 4 weeks follow-up (6MWT SMD=−1.46, 95% CI −2.24 to −0.68) were found by Grecco *et al* (overground walking vs treadmill training).[28] Non-significant mixed effects were found at both time points in the remaining two studies.[26 30] Swimming performance was measured using the Water Orientation Test Alyn-2nd edition in one study[26] and sit-to-stand performance was measured using the five-times sit-to-stand test in another.[29] There was no between-group effect found for TST on sit-to-stand[29] or swimming performance at T1.[26] Within-group effects of TST were positive for all skill performance outcomes in each study, except for the GMFCS III subgroup in one study.[29]

Overall gross motor function was measured in five of the eight studies[27 28 30 31 33] (table 1). Four studies[27 28 30 33] used the Gross Motor Function Measure (GMFM).[37] Three studies measured all domains of the GMFM[27 28 33] (A: lying and rolling, B: sitting, C: crawling and kneeling, D: standing, E walking running and jumping), while one study measured domains D and E only.[30] Nil or negative effect of TST was found at T1 or at T2 (figure 3a-d)[27 28 30 33] on domains D and E. Three studies demonstrated no effect of TST at T1 or T2 in either domain,[27 30 33] while the fourth study (Grecco *et al*) showed a large negative effect for both domains at T1 (GMFM-D SMD=−1.39, 95% CI −2.16 to −0.62; GMFM-E SMD=−1.97, 95% CI −2.82 to −1.12) and T2 (GMFM-D SMD=−1.32, 95% CI −2.09 to −0.56; GMFM-E SMD=−2.08, 95% CI −2.95 to −1.22).[28] No significant effects of TST were found on total GMFM score (figure 3E-F)[27 33]. Again, a large effect favouring the comparison was found for total score at both time points (T1 SMD=−1.83, 95% CI −2.63 to −1.03; T2 SMD=−1.67, 95% CI −2.48 to −0.87) by Grecco *et al*.[28] The remaining level III design study reported a significant positive effect of TST on gross motor function as measured by the Test of Gross Motor Development-2nd edition.[31] Within-group effects of TST on gross motor function were positive in all five studies.

Functional skill outcomes were measured in four of the eight studies[25 27 28 33] (table 1). Functional skills included self-care, assessed by the self-care domain of the Paediatric Evaluation of Disability Inventory (PEDI), in three studies[25 28 33]; mobility, measured by the PEDI mobility domain in two studies[28 33] and a parent questionnaire[27]; and social function assessed by the PEDI social function domain in one study.[28] A large effect (SMD=1.07, 95% CI 0.21 to 1.94) favouring TST was found for self-care skills at T1 in the study by Bleyenheuft *et al*.[25] No significant effects were found for the other two studies measuring self-care at T1 and T228[28 33] (figure 4A-B). No effect of TST on mobility skills (PEDI mobility domain) was found at T1 (figure 4C), but a moderate effect was found at 6 months (T2 SMD=0.58, 95% CI 0.04 to 1.12) and a moderate-to-large effect found at 12-month follow-up (T3 SMD=0.76, 95% CI 0.21 to 1.31) by Ketelaar *et al* (functional therapy vs routine physiotherapy).[33] In contrast, a large negative effect (SMD=−1.32, 95% CI −2.09 to −0.56) was found for mobility skills at T1 by Grecco *et al*[28] (figure 4C). No effect on mobility on the parent questionnaire was found at T1 or 6 months post[27] or on social function T1 or at 4 weeks follow-up.[28] Within-group effects of TST on all functional skills outcomes were positive in all four studies.

### *Participation-related outcomes*

Participation-related outcomes were measured in only three of the eight studies[25 26 31] (table 1). Measures included social participation using the Life-HABITS performance and satisfaction questionnaire,[25] adherence and enjoyment[26] and participation in physical activity using an accelerometer.[31] Large effects favouring TST were found for both social participation performance (SMD=1.19, 95% CI 0.31 to 2.07) and satisfaction (SMD=1.29, 95% CI 0.40 to 2.18) by Bleyenheuft *et al*.[25] Positive effects of TST on weekend physical activity were reported at T1.[31] High adherence rates and enjoyment

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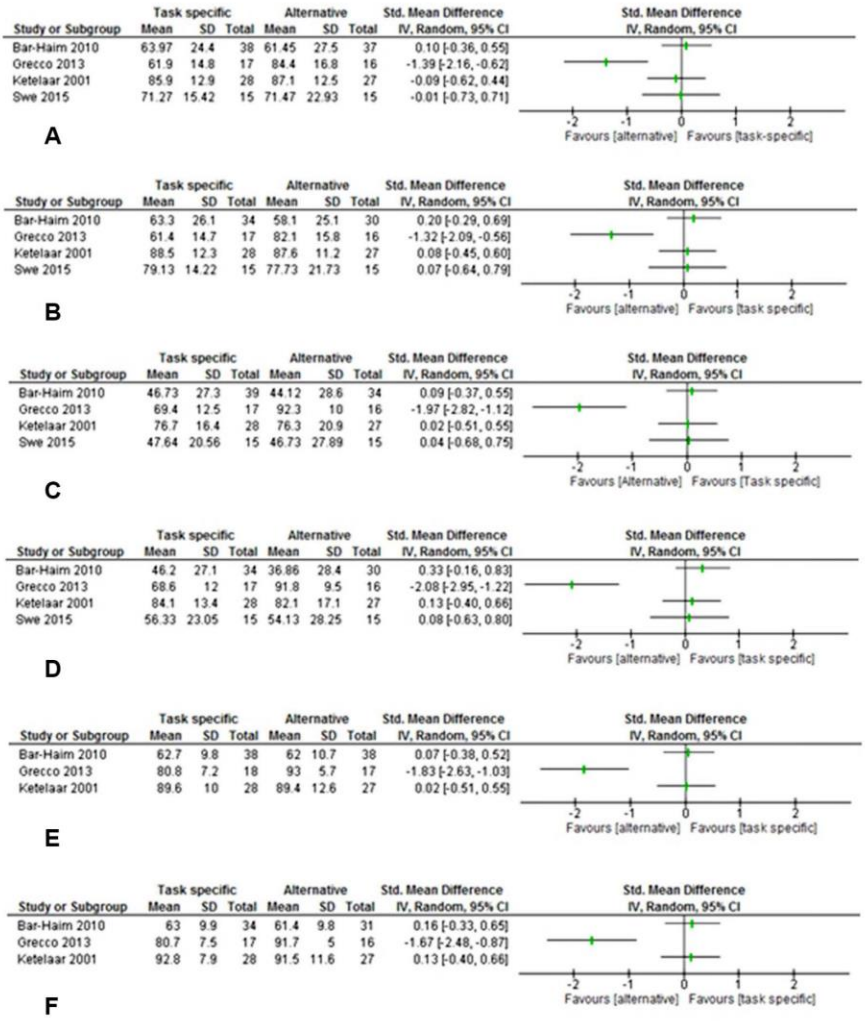



**Figure 3** (A) Gross motor function—task specific vs alternative: standing ability (Gross Motor Function Measure (GMFM) domain D) at T1. (B) Gross motor function—task specific vs alternative: standing ability (GMFM domain D) at T2. (C) Gross motor function—task specific vs alternative: walking, running and jumping ability (GMFM domain E) at T1. (D) Gross motor function—task specific vs alternative: walking, running and jumping ability (GMFM domain E) at T2. (E) Gross motor function—task specific vs alternative: overall GMFM score at T1. (F) Gross motor function—task specific vs alternative: overall GMFM score at T2.

levels for TST were also reported but without comparison group data.[26]

### Task-specific versus task-specific

#### *Interventions*

Three studies compared two or more task-specific interventions including one RCT (level II)[32] and two comparative studies (level III).[34 35] Two studies[32 34] compared the effect of TST programmes with different feedback conditions on a throwing task (table 1). The throwing task training was informed by motor learning principles but was not goal-directed (table 3). The intensity of TST in both studies was approximately 2 hours over 2 days.[32 34] The other study examined the effects of goal-directed TST compared with activity-based TST,[35] with 18 sessions over 12 weeks with parent involvement across various settings.[35] The goal-directed programme involved group and individual sessions while the activity programme was individual only.

#### *Outcomes*

Gross motor skill performance (specific to the trained task), assessed by throwing accuracy, was the sole outcome in both throwing studies.[32 34] This was assessed immediately postintervention (T1—acquisition) in both studies, 24 hours later in one[32] (T2—retention) and 3 days later

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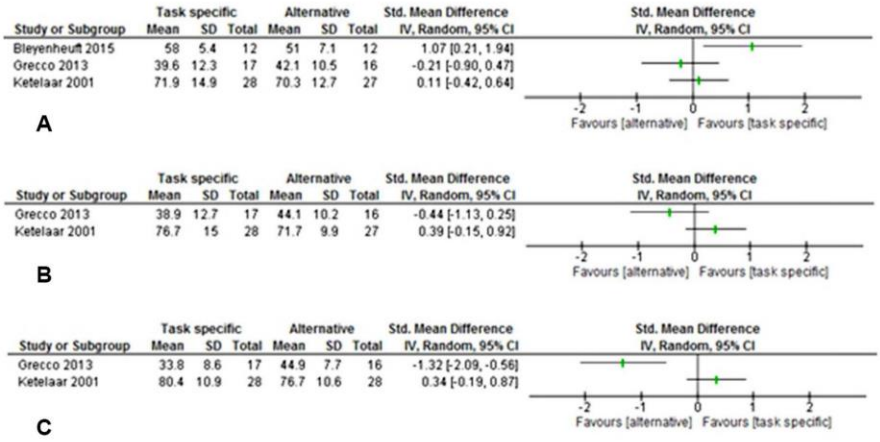



**Figure 4** (A) Functional skills—task specific vs alternative: self-care at T1. (B) Functional skills—task specific vs alternative: self-care at T2. (C) Functional skills—task specific vs alternative: mobility skills at T1.

(T2) in the other.[34] Outcomes were assessed at T1 only in the remaining study: including gross motor function (total GMFM score) and functional skills (all domains of the PEDI). The participation-related outcome, goal attainment, was assessed in the goal-directed group only by the Goal Attainment Scale (GAS).[35]

Knowledge of results provided every trial was found to improve throwing accuracy significantly more than 50% or no feedback at T1. However, at T2, those receiving knowledge of results 50% of the time performed significantly better than those who received feedback every trial or no feedback at all.[34] Self-controlled feedback was found to improve throwing accuracy significantly more than yoked feedback at T2 but not at T1.[32] Goal-directed training improved overall gross motor function and the functional skills of mobility and self-care significantly more than activity-based training.[35] There was no difference in social function between the groups. Eighty-five per cent of goals were attained with goal-directed TST.[35]

### Level IV group design

One repeated measures study involving goal-directed, group-based, intensive TST was included[36] (table 1, table 3). Gross motor function (overall GMFM score), functional skills (mobility, self-care and social function using the PEDI) and goal attainment (GAS) were measured at three baseline time points, immediately (T1) and 3 weeks (T2) after the intervention. Positive effects were reported for overall gross motor function at both T1 and T2, and for self-care skills at T2, while 66% of goals were attained at T2.

### Single-subject design

One level I single-subject design study involving randomisation to one of three feedback protocols for learning to move an exercise vehicle backward was included[14] (table 1). Gross motor skill performance (specific to the trained task) was measured using backward displacement of the vehicle 2 days following the training (T1). Eight of the 13 participants demonstrated significant improvement at T1. No specific feedback protocol was clearly superior.

### Intervention replicability

Reporting of intervention characteristics varied widely (table 3). Four studies reported intervention providers were trained in the intervention protocol,[25 27 33 36] five studies reported the format of the intervention[14 25 26 35 36] while participant adherence was described in five studies.[25 26 30 31 36] No study reported provider fidelity while comparison interventions were generally ambiguously reported.

### Quality of the evidence by outcome

The overall quality of evidence was moderate for gross motor skill performance and functional skills and low for gross motor function and participation-related outcomes.

## DISCUSSION

To our knowledge, this is the first systematic review to evaluate and synthesise the evidence for task-specific gross motor skills training in ambulant school-aged children with CP. Given the focus of TST is on practice of tasks rather than remediating impairments, and the increasing recognition of importance of child and family centred effects of interventions, this review explicitly focused on activity and participation outcomes. In general, positive within-group effects of TST were reported across outcomes of interest. However, in RCTs where TST was compared with comparison interventions, between-group effects were largely non-significant with the exception of two studies[25 28] reporting large but conflicting effects. Overall, there was moderate quality evidence for conflicting effects of TST to improve specific

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skills performance and functional skills, but low quality evidence showing no difference or negative effects on gross motor function. For participation-related outcomes, low quality evidence for positive effects of TST was found. Positive effects across all outcomes were generally found immediately following TST, with evidence of long-term retention lacking.

The secondary aim of this study was to identify motor learning strategies reported in TST and assess any relationship to outcome. The overall poor reporting of motor learning strategies and heterogeneity in interventions and outcomes limited definitive conclusions. However, this review provides some insight into potentially important characteristics of interventions, in particular practice dosage, feedback and goals. Unsurprisingly, given its intrinsic relationship to TST, the most consistently reported motor learning strategy was repetitive practice. While reporting of dosage was variable, the largest positive effects of TST were found where dosage was highest.[25] Although no specific feedback condition emerged as clearly superior in the three studies comparing these,[14 32 34] results suggest different feedback conditions may influence the phases of training differently. Replication of these studies for varying tasks is required to provide further clarity into the role of feedback. Although causal inferences are limited due to lack of randomisation, better activity outcomes were found when the TST was goal-directed.[35] These results are consistent with the growing evidence base for interventions targeted towards the goals of children with CP and their families.[6 16]

Our review found some conflicting results between studies, with large positive and negative effects found in the studies by Bleyenheuft *et al*[25] and Grecco *et al*,[28] respectively. The TST in the study by Bleyenheuft *et al*[25] was high in dose, explicitly driven by motor learning principles and involved a wide range of gross motor tasks including ball skills, cycling and walking. By contrast, in the study by Grecco *et al*,[28] dosage of TST (overground walking) was low in comparison and limited motor learning strategies were reported. The comparison treatment in this study was treadmill training without body weight support—training that may be regarded as task-related but was not deemed TST given the primary goal of the study was to improve overground walking. While practice dose was equal between the groups in the study by Grecco *et al*,[28] task progression was potentially greater with the use of treadmill functions. These results suggest task-related adjuncts, such as treadmill training, may be more beneficial than pure TST for developing (rather than acquiring) gross motor skills, such as walking in this already-ambulant population.

### Limitations across the included studies

Low-to-moderate quality of evidence was found across the outcomes due to significant methodological limitations in the studies, in addition to risk of bias. First, sample size calculation was not reported in most studies (table 1). Wide CIs found for outcome data from level II to III studies (figures 2-4) suggest inadequately powered samples may have reduced precision and thus limit the generalisability of the findings. Second, the limited detail in reporting potentially reduces the robustness of research findings if the study cannot be replicated, and limits implementation. Third, limitations in outcome measurement selection may have influenced the generalisability and transferability of some findings. The GMFM and the PEDI are a well-established tools for evaluating change in children with CP,[38] and were used in six[27 28 30 33 35 36] and four studies[25 28 33 35], respectively. However, concerns have been raised about the responsiveness of these measures in higher functioning children,[38] the target population of this review. While common use makes them appealing, their broad focus means skills targeted by the TST may not have been adequately captured. Finally, far fewer participation-related outcome measures were used compared with activity outcomes thus any evidence that improved activity through TST leads to improved participation is weak at best.

### Limitations of this review

This review chose to narrow the inclusion criteria to ambulant school-aged children with CP, interventions of gross motor skill TST and activity and participation domains outcomes, in order to draw specific conclusions. This was not possible for all questions posed by this review due to study design issues and heterogeneity, which also precluded meta-analysis. Furthermore, the methodology of the review itself has some limitations. The impact of publication bias was not evaluated. Inclusion was limited to published articles in English meaning some studies, including grey literature, may have been missed. Furthermore, non-randomised studies were retained in this review. Although the Cochrane Risk of Bias tool and the GRADE system are the most widely used systems for assessing risk of bias and the quality of the evidence across outcomes, respectively,[20 24] these tools do emphasise randomised studies.

### Conclusions and recommendations

Despite the largely positive within-group effects of TST over time across each outcome of interest, between-group effects were conflicting for skill performance and functional skills, positive for participation-related outcomes while no difference or negative effects were found for gross motor function. Given the low-to-moderate quality of this evidence, there is currently limited evidence to support task-specific gross motor skills training for improving these activity and participation-related outcomes in children with CP. Clear recommendations around whether TST is superior to other interventions cannot be made. Before conclusions can be made about any relationship of motor learning strategies to outcome, more consistent reporting and studies designed to test this are required.

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The importance of tailoring motor interventions to individual goals and lifelong physical activity is increasingly being recognised.[4 39] Thus, to enable specific recommendations, strengthening the evidence base is imperative. Adequately powered samples, rigorous study design and consistent reporting with attention to reporting interventions to allow for reproducibility and appropriate evidence synthesis is required. Future challenges also include considering issues with outcome measure responsiveness and intervention heterogeneity, and optimising TST through the use of motor learning strategies.

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**Contributors** All named authors contributed to the design of this review. RT collected data, carried out data analyses and led interpretation and reporting. CB collected data and contributed to interpretation and reporting. ARH, JLM and AJS contributed to data analysis, interpretation and reporting. All named authors approved the final manuscript as submitted.

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**Data sharing statement** Data relating to the quantitative synthesis are available on request.

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## 3.2. Supplementary appendix: Search strategy for Ovid databases

1. cerebral palsy.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
2. child*.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
3. (Task specific or skill specific or task oriented or activity focussed or task practice or goal directed).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
4. (motor imagery or implicit or error reduced or neuromotor task or parent assisted or group or cognitive orientation or repetit*).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
5. (perceptual motor or kinaesthetic explicit or action observation or dual task or trial and error or practice or imitation or experiential or discrimination or discovery or errorless or analogy or observational or exploratory).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
6. (Training or intervention or therapy or approach or learning or program).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
7. (Motor skill* or motor function* or movement skill* or Motor performance or task performance or occupational performance or motor competence or task competence or occupational competence or skill acquisition or skill attainment or skill proficiency or skill achievement or goal attainment).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
8. 3 or 4 or 5
9. 1 and 2 and 6 and 7 and 8

## 3.3. Supplementary appendix: Table 4: GRADE summary of findings

| **Outcome** | **No. of participants (studies)** | **Quality assessment** | | | | | | | | **Overall GRADE** |
|---|---|---|---|---|---|---|---|---|---|---|
| | | **Reasons to downgrade** | | | | | **Reasons to upgrade** | | | |
| | | **Study limitations** | **Consistency** | **Directness** | **Precision** | **Publication bias** | **Magnitude of effect** | **Dose response gradient** | **Bias would reduce effect** | |
| Gross motor skill performance | 122 (5) | No major limitations | Inconsistent | Direct | Precise | No evidence of bias | Cannot determine | Cannot determine | No evidence of bias | Moderate |
| Gross motor function | 217 (5) | No major limitations | Inconsistent | Indirect | Precise | No evidence of bias | Cannot determine | Cannot determine | No evidence of bias | Low |
| Functional skills | 187 (4) | No major limitations | Inconsistent | Direct | Precise | No evidence of bias | Cannot determine | Cannot determine | No evidence of bias | Moderate |
| Participation-related | 62 (3) | Major limitations | Consistent | Indirect | Precise | No evidence of bias | Cannot determine | Cannot determine | No evidence of bias | Low |

# CHAPTER 4: TRAINING TWO-WHEEL BIKE SKILLS IN CHILDREN WITH CEREBRAL PALSY: A PRACTICE SURVEY OF THERAPISTS IN AUSTRALIA

## 4.1. Preamble

A lack of knowledge regarding therapist practice when training two-wheel bike riding skills was highlighted in the literature review (Chapter 2). An understanding of current practices informs the design of the RCT (Chapters 5 and 6), especially the experimental and comparison interventions, as well as selection of clinically relevant outcome measures.

This chapter contains the manuscript of this practice survey reprinted from *Physical and Occupational Therapy in Pediatrics.* Details of the author contributions and funding sources are listed in the preface to the thesis. This manuscript uses American Psychological Association 6$^{th}$ style referencing as required by the journal with references at the end of the chapter. Co-author authorisation forms are included at the end of this thesis (Appendix IV) As the author of this article, no third-party copyright permission is required (Appendix V). The ethics approval certificate, participant information and consent form, and survey instrument pertaining to this chapter are also included at the end of the thesis (Appendices VI - VIII).

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# Training Two-Wheel Bike Skills in Children with Cerebral Palsy: A Practice Survey of Therapists in Australia

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**ABSTRACT**

*Aim*: To describe current practices of physiotherapists and occupational therapists when training two-wheel bike skills in children with cerebral palsy (CP) within an International Classification of Functioning, Disability and Health (ICF) framework. *Methods*: Ninety-five physiotherapists and occupational therapists working with children with CP in Australia completed a customized online survey. Survey questions related to: eligibility, initial assessment, intervention characteristics, and evaluation of effectiveness. Open-ended responses were analyzed using deductive content analysis. Close-ended questions were analyzed using descriptive statistics. The ICF was used as a framework for coding and reporting. *Results*: Body structure and function factors were most frequently considered in eligibility (56% of observations) and assessment (47%). Activity and participation-related factors were considered more in intervention (42%) and evaluation (75%). While functional training approaches were predominant, intervention characteristics varied markedly. Excepting goal-related tools, few measures were identified for assessment or evaluation of effectiveness. Environmental and personal factors were seldom considered across practice areas. *Conclusions*: Current two-wheel bike skills training for children with CP in Australia appears highly variable. Development and testing of bike skills-specific outcome measures and interventions and guidance for therapists on consideration of environmental and personal factors are warranted.

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Bicycle skills; cerebral palsy; children; cycling; occupational therapy; physical activity; physical therapy

## Introduction

Cerebral palsy (CP) is the most common cause of childhood physical disability affecting one in every 500 births globally (Graham et al., 2016). CP is a disorder of movement, which can affect muscle tone, coordination, movement control, balance and posture to varying degrees (Bax et al., 2005). The most widely used tool to classify the gross motor function of children with CP is the Gross Motor Classification System (GMFCS)

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(Palisano et al., 1997). Children classified as GMFCS levels I-II are generally able to walk independently without mobility aids whilst children classified as GMFCS level V are transported by wheelchair in all settings. Children with CP generally have lower levels participation in physical and recreational activities than their typically developing peers (Bjornson, Belza, Kartin, Logsdon, & McLaughlin, 2007), even those with lesser impairment at GMFCS I-II, who represent sixty per cent of Australian children with CP (Australian Cerebral Palsy Register, 2016). This is a concern because of the increased risk of chronic disease in adulthood associated with inactivity in childhood (Fernandes & Zanesco, 2010). Higher levels of physical activity has also been associated with better quality of life and happiness in children with CP (Maher, Toohey, & Ferguson, 2016).

Bike riding is a common activity for children and their families (Australian Bureau of Statistics, 2012) and is a key means of active travel—a current public health priority area for children (Australian Health Policy Collaboration, 2018). The benefits of active travel in children include cardiorespiratory fitness, improved academic performance and attention and the promotion of physical activity into adulthood (Raje & Saffrey, 2008). Learning to ride a two-wheel bike can arise as a rehabilitation goal of children with CP, in particular those classified GMFCS I-II. Given the increasing need for interventions that target physical activity and are meaningful to the child and their family (Novak et al., 2013), knowledge of training bike skills in this group is important.

Until recently, little was known about the ability of children with CP to ride a two-wheel bike. We recently found that fewer children aged 6–15 years with CP (GMFCS I-II) had learnt to ride at each age and if they were riding, they learnt later than typically developing peers (Toovey, Reid, Rawicki, Harvey, & Watt, 2017). These findings support the need for research into training approaches for ambulant children with CP who are not yet able to ride independently.

In Australian clinical practice, physiotherapists (PTs) and occupational therapists (OTs) commonly provide therapy to children with CP to improve motor skills or daily task performance, including two-wheel bike riding (Australian Institute of Health & Welfare, 2006). However, there is very little direct evidence to guide therapists how to best train two-wheel bike specific skills. Existing studies in this population on stationary bikes show positive effects on bone mineral density, muscle strength and health-related quality of life (Chen et al., 2013, 2012; Demuth, Knutson, & Fowler, 2012; Fowler et al., 2007; Johnston, Lauer, & Lee, 2008). However, there is no evidence about whether stationary bike training (a closed skill) translates to riding a two-wheel bike (an open skill). Among other factors, the high-level dynamic balance requirements of two-wheel bike riding distinguish it from stationary cycling, as well as similar activities including riding a tricycle or bike with training wheels (Halayko, Magill-Evans, Smith, & Polatajko, 2016). It is reasonable to expect each aspect of training to differ between these skills.

There is some positive evidence for task-specific gross motor skills training on motor skill performance and participation in this population, however, it is limited by intervention heterogeneity and study design issues, and not specific to bike riding (Toovey, Bernie, Harvey, McGinley, & Spittle, 2017). Cognitive, goal-directed strategies within a task-specific approach to bike riding have been tested in a small case series of youth with Down Syndrome in Canada (Halayko et al., 2016). Positive physical activity, social or skill learning outcomes are reported in several case series involving task-oriented

approaches to training bike skills in children with autism spectrum disorder and down syndrome in the United States (MacDonald et al., 2012; Ulrich, Burghardt, Lloyd, Tiernan, & Hornyak, 2011) and mixed developmental disabilities in the United Kingdom (Dunford, Rathmell, & Bannigan, 2016). To date, there are no high-quality studies specific to training two-wheel bike skills reported in children with CP.

In order to guide the development and testing of novel interventions, it is important to understand current clinical practices (Craig et al., 2013) within the context of contemporary disability and health theoretical frameworks. This is of particular importance in the pathway to randomized controlled trials. Current practices specific to training bike skills are not known. The International Classification of Functioning, Disability and Health (ICF) offers a well-accepted framework for understanding the effect of CP on the individual (Schiariti & Masse, 2014). It can also act as a system to describe interventions, assessment procedures or outcome measures by areas of functioning (ICF domains) (Martin, Baker, & Harvey, 2010) and has been used to report current therapy practices (Anaby et al., 2017; Tantilipikorn, Watter, & Prasertsukdee, 2012).

The primary aim of this study is to describe the current practices of Australian PTs and OTs regarding two-wheel bike skills training for children with CP including; determination of eligibility, initial assessment, training characteristics and outcome measurement, using the ICF as a framework. The secondary aim is to explore the representation of ICF domains within each area of practice.

## Methods

Ethical approval for this cross-sectional survey was granted by the Human Research Ethics Committee at the Royal Children's Hospital, Melbourne, Australia.

### *Participants*

Eligible participants were PTs and OTs working with children with CP in either or both hospital and community settings in Australia. Participants were recruited via online methods and data were collected over a 6-week period in May and June 2016. Written informed consent was received from all participants. PTs and OTs registered as providers for the Better Start for Children with Disabilities Initiative (Better Start—a federally funded early intervention scheme for children with disabilities in Australia (Department of Social Services, 2017)) ($n = 300$) and those who were members of the Australasian Academy of Cerebral Palsy and Developmental Medicine ($n = 427$) were invited and sent two reminders via email. While these broad methods aim to capture the majority of therapists working with children with children with CP in Australia, not all therapists registered with Better Start or who are members of the Australasian Academy work with children with CP, affecting their eligibility to participate. To improve the representativeness of this sample, snowball recruitment methods via the professional network of the research team, and advertisements on the websites and e-newsletters of the Australian Physiotherapy Association and a nationally funded Centre of Research Excellence in Cerebral Palsy, were also employed.

A total of 194 responses to the invitation were received (27% response rate), of which 186 therapists remained eligible following screening at the start of the survey. Ineligible participants were not trained as PTs or OTs or did not work with children with CP. Characteristics of the 95 therapists who completed the full survey (51% completion rate) including 79 PTs (83% of all therapists) and 16 OTs (17%) are presented in Table 1. Participants reported a wide range of years of practice and proportion of their clinical caseload consisting of children with CP. They worked across a range of settings and locations with the highest proportion working in community private practice (24%) and the state of Victoria (45%). Fifty-five therapists (58%) had specific experience training bike skills in children with CP (Table 1). The majority of participants (77%) expected children who walk without aids (GMFCS I-II) or have somewhat reduced quality of

**Table 1.** Characteristics of physiotherapists (PT) and occupational therapists (OT) who completed the survey.

| | PTs | OTs | Total |
|---|---|---|---|
| Sample (n) | 79 (83%) | 16 (17) | 95 |
| Clinical caseload—children with CP | | | |
| <25% | 23 (29) | 8 (50) | 31 (33) |
| 26–50% | 23 (29) | 4 (25) | 27 (28) |
| 51–75% | 21 (27) | 1 (6) | 22 (23) |
| >75% | 12 (15) | 3 (19) | 15 (16) |
| Caseload of children with CP classified GMFCS levels I–II | | | |
| <25% | 31 (39) | 5 (31) | 36 (38) |
| 26–50% | 21 (27) | 3 (19) | 24 (25) |
| 51–75% | 17 (22) | 2 (13) | 19 (20) |
| >75% | 8 (10) | 5 (31) | 13 (14) |
| Do not work with children classified GMFCS I-II | 2 (2) | 1 (6) | 3 (3) |
| Years working with children with CP | | | |
| <1 year | 3 (4) | 1 (6) | 4 (4) |
| 1–5 years | 18 (23) | 3 (19) | 21 (22) |
| 6–10 years | 21 (26) | 2 (12) | 23 (24) |
| 10–15 years | 15 (19) | 3 (19) | 18 (19) |
| >15 years | 22 (28) | 7 (44) | 29 (31) |
| Primary practice setting | | | |
| Community setting | | | |
| Public | 11 (14) | 3 (19) | 14 (15) |
| Private | 16 (20) | 7 (44) | 23 (24) |
| Early childhood intervention | 14 (18) | 2 (13) | 16 (17) |
| Metropolitan hospital | | | |
| Acute department | 4 (5) | 0 (0) | 4 (4) |
| Rehabilitation department | 8 (10) | 3 (19) | 11 (12) |
| School service | 10 (13) | 0 (0) | 10 (11) |
| Regional hospital | | | |
| Acute department | 0 (0) | 0 (0) | 0 (0) |
| Rehabilitation department | 7 (9) | 1 (6) | 8 (8) |
| Other | 9 (11) | 0 (0) | 9 (9) |
| Geographical location of practice | | | |
| Victoria | 32 (41) | 11 (69) | 43 (45) |
| New South Wales | 16 (20) | 1 (6) | 17 (18) |
| South Australia | 8 (10) | 2 (13) | 10 (11) |
| Western Australia | 9 (11) | 1 (6) | 10 (11) |
| Queensland | 8 (10) | 1 (6) | 9 (9) |
| Northern Territory | 3 (4) | 0 (0) | 3 (3) |
| Tasmania | 2 (3) | 0 (0) | 2 (2) |
| Australian Capital Territory | 1 (1) | 0 (0) | 1 (1) |
| Experience providing bike skills training to children with CP | | | |
| Yes | 48 (61) | 7 (44) | 55 (58) |
| No | 31 (39) | 9 (56) | 40 (42) |

movement in their upper limbs (Manual Ability Classification System, MACS I-II) could learn to ride a two-wheel bike.

### Web-based survey

The customized web-based survey was designed by the study authors and managed using secure Research Electronic Data Capture (REDCap) tools (Harris et al., 2009). The survey was piloted with three therapists prior to use. The survey took between 10 and 15 minutes to complete and contained five main components: (1) therapist characteristics, (2) child eligibility for receiving training, (3) initial assessment, (4) training characteristics, and (5) evaluation of effectiveness.

Respondents provided demographic data regarding their profession, practice setting and experience working with children with CP. They rated their expectations and experience of training two-wheel bike riding ability in children with CP by GMFCS and MACS levels. Items related to eligibility, assessment and training included open-ended questions to describe practice, followed by closed-ended Likert scale-type frequency responses (always, sometimes, never) to defined practice descriptors (i.e. potential factors influencing eligibility, components assessed and types of training). This two-step process was designed to enable the identification of areas of practice where terminology is less developed or there are mismatches when described in clinicians' own language. Practice descriptors were selected based on review of literature, authors' clinical experience and guidance from the pilot survey reviewers. Questions related to training characteristics were also guided by the Template for Intervention Descriptions and Replication Checklist (Hoffmann et al., 2014) including: key intervention approaches, specific strategies, equipment/materials used, providers, format (individual or group), settings and dosage. Finally, respondents described methods for evaluating effectiveness of the training via open-ended responses. Further details regarding the survey can be obtained from the authors on request.

### Data analysis

Quantitative and qualitative analyses were utilized. Open-ended responses were analyzed through a qualitative approach using deductive content analysis using the ICF as a framework. Two researchers (RT and AN) conducted the analysis independently by reading responses several times, breaking the responses down into observations (for example, an outcome measure or distinct type of training), and then coding into ICF domains. When there was discrepancy between codes, both researchers discussed the results until consensus was reached with final consensus on coding reached through review and discussion between all authors.

Reponses to closed-ended questions were summarized as proportions of responses for each frequency of practice and coded to ICF domains for reporting. Quantitative analyses were conducted using Stata version 15 (StataCorp, College Station, TX). To identify the most frequently addressed ICF domains within each aspect of practice, frequency scores representing the proportion of observations within the open-ended responses related to each domain were calculated and reported.

## Results

### Eligibility

Therapists identified factors from all ICF domains as considerations for provision of bike skills training (Table 2). Therapists could report more than one consideration making the total number of observations reported 364. The most commonly reported considerations from each domain from open-ended responses were; health condition—uncontrolled epilepsy, (9% of all observations), body functions—intellectual impairment (12%), activities and participation—current bike riding related skills (6%), environmental factors—family support and engagement (4%) and personal factors—child motivation or interest in riding a bike (11%). The provided descriptor most frequently selected as "always" a contraindication was severe intellectual impairment (42% of therapists).

### Initial assessment

Components relating to all ICF domains except for health condition were identified as part of the initial assessment when training bike skills (Table 3). The body structures and body functions descriptors most frequently selected as "always" assessed were balance (86% of therapists), range of motion (83%), coordination (69%) and strength and spasticity (both 64%). Activity and participation-related descriptors; child and family goals, and bike riding skills/experience were identified as components "always" assessed by 99% and 88% of participants respectively. Components related to environmental (8% of all observations) and personal factors (7%) identified through open-ended responses included type of bike, suitable location to practice and motivation or fear association with bike riding.

### Outcome measures

A variety of outcome measures and assessment tools used to assess balance, strength, range of motion, movement disorder type, gross motor function and skills, goals and use of equipment/modifications (Table 3) were identified through open-ended responses. The frequency of these responses was not analyzed. The Performance Quality Rating Scale was reported as a tool to measure performance of two-wheel bike specific skills, however, no published measures specific to bike skills were identified.

### Intervention

#### Key intervention approaches

A range of intervention approaches to training two-wheel bike skills across most ICF domains were reported (Table 4). The most frequently described in open-ended responses were goal-directed training (22% of observations) and task-specific training (15%). Selection of these approaches from practice descriptors was higher, with 72% of respondents "always" utilizing task-specific training, 67% "always" using goal-directed training and 39% "always" implementing activity-focused training. Approaches targeting body functions were also selected including balance and strength training—"always"

**Table 2.** Eligibility considerations for children with CP to undertake two-wheel bike skills training.

| ICF domain | Eligibility considerations (open-ended) | Frequency (%) 364 observations | Potential contraindication (closed-ended) | Frequency (%)[a], $n = 92–95$ | |
|---|---|---|---|---|---|
| | | | | Always | Sometimes |
| Health condition | • Uncontrolled epilepsy | 31 (9) | | | |
| | • Other medical condition (e.g. osteoporosis or post-surgery) | 19 (5)<br>Total = 50 (14%) | | | |
| Body functions & body structures | • Intellectual impairment | 44 (12) | • Moderate intellectual impairment<br>• Severe intellectual impairment<br>• Challenging behavior<br>• Bilateral distribution | 13 (14%)<br>39 (42%)<br>11 (12%)<br>8 (9%) | 69 (73%)<br>47 (50%)<br>79 (83%)<br>44 (47%) |
| | • Visual impairment | 22 (6) | | | |
| | • Range of motion (UL & LL) | 20 (5) | | | |
| | • Challenging behavior | 19 (5) | | | |
| | • Trunk control | 17 (5) | | | |
| | • Balance | 13 (4) | | | |
| | • Orthopaedic deformity (e.g. hip displacement) | 8 (2)<br>7 (2) | | | |
| | • Type & distribution of movement disorder | 7 (2)<br>5 (1) | | | |
| | • Hearing impairment | 4 (1) | | | |
| | • Strength (UL & LL) | 1 (<1) | | | |
| | • Motor control and coordination | Total = 203 (56%) | | | |
| | • Vestibular function | | | | |
| Activities & participation | • Bike riding skills (ability to pedal, steer) | 20 (6)<br>16 (4) | • MACS III-V<br>• GMFCS III-V<br>• Poor ability to communicate with therapists | 14 (15%)<br>17 (18%)<br>10 (11%) | 69 (73%)<br>65 (68%)<br>75 (79%) |
| | • Level of UL function | 8 (2) | | | |
| | • Level of functional mobility | 3 (1)<br>Total = 47 (13%) | | | |
| | • Communication limitation | | | | |
| Environmental factors | • Family support & engagement (including family goal) | 13 (4)8 (2)<br>4 (1)<br>Total = 25 (7%) | | | |
| | • Access to bike | | | | |
| | • Safe/appropriate location to ride | | | | |
| Personal factors | • Child's motivation / interest / own goal to ride a bike | Total = 39 (11%) | • Age < 5 years | 8 (9) | 55 (59) |

[a]Only items where >50% of participants selected "sometimes" or "always" a contraindication are reported.
GMFCS = gross motor function classification system; LL = lower limb; MACS = manual ability classification system; UL = upper limb.

**Table 3.** Initial assessment clinicians would undertake when providing two-wheel bike skills training to children with cerebral palsy.

| ICF domain | Components assessed (open-ended) | Frequency (%) for ICF domain, total = 186 observations | Components assessed (closed-ended) | Frequency (%)[a], n = 94–95 | | Outcome measure/ assessment tool identified (open-ended responses) |
|---|---|---|---|---|---|---|
| | | | | Always | Sometimes | |
| Body functions & body structures | • Balance and postural control<br>• Protective reaction times / accuracy<br>• Upper & lower limb strength<br>• Neck, upper limb & lower limb ROM, muscle length<br>• Muscle tone & movement disorder type and topography<br>• Motor control, planning and coordination<br>• Cognition<br>• Cardiovascular endurance<br>• Vision - visual perception & attention oculomotor control<br>• Vestibular function<br>• Proprioception, sensation and pain | Total = 87 (47) | • Balance<br>• Strength<br>• Range of motion<br>• Spasticity<br>• Coordination<br>• Motor planning ability<br>• Learning style<br>• Aerobic capacity | 82 (86)<br>61 (64)<br>79 (83)<br>61 (64)<br>66 (69)<br>55 (58)<br>45 (47)<br>5 (16) | 12 (13)<br>33 (35)<br>15 (16)<br>31 (33)<br>27 (28)<br>38 (40) 43 (45)<br>60 (63) | • CTSIB, ECAB, Pediatric Balance Scale<br>• Manual Muscle Test, Functional Strength Test<br>• Goniometry, SAROMM<br>• ASAS, HAT, MAS, Tardieu scale |
| Activities & participation | • Gross motor skills & functional mobility e.g. sitting, standing, walking, stairs, running, climbing<br>• Bike riding skills<br>• Participation in bike riding activities<br>• Activity or participation related goal<br>• Communication skills (expressive & receptive)<br>• Ability to respond to & navigate environment | Total = 72 (39) | • Bike riding skills and experience<br>• Child & family goals[a] | 83 (88)<br>94 (99) | 11 (12)<br>(1) | • GMFM, FMS, HiMAT, 10WT, 6MWT<br>• PQRS, Observation and/or video assessment<br>• COPM, GAS |

| | | | |
|---|---|---|---|
| | • Ability to problem-solve<br>• Application of knowledge<br>• Ability to perform skills safely | | |
| Environmental factors | • Type of bike<br>• Use of specialized equipment or modifications<br>• Safe/appropriate location<br>• Access to bike<br>• Opportunities to practice | Total = 14 (8) | • Individually Prioritised Problem Assessment |
| Personal factors | • Motivation/interest to ride bike<br>• Age<br>• Fear associated with bike riding | Total = 13 (7) | |

[a]ICF domain not specified in provided practice descriptor thus goals may also relate to other ICF domains.

ASAS = Australian Spasticity Assessment Scale; COPM = Canadian Occupational Performance Measure; CTSIB = Clinical Test of Sensory Interaction & Balance; ECAB = Early Clinical Assessment of Balance; FMS = Functional Mobility Scale; GAS = Goal Attainment Scale; GMFM = Gross Motor Function Measure; HAT = Hypertonia Assessment Tool; HiMAT = High Level Mobility Assessment Tool; MAS = Modified Ashworth Scale; 6MWT = six-minute walk test; 10MWT = 10 m walk test; PQRS = Performance Quality Rating Scale; SAROMM = Spinal Alignment & Range of Motion Measure.

**Table 4.** Intervention approaches to training two-wheel bike skills.

| ICF domain | Intervention approach (open-ended responses) | Frequency for ICF domains[a] (%) (total = 221 observations) | Intervention approach (closed-ended) | Frequency (%), $n = 95$ | |
|---|---|---|---|---|---|
| | | | | Always | Sometimes |
| Body functions & body structures | Component-based approaches<br>• Balance training<br>• Functional and lower limb strength training<br>• Core/trunk control training<br>• Range of motion management<br>• Spasticity management<br>• Cardiovascular fitness training<br>• Compression garments | Total = 21 (10) | Balance training<br>Strength training | 34 (36)<br>26 (27) | 39 (41)<br>42 (44) |
| Activities & participation | Goal-directed training | 49 (22) | Goal-directed training | 64 (67) | 17 (18) |
| | Task-specific training/Motor-learning based training including<br>• Whole/part practice of bike-related skills<br>• Bike handling & bike safety<br>• Top-down approach | 34 (15) | Task-specific training<br>Activity-focused training | 68 (72)<br>37 (39) | 15 (16)<br>14 (15) |
| | Cognitive Orientation to Daily Occupational Performance (CO-OP) | 10 (5)<br>Total = 93 (42) | | | |
| Environmental factors | Modified equipment / trial of various types of bike<br>• Bike modifications e.g. training wheels<br>• Use of balance (walker) bike, trike, stationary bike | Total = 12 (5) | Provision of bike with training wheels<br>Use of balance (walker) bike<br>Context-focused therapy<br>Use of stationary bike | 27 (28)<br>20 (21)<br>13 (14)<br>11 (12) | 53 (56)<br>46 (48)<br>15 (16)<br>50 (53) |
| Other/ mixed domains | Neurodevelopmental Therapy (NDT)<br>Proprioceptive Neuromuscular Facilitation (PNF)<br>Child and/or family education<br>Hydrotherapy | Total = 14 (6) | NDT<br>Vojta therapy<br>Conductive education<br>Electrical stimulation | 8 (8)<br>0 (0)<br>2 (2)<br>0 (0) | 31 (33)<br>15 (16)<br>21 (22)<br>21 (22) |

[a]Except for Activities and Participation domain where frequency counts have been reported for key approaches identified.

used by 36% and 27% of respondents, respectively. Use of equipment as an overall approach was demonstrated including training wheels (28% "always" using), balance bikes (21%) and stationary bikes (12%).

### Specific strategies and materials

The key specific strategies described by therapists in open-ended responses most commonly related to type of task practice (21% of all observations), cognitive strategies (20%), and feedback to the child (19%). Information and equipment provision were also described. Information provided to families was largely related to topics including equipment, appropriate locations for practice and access to modifications. The specific modifications/equipment reported could be categorized into upper limb/lower limb/postural supports, basic modifications to two-wheel bike (e.g. changing handbrakes to less affected side) and additions (e.g. training wheels). Practice descriptor strategies selected by the majority (>50%) of respondents as "always" utilized were goal setting with child and family (84% of all respondents), feedback (82%), repetitive practice (80%), provision of a home program (71%), task modification (59%), physical guidance (55%) and environmental modification (53%).

### Providers, format, and setting

A large majority (85%) of therapists identified they would have assistance with delivering training including from a parent or carer (80% of therapists), another PT or OT (44%), therapy assistant (39%), or teacher/teacher's aide (11%). Individual delivery of training (clinician with one child/family) only was most commonly reported (42%) while some respondents (33%) would deliver training either in an individual or group format, with fewer delivering training to a group only (5%). Community settings such as parks/bike paths were the most frequently selected (27% of respondents "always" used) location for training, followed by the child's home (25%), the child's school (16%) and the clinic environment (14%).

### Dosage

Reported dosage of training was highly variable (Table 5). The length of training sessions ranged from 20–120 minutes with sessions of 30–60 minutes most commonly reported (59% of therapists). The number of sessions ranged from 1–20 while four to six sessions were most commonly reported (52%). The overall training period ranged from 1 week–12 months with 6–12 weeks the most commonly reported period (22%).

### Evaluation of training effectiveness

Respondents most frequently identified evaluation of goal attainment (44% of observations) as a method for evaluating the effectiveness of two-wheel bike skills training (Table 6), with the Canadian Occupational Performance Measure and Goal Attainment Scale identified as tools. Evaluation of bike skills performance through observation (18%), video analysis (4%),

and use of the Performance Quality Rating System (1%) were also described, however, no published measures specific to two-wheel bike skills were reported.

### *ICF domain representation across areas of practice*

Percentages of observations within open-ended responses within practice areas by ICF domain are demonstrated in Figure 1 and Tables 2–4, 6. Eligibility most commonly considered body structures and body functions, with 56% of observations within this domain. Similarly, initial assessment of body structures and body functions components were the most frequently reported (47%) followed by assessment of activities and participation components (39%). Intervention approaches and methods of evaluating effectiveness related to body structures and body functions were less often identified (10% and 3% respectively). Intervention approaches identified by clinicians predominantly targeted the activities and participation domains (42%) as did methods for evaluating effectiveness (70%). Across each area of practice, environmental factors were consistently less frequently identified (1–7%) while personal factors were considered in eligibility (11%) and assessment (7%) only.

## Discussion

This is the first study to describe the current practices of physiotherapists and occupational therapists for training two-wheel bike skills in children with CP in Australia. There are no other known reports of typical clinical practice of bike skills training for children with CP worldwide. The spectrum of practice from eligibility considerations to assessment, intervention and evaluation was comprehensively explored within an ICF framework. This survey had broad reach—capturing responses from participants from each state and territory in Australia and representing a diverse range of experience and settings.

We found that although initial assessment components for bike skills training most commonly related to body functions and body structures, assessment of activity and participation components, in particular child and family goals, were also frequently reported. Goal-directed and task-specific training were the predominant intervention approaches selected when provided as practice descriptors, however, were less often reported within open-ended responses. These findings are consistent with a recent broader survey of current rehabilitation practice of Canadian PTs and OTs for children with CP that found body structures and body function and activities and participation domains were the focus of assessment (Anaby et al., 2017). However, our findings regarding intervention practice differed from the Canadian study finding that both of these domains were also the focus of intervention. Our findings more clearly suggest

**Table 5.** Reported dosage of two-wheel bike skills training.

| Length of training sessions (minutes) | *n* (%) | Number of training sessions | *n* (%) | Length of overall training period | *n* (%) |
|---|---|---|---|---|---|
| ≤30 | 17 (18) | 1–3 | 8 (8) | ≤2 weeks | 5 (5) |
| 30–60 | 56 (59) | 4–6 | 49 (52) | >2–6 weeks | 13 (14) |
| >60 | 2 (2) | 7–10 | 14 (15) | >6–12 weeks | 21 (22) |
| Unspecified | 20 (21) | >10 | 3 (3) | >3 months | 11 (12) |
| | | Unspecified | 21 (22) | Unspecified | 45 (47) |

**Table 6.** Methods for evaluating the effectiveness of bike skills training in children with cerebral palsy.

| ICF domain | Methods of evaluation (open-ended responses) | Freq count (%) (total = 103 observations) |
|---|---|---|
| Body structures & body functions | Bike riding endurance/fitness | 3 (3%) |
| Activity & participation | Goal attainment | |
| | • Canadian Occupational Performance Measure | 22 (21) |
| | • Goal Attainment Scale | 17 (17) |
| | • Other goal setting e.g. SMART goals, child/family goals | 6 (6) |
| | | Total = 45 (44) |
| | Bike skills | |
| | • Observation of skill attainment | 19 (18) |
| | • Video analysis | 4 (4) |
| | • Performance Quality Reporting System | 1 (1) |
| | | Total = 24 (23) |
| | Participation in bike riding | 3 (3) |
| Environmental factors | Individual Prioritised Problem Assessment | 1 (1) |
| Other/mixed domains | Feedback | |
| | • Child/family or group report, reflection & evaluation | 15 (14) |
| | • Child/family satisfaction | 3 (3) |
| | • Follow-up phone review | 1 (1) |
| | | Total = 19 (18) |
| | Outcome measure not specified | 8 (8) |

Note: SMART = specific, measurable, achievable, realistic and time-limited.

the focus of intervention is on the activity and participation domain. This may be because our survey explored the specific practice of training bike skills (a functional activity) compared to the broader scope of the Canadian survey.

It is promising that the predominant interventions reported align with the growing evidence base for goal-directed and task-specific training in CP (Novak et al., 2013; Sakzewski, Gordon, & Eliasson, 2014; Toovey et al., 2017). Given research on teaching children with developmental disabilities to ride a bike in North America and the United Kingdom also focused on functional task-related training (Dunford et al., 2016; Halayko et al., 2016; MacDonald et al., 2012; Ulrich et al., 2011), our finding may be generalizable to therapist practice in these settings. However, before any best practice recommendations can be made, these functional approaches need testing in adequately powered, high quality studies of bike riding in children with CP.

The specific characteristics of interventions generally varied although most therapists identified they would require assistance from another person to deliver the training, most commonly in community settings with four to six sessions of 30 to 60 minutes in length as the most frequently reported dosage components. These findings have implications for practice and policy in terms of resource requirements—the personnel and infrastructure available may influence outcomes across different clinical settings and service providers. Although few studies have examined rationale for the amount of therapy services provided to children with CP, a 2012 study that described general therapy dosage in younger children in the United States and Canada also found 30–60 minute sessions were most commonly reported by parents (Palisano et al., 2012). Consensus on optimal dosage of motor training in children with CP remains undetermined. However, given repetition and intensity are key principles of motor learning and neuroplasticity

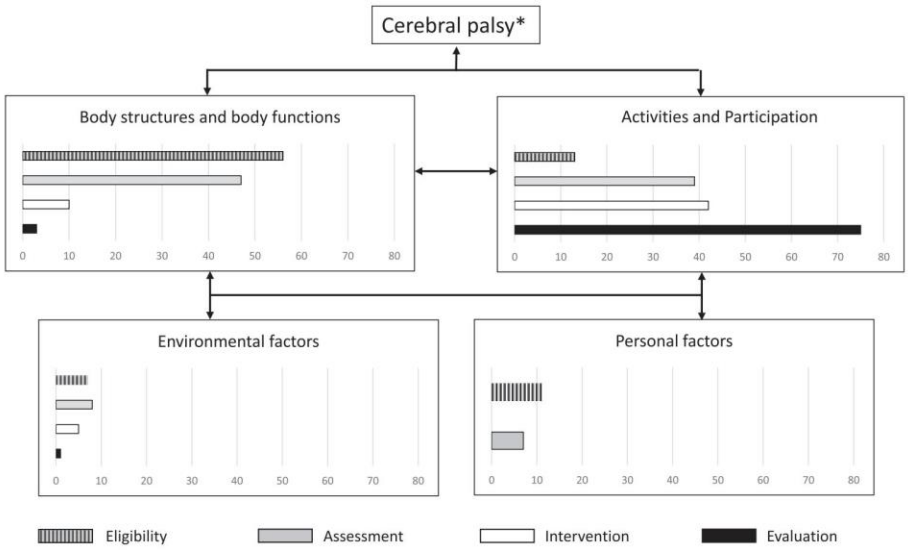



**Figure 1.** Percentage of open-ended observations within practice areas by ICF domain. Note: *14% of eligibility considerations related to cerebral palsy and associated conditions.

(Gordon, Schneider, Chinnan, & Charles, 2007; Nudo, 2003) and the emergence of evidence around intensive motor training (Sakzewski et al., 2014; Toovey et al., 2017), this finding warrants further investigation.

It is promising that evaluation predominantly related to goal attainment and bike skills performance given the recognized importance of goal-centered functional practice in rehabilitation (Brogren Carlberg & Lowing, 2013; Novak et al., 2013). Although, the findings highlight a mismatch between the frequent consideration of these factors in assessment and evaluation, and the less frequent use of established outcome tools to measure change in these areas. Despite observation and video analysis being reported as methods to assess or evaluate change, no outcome measures specific to bike skills were identified. One reason for this could be the lack of available published bike skills performance measures. Moreover, with the exception of those measuring goal attainment, our findings suggest use of outcome measures related to the activity and participation domain is limited. Poor application of activity or participation-related outcome measures is not unique to this study. A survey of upper limb therapy management in children with CP in Northern Ireland found only up to 14% of OTs or PTs used a standardized activity level measure (McConnell, Johnston, & Kerr, 2012).

Another mismatch that emerged was the frequent consideration of body structures and functions factors for eligibility and assessment yet relatively infrequent consideration of these factors in subsequent intervention and evaluation. This finding may represent a transition in clinical practice towards treatments targeting activities and participation and the functional evaluation of these. However, not for the first time, such findings raise questions around the practice of focusing assessment on impairment-related factors when training functional skills such as bike riding (Pritchard-Wiart, 2017). Environmental and personal related factors were seldom articulated across most areas of practice. Studies using the ICF or like frameworks to describe broader

rehabilitation practice in this population have had similar findings (Anaby et al., 2017; Saleh et al., 2008) raising further questions around whether these factors are considered, or if the language and tools are not available to clinicians to describe them.

### *Study limitations*

Like all surveys, this study is subject to limitations of self-reported practice and findings may not completely reflect the reality of current practice. The broad recruitment strategy and online methodology may have impacted the response rate. Therapists who received the invitation to participate through the primary strategies work with children with disabilities, but may not work with children with CP, affecting their ability to respond from the outset. While a response rate greater than 27% may have meant the findings more closely represented the practice of therapists working with children with CP in Australia, response rates for web-based surveys have been steadily declining over the last decade and a rate less than 10% is not uncommon currently (Van Mol, 2017). Despite the recruitment strategy being designed to reach across disciplines, fewer OTs were recruited limiting the generalizability of findings. This could be explained by the difference in scope of practice between disciplines and that a third fewer OTs than PTs are registered in Australia (Australian Health Practioner Regulation Agency, 2017). Just over 50% of the therapists who responded to the invitation completed the full survey, indicating that its length, structure or relevance may have limited full participation (Saleh & Bista, 2017). This drop off may also impact the representativeness of the sample and generalizability of the findings. Finally, while activities and participation are individual domains within the ICF, it was difficult to separate components of practice into either domain in the analysis and interpretation of the results. This challenge is not unique to this study, with other studies in the field also grouping these domains together for analysis and reporting (Anaby et al., 2017; Saleh et al., 2008).

## Conclusions

Overall, this study highlights variability across the spectrum of practice in two-wheel bike skills training for children with CP in Australia reflecting a lack of uniform practice. Body structures and body functions-related factors are currently primarily being considered in eligibility screening and assessment with less frequent consideration during training or evaluation. Assessment and evaluation of goals appear common and activity-related approaches to assessment and training were often reported. Consideration of personal and environmental factors was limited and participation-related practices were difficult to identify. This study highlights the need to develop and test interventions, as well as both patient-reported and performance-based outcome measures, specific to training two-wheel bike skills in this population. Guidance for clinicians on how environmental and personal factor domains of the ICF can be considered across each area of practice is also warranted.

## Acknowledgments

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## Disclosure statement

The authors report no conflicts of interest.

## Data availability statement

The data set reported in this article is available on request to the corresponding author.

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# CHAPTER 5: BIKE SKILLS TRAINING FOR CHILDREN WITH CEREBRAL PALSY: PROTOCOL FOR A RANDOMISED CONTROLLED TRIAL

## 5.1. Preamble

Drawing on evidence from the literature review (Chapter 2), systematic review (Chapter 3), the practice survey (Chapter 4), research on RCT design and conduct, as well as literature regarding outcome measurement in CP and paediatrics more broadly, this chapter details the methods for the major study within this thesis, the RCT.

This chapter contains the manuscript of this RCT protocol reprinted from *BMJ Open.* Details of the author contributions and funding sources are listed in the preface to the thesis. Non-author contributions included that of two consumer advisors Frances Wright and Donna Goldsmith whose review of the protocol informed the design of the interventions and selection of outcomes. This manuscript uses Vancouver referencing as required by the journal with references at the end of the chapter. Co-author authorisation forms are included at the end of this thesis (Appendix VIII). As the author of this article, no third-party copyright permission is required (Appendix III).

All tables and figures are included in the manuscript except for the schedule of assessments which is included in section 5.2 as a supplementary appendix following the manuscript. The ethics and governance approval certificates and participant information and consent form pertaining to this chapter, as well as the clinician training manual and information provided to parents allocated to the comparison group are also included at the end of the thesis (Appendices IX - XII)

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# Bike skills training for children with cerebral palsy: protocol for a randomised controlled trial

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## ABSTRACT

**Introduction** Two-wheel bike riding can be a goal for children with cerebral palsy (CP) and a means of participating in physical activity. It is possible for some children with CP to ride a two-wheel bike; however, currently far fewer can ride compared with their typically developing peers. Evidence supports training targeted towards goals of the child with CP and their family; yet there is little evidence to guide best-practice bike skills training. Task-specific training may lead to attainment of two-wheel bike-specific goals. This study aims to determine if a novel task-specific approach to training two-wheel bike skills is more effective than a parent-led home programme for attaining individualised two-wheel bike-specific goals in independently ambulant children with CP aged 6–15 years.

**Methods and analysis** Sixty eligible children with CP (Gross Motor Function Classification System levels I–II) aged 6–15 years with goals relating to riding a two-wheel bike will be randomised to either a novel task-specific centre-based group programme (intervention) or a parent-led home-based programme (comparison), both involving a 1-week intervention period. The primary outcome is goal attainment in the week following the intervention period (T1). Secondary outcomes include: goal attainment and participation in physical activity at 3 months postintervention (T2) and bike skills, attendance and involvement in bike riding, self-perception and functional skills at T1 and T2. Economic appraisal will involve cost-effectiveness and cost-utility analyses. Adherence of clinicians and parents to the intervention and comparison protocols will be assessed. Linear and logistic regression will be used to assess the effect of the intervention, adjusted for site as used in the randomisation process.

**Ethics and dissemination** This study was approved by the Human Research and Ethics Committees at The Royal Children's Hospital (#36209). Results will be disseminated via peer-reviewed publications and conference presentations.

**Trial registration number** NCT03003026; Pre-results.

## Strengths and limitations of this study

- ► To our knowledge, this will be the first adequately powered randomised controlled trial to evaluate the effectiveness of a novel task-specific bike skills training programme for attaining bike-specific goals in children with cerebral palsy.
- ► The range of secondary outcomes will allow for assessment of the effects of training bike skills on a range of activity and participation outcomes.
- ► Assessment of fidelity will enable evaluation of the extent to which clinicians and families adhere to the intervention and comparison group protocols.
- ► The economic appraisal will be useful for future policy and decision-making.
- ► Due to the nature of the intervention, clinicians delivering the interventions and participants will not be blind to allocation.

## INTRODUCTION

Cerebral palsy (CP) is the most common cause of childhood physical disability affecting 1 in 500 births.[1] It is a group of disorders of the development of movement and posture, causing activity limitations that are attributed to non-progressive disturbances occurring in the developing fetal or infant brain.[2] Children with CP participate less in physical and recreational activities than their typically developing peers,[3] putting them at increased risk of poor health and disease in adulthood.[4] Effective means of engaging children with CP are required to improve physical activity patterns in this population, and evidence supports training targeted towards goals of the child and their family.[5] Bike riding is a common activity for families[6] and may be an effective means of involving ambulant children with CP in physical activity that is enjoyable and meaningful to them.

The Gross Motor Function Classification System (GMFCS)[7] uses five levels (I–V) to classify children with CP according to their level of motor function. Children classified as levels I–II are independently ambulant with or without hand-held devices. Far fewer ambulant children with CP (GMFCS I–II) can ride a two-wheel bike at any given age compared with their typically developing peers, and if they do, they learn later in life. However, it is possible for children with CP at GMFCS levels

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I and II to learn to ride and the majority who do so, learn at home with their parents.[8]

Physiotherapists and occupational therapists routinely implement training to improve motor skills in children with CP. However, there is very little specific evidence to guide best practice in training of bike riding skills. The studies that do exist specific to children with CP have been conducted on stationary bikes[9–11] with no evidence to suggest this translates to riding a two-wheel bike in the community. Furthermore, the current practices of Australian physiotherapists and occupational therapists for training two-wheel bike skills in children with CP are not well understood. Importantly, there does not appear to be a standard or usual care.

The development and testing of approaches to training bike skills is required to provide clinicians and families with evidence-based guidance when working with children with CP with two-wheel bike-specific goals. Strong evidence exists for task-specific training to improve general upper limb function in this population[5 12] and gross motor skills in adults following stroke.[13] Task-specific training involves practice of context-specific tasks where the intervention focuses on the skills needed for a task(s).[14] It is informed by principles of motor learning[15] and dynamic systems theory[16] and involves a dynamic interaction between the task, the child and the environment to achieve a motor skill in a task-specific context.[17] Evidence for task-specific training to improve gross motor skills in ambulant children with CP exists,[18 19] but is currently limited by poor study methodology and intervention heterogeneity.[20] An intensive task-specific approach to training bike skills has seen promising outcomes in a group setting at the two main paediatric rehabilitation settings in Victoria, Australia demonstrated through results from a small pilot case series (n=5).[21] While this clinical evidence supports the safety and feasibility of task-specific training in bike riding in a group setting, an adequately powered study with a comparison group is required to ascertain the effectiveness of such an approach.

### Objectives

The primary objective of this study is to determine if a novel task-specific approach to training bike skills is more effective than a parent-led home programme in ambulant children with CP (GMFCS I–II) aged 6–15 years, for attaining individualised two-wheel bike-specific goals immediately following the intervention period (T1).

The secondary objectives of this study are:

1. To determine if a novel task-specific approach to training bike skills is more effective compared with a parent-led home programme in children with CP (GMFCS I–II) aged 6–15 years on:
   a. Goal attainment at 3 months following the intervention (T2);
   b. Acquiring and retaining two-wheel bike skills at T1 and T2;
   c. Functional skills at T1 and T2;
   d. Physical activity behaviour at T2;
   e. Self-perception at T1 and T2;
   f. Self-perceived bike riding competence at T1 and T2.
2. To compare attendance and involvement in bike skills training between the intervention and comparison groups during the intervention and follow-up periods.
3. To conduct an economic appraisal, involving assessment of quality of life, of the intervention compared with the comparison programme.
4. To examine clinician and parent fidelity with delivery of both group protocols.

## METHODS AND ANALYSIS

### Design

Assessor-blinded, parallel-group, randomised controlled, multicentre, superiority trial comparing a novel task-specific approach to a parent-led home programme for training bike skills. This study involves a 1-week intervention period and 3-month follow-up period (figure 1).

### Setting

The study will be conducted through the Victorian Paediatric Rehabilitation Service (VPRS: a state-wide rehabilitation service for children with rehabilitation goals including children with CP) at The Royal Children's Hospital and Monash Children's Hospital in Melbourne, Australia.

### Participants

Sixty participants will be recruited from the Victorian Cerebral Palsy Register (VCPR: a register of children with CP who were born in Victoria or receive health services in Victoria) and the VPRS. Approximately 30 children will be randomised to the intervention group and 30 children will be randomised to the comparison group (figure 1). Each participant must meet all of the inclusion criteria and none of the exclusion criteria to be enrolled in this study (box).

### Recruitment procedures

Participants will be identified and recruited through the VCPR and the VPRS at The Royal Children's Hospital and Monash Children's Hospital. The study will also be advertised on the National Health and Medical Research Council Centre of Research Excellence in Cerebral Palsy (CRE-CP) newsletter and website.

#### Victorian Cerebral Palsy Register

Within this register, it is recorded whether parents or primary care givers have consented to being contacted for research purposes. Invitations to participate in the study will be sent by VCPR staff to potentially eligible participants whose parents/primary care givers have provided consent by email or letter including a full participant information and consent form. Families will have the opportunity to contact the VCPR to request that their contact details not be passed onto the study team for follow-up and screening for eligibility which will occur by email and phone.

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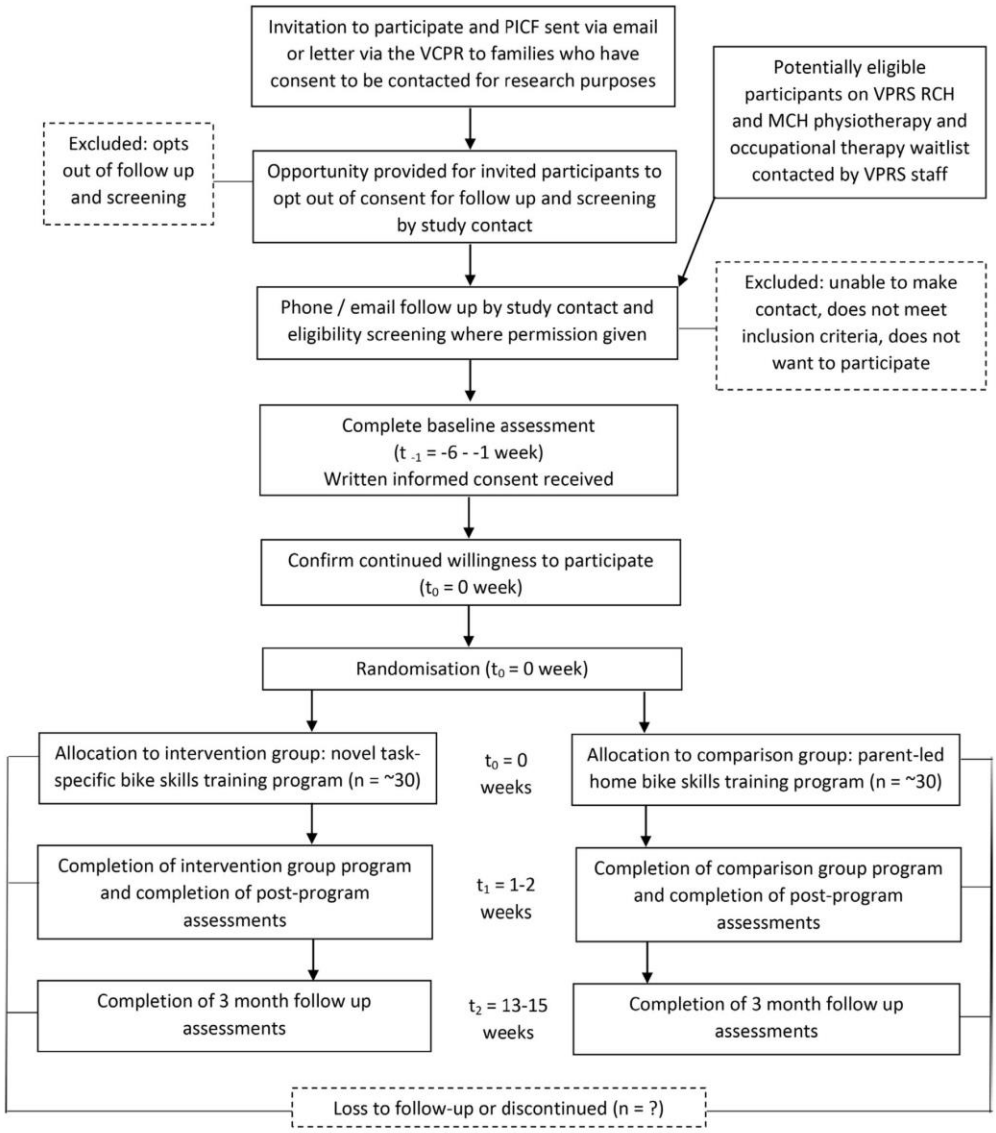



**Figure 1** Study timeline. $t_{-1}$, enrolment and baseline assessment time point; $t_0$, randomisation and allocation time point; $t_1$, first follow-up assessment time point; $t_2$, final follow-up assessment time point; MCH, Monash Children's Hospital; PICF, participant information and consent form; RCH, The Royal Children's Hospital; VCPR, Victorian Cerebral Palsy Register; VPRS, Victorian Paediatric Rehabilitation Service.

### The Victorian Paediatric Rehabilitation Service

Waitlists for services and clinics at VPRS sites at The Royal Children's Hospital and Monash Children's Hospital, will also be used to identify potentially eligible participants. A VPRS clinician will contact the parents of potentially eligible participants as per respective VPRS site physiotherapy waitlists. Potentially eligible participants who attend VPRS clinics at both hospitals during the recruitment period but are not yet on the respective VPRS physiotherapy waitlists will also be identified by VPRS clinicians. Interested families will be given the study contact's details or permission will be sought by the VPRS clinicians to pass their contact information on to the study contact for screening and follow-up.

### The Centre for Research Excellence in Cerebral Palsy website and e-newsletter

An advertisement inviting eligible families to participate in the study will be posted on a parent, clinician and researcher website for the management and treatment of CP (http://www.cre-cp.org.au) and in the website's e-newsletter during the recruitment period.

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**Box Inclusion and exclusion criteria**

**Inclusion criteria**

- Is between age 6 and 15 years at the time of randomisation.
- Has cerebral palsy as determined by the Victorian Cerebral Palsy Register (VCPR) or in writing from the child's general practitioner or paediatrician if not on the VCPR.
- Is independently ambulant without aids (Gross Motor Function Classification System I–II).
- Has goals related to improving two-wheel bike skills.
- Has a primary caregiver available to participate in the intervention.
- Has a legally acceptable representative capable of understanding the informed consent document and providing consent on the participant's behalf.
- Lives in Victoria or close to the Victorian border in New South Wales and receives health services in Victoria.
- Has access to an appropriately sized two-wheel bike and helmet.
- Has medical clearance to participate in the study from the child's general practitioner, paediatrician or paediatric specialist.
- Participant and primary caregiver able to understand English.

**Exclusion criteria**

- Has a moderate-to-severe intellectual impairment.
- Has a dual diagnosis with another developmental disability or medical condition that may impact on their ability or safety to train two-wheel bike skills.
- Had musculoskeletal surgery, or other major surgery including insertion of a baclofen pump that may affect their physical ability in the 6 months prior to randomisation.
- Had Botulinum toxin-A injections to the lower limbs and/or upper limbs in the 6 months prior to randomisation.

### Baseline study visit

Eligible participants will be enrolled in the study at the baseline (T0) assessment visit up to 6 weeks prior to the intervention period. Written informed consent will be obtained prior to performing any assessments and randomisation by the Principal Investigator or trained outcomes assessor. The following will be collected at the T0 assessment (see also supplementary appendix 1):

- Age, intellectual impairment (if any) and description of the CP including: topography, motor type, GMFCS level and Manual Ability Classification System level.
- Previous time spent practising bike skills on average per week or month since commencement of bike skills practice.
- Parent rated importance of their child attaining their goals, competence of their own bike skills and family interest in bike riding on a five-point scale.
- Family social risk as measured by a questionnaire comprised six questions regarding social status including family structure, education of primary caregiver, occupation of primary income earner, employment status of primary income earner, language spoken at home and maternal age at birth.[22]
- Goals will be set by the child, parent and outcomes assessor together using the Goal Attainment Scale (GAS).[23]
- Baseline data for secondary outcomes will be collected including: two-wheel bike skills, functional skills, physical activity behaviour, self-perception, self-perceived two-wheel bike riding competence and health-related quality of life as assessed by the measures detailed below under the 'Primary and secondary outcome measures' section.

### Randomisation and blinding

A statistician not directly involved in the study will prepare the randomisation schedule using computer-generated block randomisation with variable block sizes. Randomisation will be stratified by site. The statistician will generate opaque, numbered, sealed envelopes according to the randomisation schedule. In the week prior to the intervention period the participant will be allocated a sequential study number within the appropriate strata. Participants will then be randomised by a study investigator not involved in assessment procedures who will open the envelopes and inform participants of their allocation via phone or email. Participants who are already known to either site will be randomised within that site, otherwise families will be randomised within a site based on family preference or home location. The outcome assessors will be blind to group allocation, but it will not be possible to blind the treating clinicians or participants.

### Details of the intervention and comparison

#### The intervention: novel task-specific bike skills training programme

Participants randomised to the intervention group will participate in a novel bike skills training programme conducted over three consecutive days, with a further 4 days for practising the learnt skills at home (7-day intervention period). This approach involves seven key components:

1. Task-specific: training will be informed by the dynamic systems theory and principles of motor learning. The dynamic interaction between systems including the task, the child and the environment is considered to achieve motor skills in a task-specific context.[16] Each of these systems is considered at the stages of the motor learning process. Initially, new motor tasks are scaffolded, so that the participant will always actively complete at least part of the task. This may involve task demonstration or physical guidance. As performance improves, the task and/or environment is altered to encourage problem solving and increase the motor challenge. This may include modifying the bike (eg, seat height, location of the brakes, basic straps for hand or feet) and reducing the physical guidance in order to achieve each progression of the skill/s. Once a motor skill is acquired, variability and randomness of practice in terms of task difficulty and environmental challenge will be introduced to increase the complexity and generalisability of the skill.[24] Overall practice will be repetitive, progressive, variable and favour whole skill practice rather than part practice.[24] The amount and type of feedback from the trainer

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will be guided by participant preference, and will focus on knowledge of results or performance for each new skill,[25] for example, getting on and off the bike. Participants will use their own two-wheel bike without training wheels and helmet where possible. Cones or markers will be used as visual cues for skill practice.

2. Group-based: training will be delivered to groups of up to six participants. There is evidence to suggest group-based rehabilitation programmes improve functional skills, self-perceived performance and cost-effectiveness of treatment as much, or more than individual therapy.[26]
3. Clinician-mediated: each programme will be conducted by at least one physiotherapist and one other clinician (physiotherapist, occupational therapist or allied health assistant). There will be a minimum ratio of one clinician to three child participants in each group. All clinicians will be employed by VPRS and will undertake 6–8 hours training in the intervention protocol in the 4 months prior to delivering the intervention. The same two clinicians will lead the 3 days of each programme where possible.
4. Intensive: each programme will run for 2 hours/day over three consecutive days during 1 week of the school holiday period. This intensity is supported by motor learning literature, in particular the benefits of repetitive practice in the skill acquisition phase.[27] This intensity allows for repetitive practice,[18] including repetitive practice in the home environment following the programme and has been supported by parent evaluation of the intensive programme delivered as part of the pilot case series.[21] Breaks from physical activity will be offered at least every 30 min and families can request additional rests. Participants will also be given a home programme of one to three bike skills practice exercises following each session and encouraged to practice these up to 30 min/day during the week-long intervention period and three to five bike skills to practice when able in the 3-month follow-up period.
5. Goal-directed: evidence suggests interventions that are goal-directed improve gross motor function more than those that are not.[28] Goal setting is a key component of paediatric rehabilitation and has been well established in the literature.[29] GAS will be used as an outcome measure and as a process for setting goals related to bike skills training. Clinicians delivering the intervention will be aware of each participant's goals, which will be used to provide individualised opportunities for problem solving and drive the movements required to meet the task demands.[30]
6. Parent or caregiver involvement: at least one parent or caregiver will be required to attend each session of the programme. Parent involvement and education is recognised as a key component in family centred practice.[31] It facilitates a partnership between the clinician and parent towards achieving the child's goal. Parents will be coached by the clinician during the 3-day intervention regarding approaches to motor learning, including gradually increasing the difficulty of the task while ensuring this intersects with success. Parents will be provided verbal guidance regarding strategies and safety of practice in the home environment.[24]
7. Ecological setting: when possible the programme will be conducted in outdoor recreation or community reserves at or in close proximity to the rehabilitation service. This aligns with dynamic systems theory and task-specific training in terms of the role the environment has in promoting motor learning. Different surfaces and gradients will be available to individualise the environment based on each participant's stage of motor learning and to promote successful problem solving. All programme settings will be conducted away from roads and busy public spaces. Participants will be encouraged to practice outside of the programme in similar environments and advised to avoid practice on roads, busy bike paths or other risky environments during the intervention and follow-up periods.

### The comparison: parent-led home bike skills training programme

Current bike skills training for children with CP is not well understood. Given the lack of specific evidence, current practice is not likely to be uniform in approach, dosage or setting. While the majority of ambulant children with CP (GMFCS I–II) are currently not able to ride a two-wheel bike, many of those who can ride learnt in informal settings with their families.[8] There also is evidence to support home-based therapy programme involving parent education for goal attainment in children with CP.[5 32] Given this, it seemed appropriate that the comparison group for the intervention was a parent-led, home-based programme.

Participants randomised to the comparison group will receive written general information on training bike skills either in person or via email dependent on consenting and baseline assessment location. Families will receive this information at the start of the 1-week period of training during the school holidays. Parents will be encouraged to work with their child on two-wheel bike skills goals guided by the written information (available on request). This information involves:

1. Intensity: families will be encouraged to practice at least 30–45 min on each of the 7 days of the 1-week period.
2. Safety: families will be encouraged to practice in settings away from roads and busy public spaces. They will also be advised to perform a risk assessment of the location prior to commencing. Information on appropriate weather and adequate hydration will also be included.
3. Appropriate bike and helmet fit: information regarding fitting the bike and helmet to the child for skill development, safety and potentially useful modifications will be provided.

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A trained VPRS physiotherapist will also telephone families in the comparison group between 3 and 5 days into the training period. The purpose of this phone call will be to inquire about how the family is managing with the training programme and to offer general advice regarding practice for the remaining 2–4 days of the training period.

### Primary and secondary outcome measures

Outcomes will be measured in the week following (T1) and 3 months (12–14 weeks) following (T2) the intervention period (see online supplementary appendix 1). Outcomes will be assessed by the Principal Investigator (RT) or a physiotherapist trained in the outcomes assessment, both blinded to group allocation.

The primary outcome, goal attainment at T1, will be measured using GAS, a criterion referenced tool for individualised and collaborative goal setting between the child, family and clinician.[23 29 33] GAS is commonly used in rehabilitation for children with CP because it is valid,[33] reliable and responsive[34] in this heterogeneous population. GAS will be facilitated by the blinded outcomes assessors, trained in administering GAS. Two to three individualised and measurable two-wheel bike-specific activity or participation goals per participant will be set at the baseline visit (T0). Six potential outcomes will be specified for each goal: −3 (deterioration), −2 (equal to start), −1 (less than expected), 0 (expected), 1 (somewhat more than expected), 2 (much more than expected).[35] Children aged 8–15 years will lead the goal setting at T0 and scoring of goal attainment, while children aged 6–7 years will complete the process with their parent and assessor. The primary outcome, goal attainment, is defined as attainment of at least one goal to an expected (score of zero) or greater level. While varied interpretations of goal attainment have been used, including averaging the number of goals achieved, recent literature in rehabilitation suggests that the chosen definition reflects a clinically relevant change and allows for appropriate statistical analysis, in that it is not treated as a continuous variable.[35–37]

The secondary outcomes will be assessed as follows:

- Goal attainment at T2 measured using GAS.[29]
- Bike skills acquisition and retention measured using the subscale items related to bike skills in the mobility domain of the functional skills in the Dutch calibration of Paediatric Evaluation of Disability Inventory (PEDI-NL)[38] and the Cycling Skills Checklist[39] at T1 and T2. The PEDI is a commonly used scale to measure functional status across the domains of self-care, mobility and social function in children with disability. As part of its calibration for use in the Netherlands, a subscale was added to the mobility domain involving four levels of bike riding skill. The PEDI-NL has good content and discriminative validity and is reliable in children with disabilities.[40] The Cycling Skills Checklist is a 20-item checklist of beginner bike skills, where a score out of five is given for each skill. The maximum score for the highest level of bike skills is 100. It has not been validated in children with CP, however has been used in research with youth with Down syndrome.[41]
- Functional skills measured using the Paediatric Evaluation of Disability Inventory-computer adaptive test (PEDI-CAT)[42] at T1 and T2. The PEDI-CAT comprises a comprehensive item bank of 276 functional activities acquired throughout infancy, childhood and adolescence. The PEDI-CAT measures function in four domains: (1) daily activities; (2) mobility; (3) social/cognitive and (4) responsibility. It is valid and reliable for use in parents of children with all ages with CP. The content-balanced version of the PEDI-CAT will be used.
- Physical activity behaviour measured using a triaxial accelerometer[43] and the Physical Activity Questionnaire for Children (PAQ-C)[44] at T2. Accelerometry is a feasible, reliable and validated method of measuring activity in children and young people with CP.[45] The Activ8 will be used as it is able to distinguish cycling as a different type of physical activity from walking, running, standing and sitting.[43] The Activ8 will be worn by each participant for 7 days at T0 and at T2. PAQ-C is a valid and reliable[46] self-report 7-day recall assessment of physical activity in children aged 8–20 years.
- Overall self-perception measured with the Pictorial Scale of Perceived Competence and Social Acceptance for Young Children[47] (aged 6–7 years) or the Self-Perception Profiles for Children[48] (aged 8–13 years) and Adolescents[49] (aged 14–15 years) at T1 and T2. These self-perception scales have good validity in children without intellectual impairment.[47–49]
- Self-perceived bike riding competence measured with the bike riding item of the Pictorial Scale of Perceived Movement Skill Competence.[50] The scale from which this item is drawn has good reliability, and face and construct validity in children.[50 51]
- Attendance and involvement for participants in the intervention group during the 3-day programme as recorded by clinicians delivering the intervention group protocol. Any home-based bike skills training during the intervention period in both groups will be recorded by participants and parents each day of the intervention period and each week during the follow-up period in a participant diary. Families will also be asked to assess the involvement of the child of a five-point adjectival scale from minimally involved to very involved in the practice for each day of the 7-day intervention period.
- Quality of life measured by Child Health Utility-Nine Domains (CHU-9D)[52] at T1 and T2. The CHU-9D is a paediatric generic preference-based measure of health-related quality of life.[52] It consists of a descriptive system and a set of preference weights, giving utility values for each health state described by the descriptive system, allowing for calculation of quality-adjusted life-years for cost-utility analysis. It consists of nine domains and has been validated in

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children aged 7–17 years. Data of resources and time used to deliver the task-specific approach to training bike skills and the parent-led home programme will be collected by clinicians and parents and used for cost-effectiveness analysis.

- Fidelity assessed by examining the adherence of the clinicians and parents to the intervention and comparison group protocols. The amount of time practising bike skills will be measured by participant diaries in both groups. Clinicians will also complete attendance logs for participants in the intervention group and will document adherence to the protocol as reported by the parent on the comparison group phone call. Specific fidelity to the intervention protocol will be by video analysis. One session of the intensive programme per participant will be videoed and analysed for adherence to the protocol using the Motor Learning Strategies Rating Instrument-20 Items.[24]

Participating families will be asked to document any other therapy, health or medical interventions they receive during the study period on the participant diaries.

### Exclusion during the study

All outcome data will be attempted to be collected for all enrolled participants with the exception of those who withdraw consent.

### Treatment discontinuation

Participants in the intervention group or their parents may decide to stop the intervention at any time during the study. If a participant stops the intervention for any reason, all evaluations required for the immediate and final study visit will still be offered to the participant (unless the participant formally withdraws from the study).

## Data analysis plan

### Sample size calculation

Results of a survey conducted by the research team indicate that approximately 25% of children with CP (GMFCS I–II) had learnt to ride a two-wheel bike in the home environment led by their parents or caregivers,[8] which is likely to be the key goal of many of the study participants. Within previous studies using GAS to assess the effectiveness of similar interventions in children with CP, the proportion of goals attained or participants who have reached goal attainment has been reported between 66% and 86%.[29 35 53 54]

Using the results of previous studies, this study is powered to find an absolute difference of 50% (from 25% in the home programme/comparison group to 75% in the intervention group) in the proportion of participants who reach goal attainment following the intervention. Assuming independent observations from individuals, a sample size of 19 in each group (38 in total) would be required to identify a difference in proportions of 50% with 80% power (based on a two-sided test with a 5% level of significance). In this study, participants in the intervention group will receive the intervention in groups. It is likely that the outcomes for participants in the same group will be correlated or clustered hence the sample size has been inflated to account for this correlation. Assuming a small intracluster correlation of 0.1 between individuals within a cluster, and assuming an average cluster size of five, this equates to a design effect of 1.4, hence we will need to recruit 27 participants per arm (54 participants in total) to obtain the effective sample size of 38. Finally, we inflate the required sample size to allow for 10% loss to follow-up, hence we plan to recruit a total of 60 participants (approximately 30 per group).

### Statistical analysis

All statistical analysis will be conducted on an intention-to-treat basis where outcome data are available using STATA statistical software V.14.[55] Descriptive statistics will be used to characterise each group. Logistic regression will be used to assess the effect of providing the novel task-specific intervention compared with the parent-led home programme on the primary outcome, bike-specific goal attainment, adjusted for site as used in the randomisation process. Logistic regression will also be used to compare secondary binary outcomes between each group and linear regression will be used to compare secondary continuous outcomes between groups.

All analyses will be conducted using mixed effects models including a random effect to allow for the clustering of participants within therapy groups in the intervention arm. As a secondary analysis, all analyses will be repeated using a per-protocol analysis. In this analysis, participants in the intervention group who discontinued the intervention prior to completing the 3-day programme will be excluded from the analysis.

Participants will also be excluded from per-protocol analysis in either treatment group if any of the following protocol violations occur following randomisation and during the intervention and follow-up periods:

- Botulinum toxin-A injections to the lower or upper limbs;
- Musculoskeletal surgery or other major surgery that may affect their physical ability;
- Insertion of an intrathecal baclofen pump;
- Occupational therapy or physiotherapy related to training two-wheel bike skills other than the intervention or comparison group protocols.

The economic appraisal will be conducted from a societal perspective. Cost-consequence analysis, including cost-effectiveness analysis and cost-utility analysis, will be carried out by comparing the incremental cost with the incremental benefit. The cost-effectiveness analysis will compare the costs with the primary and secondary outcomes demonstrating significance, and the cost-utility analysis will compare the costs with the outcomes as measured by the CHU-9D.[52] The costs associated with resources and time used for each group will be assessed and compared.

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### Handling of missing data

Prior to analysis, the amount of missing data will be explored, along with a comparison of distribution of key variables in individuals with and without missing data. If there is a reasonable amount of missing data and the data summaries suggest that the data are missing at random then all analyses will be presented following multiple imputation for missing data using baseline variables as auxiliary variables. Complete case analysis will also be conducted and reported. In the case there is little missing data, a complete case analysis will form the primary analysis.

## Ethics and dissemination

The trial is registered with the US National Institutes of Health (NCT03003026) and recruitment is ongoing.

Data collected as part of this study will be entered and stored in electronic format on a REDCap secure, web-based database.[56] All other relevant electronic and paper data files will be stored securely and accessible only to study investigators. Participant confidentiality and privacy will be strictly held in trust by all study personnel.

Given the low risk nature of trial, a data monitoring committee is not required. Adverse events will be recorded from the time the participant signs the informed consent form until the end of the last study visit. Any serious adverse events occurring in a study participant will be reported to all involved ethics committees within 72hours of occurrence.

This study is being completed as part of RT's Doctor of Philosophy (PhD—physiotherapy) at the University of Melbourne. It will form a major part of her thesis. The results of this study will be submitted to peer-reviewed journals and presented to national and international conferences. Participating families will receive detailed summaries of the results of the study and a brief summary of the results will be distributed through the VCPR biannual newsletter and the CRE-CP e-newsletter/website.

## Significance

This study will contribute to the evidence base regarding the effectiveness of approaches to training bike skills in children with CP for attaining bike-specific goals. Furthermore, the range of secondary outcomes will allow for assessment of the effect of training bike skills on a range of meaningful outcomes for children and their families. The results of the economic evaluation will be used for policy and decision making.

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**Contributors** All named investigators contributed to the design of this trial protocol, to drafting and revising the manuscript and have approved this version for submission. Lead investigator RT is responsible for all aspects of study conduct with a particular focus on study oversight, recruitment, clinician training, reporting of adverse events, conducting study visits, outcome assessment, data management and statistical methods. Dr ARH, Associate Professor JLMcG and Associate Professor AJS are responsible for selected study procedures (including randomisation allocation) and study oversight. Associate Professor KJL has contributed to statistical methods and will be involved in interpretation of the results. Dr STFS will contribute to economic appraisal. RT will lead the dissemination and translation of results, with contributions from all investigators.

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**Competing interests** None declared.

**Patient consent** Not required.

**Ethics approval** This study was granted multisite approval by the Human Research and Ethics Committee at the Royal Children's Hospital (#36209).

**Provenance and peer review** Not commissioned; externally peer reviewed.

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## 5.2. Supplementary appendix: Table 1: RCT schedule of assessments

| VARIABLES | STUDY PERIOD | | | | |
|---|---|---|---|---|---|
| | **Initial Eligibility Screen** | **Baseline Assessment** | **Allocation** | **Follow up study visits** | |
| **TIME POINT**** | | **T0 = -6 to -1 weeks** | **-1 to 0 weeks** | **T1 = 1 to 2 weeks** | **T2 = 13 to 15 weeks** |
| Confirmed CP | X | X (Confirm) | | | |
| GMFCS | X | | | | |
| Age | X | | | | |
| Intellectual ability | X | | | | |
| Healthy care giver available | X | | | | |
| Live in Victoria / near border | X | | | | |
| Appropriate bike and helmet | X | | | | |
| Medical clearance | X | | | | |
| BonT-A injections or surgery (including insertion of baclofen pump) in last 6 months | X | X | | X | X |
| No other bike related therapy during intervention and follow up period | | | | X | X |
| Informed Consent | | X | | | |
| Allocation | | | X | | |
| Topography and motor type | | X | | | |
| MACS | | X | | | |
| Previous bike riding practice | | X | | | |
| Parent rated importance of bike skills goal attainment | | X | | | |
| Parent bike skills competence and interest | | X | | | |

| | | | | | |
|---|---|---|---|---|---|
| Parent social risk questionnaire | | X | | | |
| Goal attainment (GAS) | | X | | X | X |
| Two-wheel bike skills (PEDI-NL & Cycling skills checklist) | | X | | X | X |
| Functional skills (PEDI-CAT) | | X | | X | X |
| Physical activity behaviour (accelerometer & PAQ-C) | | X | | | X |
| Self-perception (SPP-YC/C/A) | | X | | X | X |
| Self-perceived bike riding competence | | X | | X | X |
| Health-related quality of life (CHU-9D) | | X | | X | X |
| Attendance and involvement in intervention group | | | | X | X |
| Practice in intervention and comparison group | | | | X | X |
| Child involvement in intervention and comparison group | | | | X | X |
| Other therapy or medical interventions | | | | X | X |
| Adverse events | | X | | X | X |

Table 5.1 notes: BonT-A (Bontulinum Toxin-A)

# CHAPTER 6: A TASK-SPECIFIC TRAINING PROGRAM IS MORE EFFECTIVE THAN A PARENT-LED HOME PROGRAM FOR TWO-WHEEL BIKE RIDING GOAL ATTAINMENT IN CHILDREN WITH CEREBRAL PALSY: A RANDOMISED CONTROLLED TRIAL

## 6.1. Preamble

This chapter contains a manuscript prepared for submission to the *Journal of Physiotherapy* which reports the primary and secondary outcomes from the RCT (protocol detailed in Chapter 5). The economic evaluation and analysis of fidelity to the interventions outlined in the protocol (pages 64 and 65) are beyond the scope of the PhD and are not reported on in this chapter. Details of the author contributions and funding sources are listed in the preface to the thesis. As the manuscript has not yet been submitted, co-author authorisation forms are not required.

Other non-author contributions to this manuscript were as follows: Consumer advisors Frances Wright and Donna Goldsmith reviewed the protocol to inform intervention design and selection of outcomes; Kim Jachno was an independent statistician who prepared the randomisation schedule and concealed opaque envelopes containing allocation information; Dr Sue Reid prepared the VCPR invitation list; Clare McKinnon was an outcomes assessor involving study enrolment and data collection; Matthew Francis, Amber Bennett, Tarli Hall, Sinead Beynon, Jenny Shing and Jinseo Yang were research selective students who contributed to data entry, Jason Mihalopoulos was a research assistant who assisted with management of accelerometer data and Diana Zannino provided statistical consultation while co-author A/Prof Katherine Lee was on parental leave.

This study is reporting according to the Consolidated Standards of Reporting Trials (CONSORT) Non-Pharmacologic Treatment guidelines. All tables and figures are included in the manuscript except for Tables 6.3, 6.5 and 6.7 which are included in section 6.8 as a supplementary appendix following the manuscript as required by the journal. The ethics and governance approval certificates and participant information and consent form

pertaining to this chapter, as well as relevant sections of the clinician training manual and information provided to parents allocated to the comparison group are included at the end of the thesis (Appendices IX - XII)

## 6.2. Abstract

**Question**

Is a task-specific training (TST) approach more effective than a parent-led home program for attaining two-wheel bike riding goals in ambulant children with cerebral palsy (CP) one week after the intervention?

**Design**

Multi-centre assessor-blind parallel superiority randomised controlled trial involving concealed allocation and intention-to-treat analysis.

**Participants**

Sixty-two ambulant children with CP aged six to fifteen years with goals related to two-wheel bike riding participated across two paediatric rehabilitation centres in Australia.

**Interventions**

Children in the intervention group participated in a TST program involving a physiotherapist-led, group-based, intensive component at a park adjacent to each site. Children in the comparison group trained in home and community settings with their parents who were provided with a recommended practice schedule and generic support via written information and a phone call. Both programs involved a one-week intervention period.

**Outcome measures**

Outcomes were assessed at one week and three months post the intervention period. The primary outcome was goal attainment at one week measured using the Goal Attainment Scale. Secondary outcomes were: bike skills, participation in bike riding, functional skills, self-perception and health related quality of life at both time points, and goal attainment and physical activity at three months.

**Results**

Children in the task-specific group had greater odds of goal attainment than children in the home program at one-week post-intervention (odds ratio (OR) =11.9, 95% confidence interval (CI) =3.1-46.7, $p<0.001$) with effects retained at three months (OR=4.1, 95% CI 1.2-14.5, $p=0.03$). There was also evidence for better outcomes following the task-specific program for outcomes related to: participation in bike riding, physical activity and self-perception, mixed effects for functional skills and little evidence of an effect in bike skills and health-related quality of life.

**Conclusion**

The task-specific approach was more effective for attaining two-wheel bike riding goals than a parent-led home program in ambulant children with CP. Secondary outcome results generated hypotheses for future research.

**Trial registration:** NCT03003026

## 6.3. Introduction

Effective means of engaging children with cerebral palsy (CP) in physical activities are required to reduce the risk of poor health outcomes in adulthood including increased risk of chronic disease (Novak et al., 2013; Peterson et al., 2015). Training directed towards goals that are meaningful and engaging for children with CP is supported by strong evidence for improved motor activities, function and self-care (Novak et al., 2013). The International Classification of Functioning, Disability and Health (ICF) provides a useful framework to consider activity limitations and participation restrictions when setting these goals and measure the impact of training beyond body structures and function (World Health Organisation, 2001)

Riding a two-wheel bike is a popular activity for Australian children (Australian Bureau of Statistics, 2012), a form of active transport and can be a goal for children with CP (Toovey, Reid, et al., 2017). It is a potential means of engaging this population in a physical activity that is enjoyable, functional, and can be done with family and friends (Raje & Saffrey, 2016). Bike riding is also a complex gross motor task (Wierda & Brookhuis, 1991) – we found a lower proportion of ambulant children with CP can ride at each age compared with their typically developing (TD) peers, and those who do ride, learned later (Toovey, Reid, et al., 2017). The reasons for this likely go beyond the motor impairments inherent to CP. Physiotherapists (PTs) and occupational therapists are routinely involved in training new motor skills (Australian Institute of Health and Welfare, 2006), however, there is a paucity of evidence to guide therapists and families in training two-wheel bike skills. While a recent systematic review suggests cycling can improve strength, balance and gross motor function in this population (Armstrong, Spencer, et al., 2019), all but one (Mohanty et al., 2015) of the nine included studies involved stationary cycling and no studies focused on two-wheel bike riding goals.

Task-specific training (TST) is a rehabilitation approach in which individuals actively practice the goal or task they wish to achieve (Bayona et al., 2005; Hubbard et al., 2009). It is grounded in motor learning and dynamic-systems theory and involves repetitive practice of context-specific tasks (Shumway-Cook & Woollacott, 2012; Thelen & Smith, 2007).

The evidence for TST for improved upper limb (UL) function in children with CP (Sakzewski, Gordon, et al., 2014; Sakzewski, Ziviani, et al., 2014) and locomotor function in adults post-stroke (French et al., 2007) is strong. Evidence for TST of gross motor skills for improving activity and participation-related outcomes does exist in ambulant children with CP but is limited by study design issues including underpowered samples (Toovey, Bernie, Harvey, McGinley, & Spittle, 2017). While no uniform approach to training bike riding in this population is apparent, TST and using goals to evaluate effectiveness are predominant practices reported by Australian therapists (Toovey, Spittle, Nicolaou, McGinley, & Harvey, 2019). This clinical uptake is promising; however, to date, no RCT has been conducted to determine the effectiveness of this approach.

Thus, the primary research question for this study was: Is a task-specific approach more effective than a parent-led home program for attaining two-wheel bike riding goals in ambulant children with CP? Our primary hypothesis was that the task-specific program would be more effective at one week post-intervention. The secondary aims were to explore the effects of the task-specific program compared with the home program on bike skills, participation in bike riding, functional skills, physical activity, self-perception and health-related quality of life.

## 6.4. Method

### 6.4.1 Study design and setting

This assessor-blinded, parallel-group, 1:1 superiority randomised controlled trial (RCT) was conducted across two sites of the Victorian Paediatric Rehabilitation Service (VPRS) - the Monash Children's Hospital (Monash) and Royal Children's Hospitals (RCH). In addition to prospective registration with the US National Institutes of Health (NCT03003026), the detailed study protocol has been published (Toovey et al., 2018). Review and advice from two parents whose children participated in pilots of the intervention informed the intervention design and outcome selection. Recruitment was open November 2016 – November 2017 and data were collected from December 2016 - April 2018.

### 6.4.2 Participants

Participants were identified and recruited through the VCPR and the Monash and RCH sites of the VPRS. Invitation to participate was also advertised via the website and e-newsletter of the nationally funded CRE-CP. Eligible participants were children with CP aged six - fifteen years old who were independently ambulant (Gross Motor Function Classifcation System [GMFCS] I-II), had goals related to two-wheel bike riding, could access a bike and helmet and lived in the state of Victoria, Australia. Children were also required to have medical clearance and a parent or caregiver able to participate, and both the children and parents were required to understand English as funding was not available for translation. Exclusion criteria were a moderate-severe intellectual impairment, dual diagnosis with another developmental disability or condition that may have impacted their ability to participate safely and BonT-A injection/s, or musculoskeletal (or other major) surgery, in the six months prior to randomisation. An outcome assessor obtained informed consent from eligible participants at the baseline assessment (T0), conducted at either study site. This was done prior to collecting participant characteristics and baseline outcome data at the T0 assessment.

### 6.4.3 Randomisation and blinding

Computer-generated block randomisation with variable block sizes (2, 4 or 6), and stratified by site, was prepared by an independent statistician. Allocation was concealed using sequentially numbered opaque envelopes. Randomisation and allocation were conducted following consent and baseline assessment by a study investigator not involved in outcome assessment in the week prior to the intervention period by opening the next available envelope in the appropriate strata. Outcome assessors were blind to allocation but blinding participating families or treating clinicians was not possible. Families and clinicians were informed of participant allocation by telephone and/or email.

### 6.4.4 Interventions

Both the experimental and comparison groups involved a one week intervention period during school holidays. The interventions have been described in detail in the protocol

paper and are summarised below (Toovey et al., 2018). The intervention clinicians were five PTs and two allied health assistants. In the three months prior to the first intervention period each clinician undertook eight hours of training led by the primary author in delivering both programs, including a training manual (See Appendix XI).

### 6.4.5 Experimental group

Children in the experimental group participated in a task-specific bike skills training program conducted for up to two hours a day over three consecutive days at an outdoor community park adjacent to each site. Thirty minutes of home practice per day was prescribed for the four remaining days. Each program was led by a PT, supported by an allied health assistant and delivered to a group of three to six children. At least one parent or caregiver attended the program for each child. This motor learning-based program involved task analysis and child-led problem solving to inform modification of the task and environment while progressing through gradually more challenging bike skills. Each child's program was individualised to goals set at baseline. While the focus was on practice of whole skills, adaptive part-practice was used to enable progression. New tasks were scaffolded so participants could achieve at least some skills, for example by removing pedals, the therapist stabilising the bike or changing the gradient/surface of the practice environment. As skill competence developed, the task and/or environment continued to be modified so the activity was challenging enough to require new learning towards attainment of bike riding goals. Clinicians were trained to enable repetitive, progressive and variable practice with amount and type of feedback guided by the participant. Clinicians coached parents on TST and safe practice in home and community environments.

### 6.4.6 Comparison group

Current bike skills training practices by therapists in Australia lack uniformity meaning "standard" or "usual" care is difficult to specify (Toovey et al., 2019). Prior to this study, it was found that many of the ambulant children with CP in Victoria who had learnt to ride, did so in informal settings with their parents (Toovey, Reid, et al., 2017). Thus, the comparison group involved a parent-led home program. Families allocated to this group received written information on two-wheel bike riding, including safety considerations,

bike and helmet fit and suggested practice location (see Appendix XII). Parents were encouraged to work towards their child's bike riding goals for 30 to 45 minutes each day of the intervention period, but no specific training methods were detailed. They also received a phone call from a PT trained in providing general support but not task-specific guidance.

### 6.4.7 Compliance measures

Clinicians completed attendance logs for the three-day program within the experimental group and documented adherence to the comparison group protocol as reported by parents on the phone call. Families in both groups completed diaries documenting time practicing bike skills in home or community environments, and any other physiotherapy or occupational therapy received, each day during the intervention period. Video analysis of clinician adherence to the experimental group protocol will be reported in a different paper as this is beyond the scope of this thesis.

### 6.4.8 Outcome measures

Outcomes were assessed at one week (T1) and three months (T2) at either study site following the intervention period by one of two PTs trained in measurement procedures and blind to group allocation.

#### 6.4.8.1. Primary outcome

The primary outcome, goal attainment at T1, was measured using the Goal Attainment Scale (GAS). The GAS is a criterion-referenced tool that is valid and reliable in children with CP (Kiresuk & Sherman, 1968). Two to three goals specific to two-wheel bike riding were set in collaboration between the child, family and assessor at baseline using proposed criteria for appraising GAS in rehabilitation research (Krasny-Pacini, Evans, Sohlberg, & Chevignard, 2016). Six levels of attainment were determined for each goal (examples in Table 6.1) (Steenbeek, 2010). The primary outcome was defined as attainment of at least one goal to an expected, or greater than expected level at T1. This binary interpretation is consistent with recommendations for using the GAS in paediatric rehabilitation as it allows for appropriate statistical analysis and represents a clinically meaningful improvement (Krasny-Pacini, Hiebel, Pauly, Godon, & Chevignard, 2013; Steenbeek, 2010).

**Table 6.1: Goal attainment scale examples**

| Score | Predicted attainment at T1 | Activity-related example | Participation-related example |
|---|---|---|---|
| -3 | Deterioration | Can ride two-wheel bike in straight line on flat grassy surface 50m with assistance at seat and handlebars from two people | Never initiates bike riding practice and no longer has goal to ride a two-wheel bike to school |
| -2 | Much less than expected (equal to start) | Can ride two-wheel bike in straight line on flat grassy surface 50m with assistance at seat and handlebars from one person | Does not initiate bike riding practice despite goal to ride a two-wheel bike to school |
| -1 | Less than expected | Can ride two-wheel bike in straight line on flat grassy surface 50m with assistance at seat from one person | Rarely initiates two-wheel bike riding practice in order to achieve goal |
| 0 | Expected | Can ride two-wheel bike in straight line on flat grassy surface 50m with close supervision | Initiates two-wheel bike riding practice in order to achieve goal some of the time |
| +1 | More than expected | Can ride two-wheel bike in straight line on flat grassy surface 50m with distant supervision | Initiates two-wheel bike riding practice in order to achieve goal most of the time |
| +2 | Much more than expected | Can ride two-wheel bike in straight line on flat grassy surface 50m independently | Initiates two-wheel bike riding practice in order to achieve goal all of the time |

#### 6.4.8.2. Secondary outcomes

Secondary outcomes included; goal attainment at T2, bike skills, participation in bike riding, functional skills, physical activity, self-perception and health-related quality of life. The GAS T-score was also calculated as a secondary interpretation of goal attainment at both timepoints (Kiresuk & Sherman, 1968). While there is conjecture around use of the T-score as it assumes that the GAS in an interval, rather than an ordinal, scale, it is commonly used in paediatric rehabilitation research so was included to enable comparision with other studies (Krasny-Pacini et al., 2013). Secondary outcomes were measured at T0, T1 and T2, except physical activity, which was assessed at T0 and T2 only. Outcome selection was informed by the ICF framework (World Health Organisation, 2001) and the family of participation-related constructs for childhood disability (Imms, Granlund, et al., 2016) in order to generate hypotheses about the impact of the interventions on bike riding (activity) competence and participation-related factors. The family of participation-related constructs model proposes that participation is more than attendance but also includes involvement in a life situation – and that there is a dynamic interaction between participation and, sense of self, activity competence and preferences, as well as the context and environment specific to each child (Imms, Granlund, et al., 2016).

Bike skills were assessed using the Cycling Skills Checklist (score out of 100 - higher scores indicating better performance) (Halayko et al., 2016) and two cycling items of the Dutch Pediatric Evaluation of Disability Inventory (PEDI-NL) (Custers, 2001): whether the child "can ride 50m independently but can have help to start" and/or "can ride 100m without help to start." Both bike skills measures were completed by the outcomes assessor. Participation in bike riding was assessed by parent-recorded time practicing (hours per day / per week) and level of involvement (scale 1 - 5 from minimally involved to very involved) in bike riding in home and/or community settings.

Functional skills were measured using the scaled scores for the four domains of the parent-completed iPad-administered PEDI-Computer Adapted Test (PEDI-CAT) (Haley, Coster, Dumas, Fragala-Pinkham, & Moed, 2012): daily activities, mobility, social/cognitive and responsibility. The content-balanced version of the PEDI-CAT was used.

Physical activity was assessed using the Physical Activity Questionnaire for Children (PAQ-C) score (Crocker, Bailey, Faulkner, Kowalski, & McGrath, 1997) and active time as measured by an Activ8™ triaxial accelerometer (Activ8, 2017). Each child and parent completed the PAC-Q together. The Activ8™ was fitted to the lateral upper third of the non-affected or preferred thigh using waterproof tape or a leg strap. Active time was defined as average time standing, walking, running and cycling in hours/day conditional upon at least 15 seconds of included minutes being recorded as active. While the Activ8™ is a valid and reliable measurement tool for physical activity in CP, it may not accurately differentiate running from cycling well (Lankhorst et al., 2019), thus combined active time was utilised. Participants were asked to wear accelerometers for seven days and data were included in the analysis if at least three days involving at least 10 hours wear time was available, consistent with published literature (Lankhorst et al., 2019; Migueles, Ulf, & Nystro, 2017; Trost, Fragala-Pinkham, Lennon, & O'Neil, 2016).

Depending on participant age, self-perception was assessed using the self-perception profile for young children (SPP-YC) (Harter & Pike, 1983), children (SPP-C) (Harter, 2012b) or adolescents (SPP-A) (Harter, 2012a). Comparable domains from each profile were combined to determine a mean domain score (1, low competence - 4, high competence) for each group. Combined domains included cognitive (cognitive (SPP-YC) and scholastic (SPP-C/A) competence), social (peer acceptance (SPP-YC) and social competence (SPP-C/A)), physical (physical (SPP-YC) and athletic competence (SPP-C/A)) while the global self-worth domain was only measured in children aged 8 years and over (SPP-C/A). Self-perceived bike riding competence was measured using the cycling item of the Pictorial Scale of Movement Skill Competence (score 1 or 2 = low competence, 3 or 4 = high competence) (Barnett, Ridgers, Zask, & Salmon, 2015; Barnett et al., 2016).

Finally, health-related quality of life was measured using the Child Health Utility Nine-Domains (CHU-9D) utility score (0 – 1) (Stevens, 2011). This score allows for calculation of quality-adjusted life years for cost-utility analysis, which will be reported alongside cost-effectiveness analysis separate to this paper. The CHU-9D and SPP were completed through interview with each child.

### 6.4.9 Adverse events

Adverse events were defined as any untoward medical occurrence related to the interventions during the study period.

### 6.4.10 Statistical analysis

The study was powered to find a between-group difference of 50% in the proportion of children who reach goal attainment. A sample of 60 children (30 in each group) was required to identify a 50% difference in proportions of goal attainment with 80% power (based on a two-sided test with a 5% level of significance) while accounting for 10% loss to follow up and clustering (groups of 3-6) within the experimental group.

The primary analysis of all outcomes was conducted by intention-to-treat (ITT) using Stata statistical software v15.1 (StataCorp, 2017). Comparisons were made using mixed effects models fitted to each outcome separately with a random effect to allow for clustering of participants within TST groups and adjusted for site as used in the randomisation process. Participants in the comparison group were analysed as a single cluster.

Logistic regression was used to assess the effect of the task-specific program compared to the parent-led home program on the primary outcome and other secondary binary outcomes. We report proportions for each outcome separately in the two treatment groups, with estimated effect sizes presented as odds ratios (ORs) with 95% CIs and p-values. Pre-planned subgroup analysis by GMFCS and sensitivity analyses for the attainment of a least one goal to more and much more than expected levels on the GAS for the primary outcome were conducted.

Linear regression was used to compare secondary continuous outcomes between groups. Summaries of the outcome data are reported separately in the two treatment groups, with estimated effect size presented as a mean difference (MD) with its 95% CI and p-values.

In accordance with the statistical analysis plan, given data summaries suggested primary outcome data was missing at random, analyses are presented following multiple imputation

using chained equations (Jakobsen, Gluud, Wetterslev, & Winkel, 2017; Sterne et al., 2009). Complete baseline variables that were correlated with the primary outcome were used as auxiliary variables and separate imputation models were run for each time point. Analysis of the complete case population is presented as the primary analysis for outcomes where convergence to the imputation model was not achieved or greater than 40% of data was missing (Jakobsen et al., 2017). All analyses were repeated using a complete case analysis and the per-protocol populations as defined in the published protocol and are presented as supplementary tables (see 6.8 Supplementary appendices).

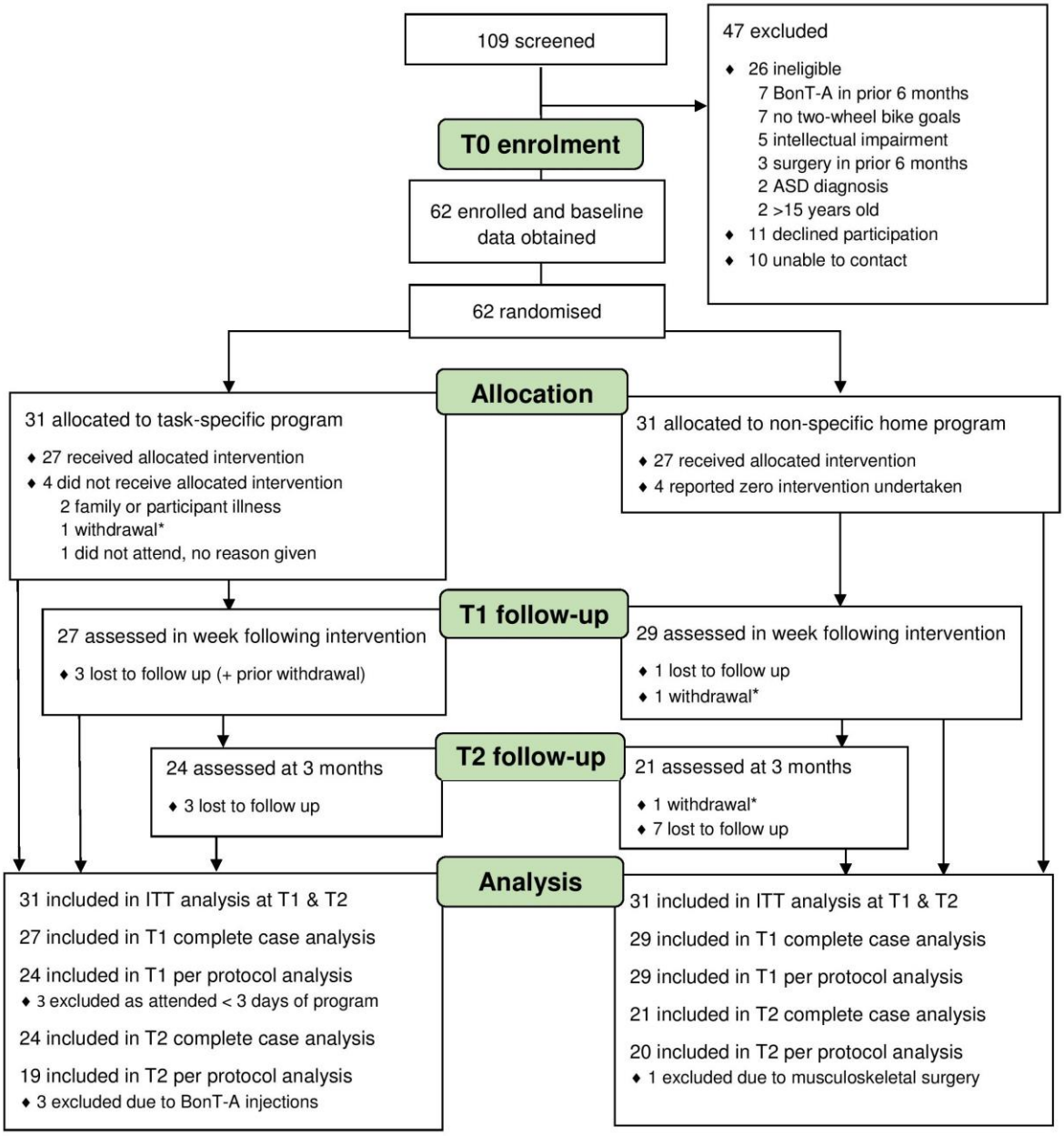



**Figure 6.1:** Design and participant flow through the study

Figure 6.1 notes: *Consent provided to use already collected data

## 6.5. Results

Sixty-two of the 109 children screened were randomly assigned to the task-specific program (n=31) or the comparison program (n=31). Figure 6.1 shows the design and flow of participants through the study. Demographics and baseline outcomes were similar between the groups except for level of bike riding skill (cycling skills score, PEDI-NL cycling items), and self-perceived bike riding competency, which were higher in the comparison group (Table 6.2). Primary outcome data were obtained from 90% (n=56) of participants and 73% (n=45) of participants were included in the 3 month-follow up assessment.

### 6.5.1 Compliance with the interventions

Eighty-eight percent (n=27) of children in either group participated in their allocated program (Figure 6.1). Twenty-four participants allocated to the task-specific program attended for all six hours of the centre-based component and completed a median of 19.4 minutes (interquartile range (IQR) 10.0-46.9) of home practice per day for the remaining four days of the intervention period. In the comparison group, the median amount of practice per day during the intervention period was 17.1 minutes (IQR 11.1-26.8, n=25) and clinicians were able to contact 26 out of 31 families by phone but were unable to contact the remaining five families. Three task-specific program participants and two comparison group participants attended other therapy during the intervention period (range = 20-120 mins). As this therapy did not involve bike riding these participants remained in the per-protocol analyses (see 8.8 supplementary appendices).

**Table 6.2: Characteristics of participants at baseline**

| | Task-specific program, n (%) n = 31 | Comparison program, n (%) n = 31 |
|---|---|---|
| Site | | |
| Monash Children's Hospital | 18 (58) | 19 (61) |
| The Royal Children's Hospital | 13 (42) | 12 (39) |
| Age in years, mean (SD) | 9.9 (2.7) | 9.1 (2.3) |
| Sex | | |
| Male | 16 (52) | 17 (55) |
| GMFCS level | | |
| I | 17 (55) | 18 (58) |
| II | 14 (45) | 13 (42) |
| MACS level | | |
| I-II | 28 (90) | 30 (97) |
| III | 3 (10) | 1 (3) |
| Distribution | | |
| Unilateral | 16 (52) | 15 (48) |
| Bilateral | 15 (48) | 16 (52) |
| Predominant motor type | | |
| Spastic | 25 (81) | 24 (78) |
| Dyskinetic | 2 (6) | 1 (3) |
| Ataxic | 1 (3) | 1 (3) |
| Hypotonic | 3 (10) | 5 (16) |
| Mild intellectual impairment (IQ > 70) | 5 (16) | 3 (10) |
| Low social risk (score 0 or 1) | 18 (58) | 22 (71) |
| Previous bike-related experience, n (%) | | |
| Two-wheel bike with training wheels | 21 (68) | 23 (74) |

| | | |
|---|---|---|
| Balance bike | 3 (10) | 4 (13) |
| Upright trike | 11 (36) | 17 (55) |
| Recumbent trike | 4 (13) | 0 (0) |
| Time spent practicing two-wheel bike skills in last month in hours, mean (SD) | 0.2 (0.3) | 0.3 (0.5) |
| Family interest in bike riding | | |
| Not at all / not really interested | 2 (6) | 2 (6) |
| Somewhat interested | 9 (29) | 6 (19) |
| Interested / really interested | 20 (65) | 23 (75) |
| Parent rated importance of child attaining two-wheel bike riding goals | | |
| Not at all / not really important | 0 (0) | 0 (0) |
| Somewhat important | 2 (6) | 2 (6) |
| Important / really important | 29 (94) | 29 (94) |
| Parent self-perceived two-wheel bike riding competence | | |
| Not at all / not really competent | 2 (6) | 2 (6) |
| Somewhat competent | 2 (6) | 5 (16) |
| Competent / really competent | 27 (88) | 24 (78) |
| Cycling skills checklist score, mean (SD) | 38.9 (11.9), n=29 | 50.0 (23.9) |
| PEDI-NL cycling items | | |
| Can ride 50m without training wheels, may have help to start | 1 (3) | 8 (26) |
| Can ride 100m without training wheels, no help* | 0 (0) | 5 (16) |
| PEDI-CAT domain scaled score, mean (SD) | | |
| Daily activities | 55.8 (3.1) | 56.4 (2.85) |
| Mobility | 64.6 (2.6) | 64.6 (2.3) |
| Social/cognitive | 67.5 (4.1), n = 28 | 67.5 (3.5) |
| Responsibility | 50.3 (5.6), n = 28 | 49.8 (4.9) |

| | | |
|---|---|---|
| Active† time per day in hours, mean (SD) | 3.3 (1.2), n = 26 | 3.7 (1.3), n = 24 |
| PAQ-C score, mean (SD) | 2.5 (0.7), n = 25 | 2.5 (0.5), n = 25 |
| SPP domain score, mean (SD) | | |
| Cognitive / scholastic competence | 2.9 (0.8) | 2.9 (0.7) |
| Peer acceptance[#] / social competence | 3.0 (0.7) | 3.0 (0.7) |
| Physical[#] / athletic competence | 2.6 (0.9) | 2.5 (0.8) |
| Global self-worth^ | 3.3 (0.5), n = 22 | 3.2 (0.5), n = 17 |
| High self-perceived bike riding competence (score 3-4) | 5 (16) | 11 (37), n = 30 |
| CHU-9D HR-QoL utility score, mean (SD) | 0.88 (0.1) | 0.92 (0.1) |

Table 6.2 notes: SD; standard deviation, IQ, Intelligence Quotient; *all had also achieved "Can ride 50m without training wheels, may have help to start"; † active defined as walking, running or cycling; [#] for children aged 6 -7; ^ for children aged 8 - 15 only.

### 6.5.2 Effects of the interventions

#### 6.5.2.1. Primary outcome: Goal attainment

Participants in the task-specific group had greater odds of goal attainment at one-week than those in the home program group (OR=11.9, 95% CI=3.1-46.7, p<0.001). This finding was consistent for both GMFCS I and II sub-groups and on sensitivity analysis of the primary outcome (Figure 6.2). There was also evidence that the task-specific program resulted in better outcomes for goal attainment at three months (OR=4.1, 95% CI=1.2-14.5, p=0.03) and for GAS T-scores at both time points (T1 MD=14.3, 95% CI=8.5-20.0, p<0.001; T2 MD=11.7, 95% CI=2.3-21.0, p=0.02). On average goal attainment (mean GAS score>50) was identified at both time points in the task-specific program but not in the home program (see Table 6.3 in section 6.8 supplementary appendices). Results were consistent across ITT with multiple imputation, complete case and per-protocol analyses (Figure 6.2, Table 6.3).

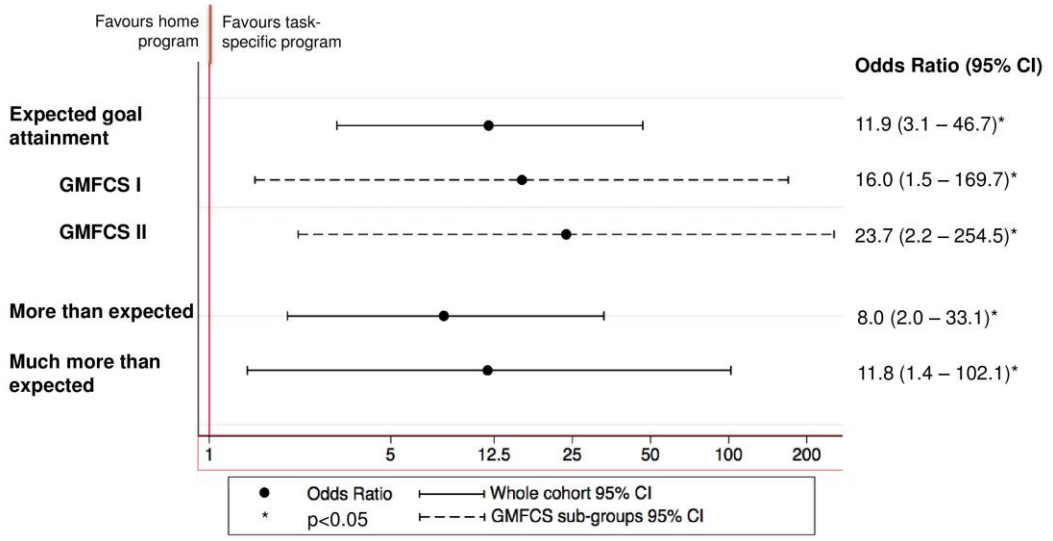



**Figure 6.2:** Primary outcome analyses – ORs and 95% CI for goal attainment at T1, task-specific program vs home program

#### 6.5.2.2. Bike riding related secondary outcomes

There were 12 participants who were newly able to ride 50m independently (with the option of help to start) following the task-specific program compared to three following the comparison program, there was little evidence of between-group differences at either time point (Table 6.4). There was greater odds of high self-perceived bike riding competence in the task-specific program compared with the home program at T1 but not at T2 (Table 6.4). For participation in bike riding, there was evidence that the task-specific group had greater involvement than the home program group at both time points but there was little difference in the amount of time practicing bike riding in home/community settings between the groups (Table 6.4). Complete case and per-protocol analyses for these outcomes are reported in Table 6.5 (see section 6.8 supplementary appendices). While effects favoured the task-specific program for involvement in bike riding at T1 following ITT analysis with multiple imputation (Table 6.4), this difference was borderline on complete case analysis and not significant on per-protocol analysis. Otherwise, results were consistent across analyses.

#### 6.5.2.3. Other secondary outcomes

There was evidence that participants in the task-specific program had better outcomes at both time points for the SPP domain of global self-worth (self-perception) and at three months only for the social/cognitive domain of the PEDI-CAT (functional skills), and physical activity when measured by the PAQ-C (Table 6.6). In contrast, there was evidence that the home program group had better outcomes for the daily activities domain of the PEDI-CAT at T1. No differences were identified for the remaining functional skills, physical activity, and self-perception or quality of life outcomes (Table 6.6). No adverse events were reported in either group. Complete case and per-protocol analyses for these outcomes are reported in Table 6.7 (see section 6.8 supplementary appendices). Results were consistent across ITT with multiple imputation, complete case and per-protocol analyses except for social/cognitive skills at T2 and physical activity at T2 when measured by the PAC-Q. In these cases, effects favoured the task-specific program on complete case analysis but not on per-protocol analysis (Table 6.7).

**Table 6.4: Bike riding related secondary outcomes for task-specific program vs home program - ITT analysis**

| Outcome | 1 week (T1) | | 3 months (T2) | |
|---|---|---|---|---|
| | **MD (95% CI)** | **p-value** | **MD (95% CI)** | **p-value** |
| Participation in bike riding | | | | |
| Average involvement in home practice | 0.4 (0.1 – 0.8) | <0.01 | 1.1 (0.6 – 1.5)^ | <0.01^ |
| Home bike skills practice (hours[#]) | 0.1 (-0.1 – 0.3)^ | 0.4^ | 0.03 (-0.7 - .1)^ | 0.5^ |
| Bike skills | | | | |
| Cycling skills checklist score | 5.1 (-3.9 – 14.2) | 0.3 | 7.7 (-2.6 - 18.0) | 0.1 |
| | **OR (95% CI)** | **p-value** | **OR (95% CI)** | **p-value** |
| PEDI-NL cycling items achieved | | | | |
| Rides 50m no training wheels, help to start | 1.5 (0.5 – 4.3) | 0.5 | 1.0 (0.4 – 3.2)^ | 1.0^ |
| Rides 100m no training wheels, no help[+] | 0.6 (0.2 – 2.1) | 0.5 | 1.1 (0.3 – 3.7)^ | 0.9^ |
| High self-perceived bike competence | 4.0 (1.1 – 15.4)^ | 0.04^ | 2.2 (0.6 - 7.8) | 0.2 |

Table 6.4 notes: [#] hours per day (T1) / per week (T2); ^complete-case analysis (see Table 5 for n for each outcome); *statistically significant; [+]all had also achieved riding 50m without training wheels but can have help to start

**Table 6.6: Other secondary outcomes for task-specific program vs home program - ITT analysis**

| Outcome | 1 week (T1) | | 3 months (T2) | |
|---|---|---|---|---|
| | MD (95% CI) | p-value | MD (95% CI) | p-value |
| Functional skills | | | | |
| PEDI-CAT scaled score | | | | |
| Daily activities | -0.6 (-1.9 – 0.8)^ | 0.4^ | -1.7 (-2.6 to -0.7)^ | <0.01^ |
| Mobility | 0.7 (-0.2 – 1.6)^ | 0.1^ | 0.05 (-0.8 – 0.8)^ | 0.9^ |
| Social / cognitive | 0.5 (-0.7 – 1.7)^ | 0.4^ | 1.4 (0.1 – 2.6)^ | 0.04^ |
| Responsibility | -0.4 (-2.2 – 1.3)^ | 0.6^ | -0.8 (-3.3 – 1.8)^ | 0.5^ |
| Physical activity | | | | |
| Active† time per day (hours) | - | - | 0.3 (-0.6 – 1.2)^ | 0.6^ |
| PAQ-C score | - | - | 0.3 (0.02 – 0.5)^ | 0.04^ |
| Self-perception | | | | |
| SPP domain score | | | | |
| Cognitive# / scholastic competence | 0.1 (-0.2 – 0.4) | 0.4 | -0.01 (-0.4 – 0.3)^ | 1.0^ |
| Peer acceptance# / social competence | 0.1 (-0.3 – 0.5) | 0.7 | 0.1 (-0.2 – 0.4) | 0.6 |
| Physical# / athletic competence | 0.05 (-0.5 – 0.6) | 0.9 | 0.1 (-0.1 – 0.3)^ | 0.4^ |
| Global self-worth~ | 0.2 (0.1 – 0.3)^ | 0.01^ | 0.2 (0.1 – 0.3)^ | <0.01^ |
| Health-related quality of life | | | | |
| CHU-9D utility score | 0.02 (0.01 – 0.04)^ | 0.2^ | -0.01 (-0.04 – 0.04)^ | 0.8^ |

Table 6.6 notes: ^complete-case analysis; †active defined as walking, running or cycling; #for children aged 6 -7; ~for children aged 8 – 15 only;

## 6.6. Discussion

This is the first RCT to focus on two-wheel bike riding in children with CP. We found that a novel task-specific program was more effective than a parent-led home program for the attainment of two-wheel bike riding goals in ambulant children with CP at one week, supporting our hypothesis. Greater odds of goal attainment were retained at three months. The rigorous study design involved an adequate sample size, concealed allocation, blinded outcome assessment and use of multiple imputation to handle missing data. Positive effects favouring the task-specific program were consistent across pre-determined primary, sub-group and sensitivity analyses of goal attainment (Figure 6.2).

There was also some evidence that the task-specific intervention may benefit children beyond attainment of their bike-riding goals, generating several hypotheses for further research. Positive effects on self-perceived bike riding competency, self-worth and social/cognitive skills align with the child-led problem-solving approach and group-based nature of the task-specific program. Conversely, effects favouring the home program were identified for daily activities, which is consistent with other studies in this population where home programs have been shown to be effective for this outcome (Novak, Cusick, & Lannin, 2009; Novak, Cusick, & Lowe, 2007). In saying this, improvements in functional skills beyond the target of the intervention (bike riding) were not expected and it is conceivable that this result is spurious or by chance given the number of analyses conducted related to functional skills. There were some inconsistencies in the results for physical activity outcomes which were not surprising given the recall bias inherent to self-report measures compared to an accelerometer (Trost, 2007). Still, the positive effect of the task-specific program for family-reported physical activity warrants further investigation. Although many participant goals targeted bike skills (see example, Table 6.1), and large within-group improvements in bike skills were found in the task-specific program (Tables 6.2, 6.5), it was not unexpected that no between-group differences in this outcome were found. This is likely to be because of the higher level of bike skills in the comparison group at baseline as well as the fact that analyses were pre-specified to be adjusted for site only. Overall, secondary outcome results should be interpreted with caution given the lack of evidence

related to what constitutes a clinically meangiful change in children with CP for the outcome measures used.

This trial is not without limitations. Several secondary outcomes either did not fit or could not be included in our multiple imputation model as over 40% of data was missing meaning complete case analysis formed the primary analysis for these outcomes (marked ^ in results tables) (Jakobsen et al., 2017; Sterne et al., 2009). Though missing data adds imprecision, it is unlikely this issue would strongly bias inferences made as we generally had consistent findings for outcomes where both imputed and complete data were available (Tables 6.3-6.7). In saying this, there is noticable imprecision associated with some goal attainment results on complete case and per-protocol analyses (Table 6.3) likely due to missing data or outliers. While this limits interpretation of these secondary analyses, it also provides further justification for using multiple imputation to account for missing data within the primary analyses. While attempts were made to dose-match training time between the two group, the home program group was, on average, unable to achieve the recommended practice time of 30 - 45 minutes per day during the intervention period. This raises a question around the extent to which effects were mediated by training dose rather than just the combined components of the task-specific program (Toovey et al., 2018). However, it may also indicate that relatively independent practice of a new and complex skill is challenging for many families in contrast to the supportive and specialised task-specific program environment. In any case, this study aimed to address whether the task-specific program was more effective than a pragmatic comparison – it did not seek to determine which program components were the active ingredients.

At up to eight hours high-density training over a week, the experimental program was low dose compared to other motor learning-based approaches that have been used in children with CP such as Hand-Arm Bilateral Intensive Therapy Including Lower Extremity (HABIT-ILE), which involves 90 hours over 10 days (Bleyenheuft et al., 2015, 2017). While HABIT-ILE addresses participant goals across a range of activities, the current task-specific program focused specifically on two-wheel bike skills. The positive effects found in our study suggest that short bursts of high-density training can be effective for children with goals related to specific complex motor skills. This is consistent with evidence of benefit from bike skills programs delivered in a week or less

in other populations including children with Trisomy 21 and ASD (MacDonald et al., 2010; Ulrich et al., 2011).

Good participant adherence, successful embedding across two clinical sites and a lack of adverse events indicate that the task-specific program was clinically feasible and safe. However, when considering clinical implications of RCT findings, care needs to be taken with interpreting mean group differences, especially in heterogeneous populations like CP (Damiano, 2014). A specialised program may not be necessary for all – we found some participants did achieve their goals at home with their parents, so next steps could involve exploring best responders in each group. This study was also limited to three months follow up and to children classified GMFCS I-II. Future work should explore longer term involvement in community cycling and enablers of this outcome, while an in-progress trial testing the efficacy of functional electrical stimulation and adapted cycling in children classified GMFCS II-IV may help address the knowledge gap across the spectrum of CP (Armstrong, Boyd, et al., 2019).

In the context of the focus on goals in paediatric rehabilitation (Oien et al., 2010) and the recently introduced goal-directed disability insurance funding model in Australia (National Disability Insurance Agency, 2016), the findings of the current study have the potential to translate into clinical practice. However, to optimise knowledge translation more broadly than health and disability – it will be important to also direct efforts across settings and sectors including to schools and sport associations. Analysis of the economic appraisal and fidelity study embedded within our protocol (Toovey et al., 2018), though beyond the scope of this paper, will also provide further useful information to guide translation into policy and practice.

In summary, this study has shown that a task-specific program is more effective than a parent-led home program for attaining two-wheel bike riding goals in ambulant children with CP in the short term. Furthermore, the task-specific program may lead to additional short and medium-term activity and participation-related benefits. Our findings provide some specific evidence-based guidance to clinicians working with children with CP who have bike riding goals. This is important in the context of rehabilitation increasingly being directed towards the activity and participation goals of the individual. Future work should explore longer-term participation in cycling and identify factors that

support goal attainment so interventions can be targeted to individuals and findings can be translated across settings.

## 6.7. Additional information for submission

**What was already known on this topic**: Many ambulant children with CP have two-wheel bike riding goals yet little guidance was available to clinicians on how best to train bike skills. Evidence supports TST for improving UL function and gross motor skills in this population, but this approach has not been specifically tested for bike riding goals.

**What this study adds:** A TST program was more effective than a parent-led home program for attaining two-wheel bike riding goals in ambulant children with CP at one week post-intervention. Positive effects were also observed in secondary outcomes related to retention of goal attainment, participation in bike riding, self-perception, functional skills and physical activity have generated hypothesesfor future research.

**Ethics Approval:** The Royal Children's Hospital Melbourne provided multisite approval for this study (36209A). Written informed consent was provided by a parent or guardian for all participants prior to data collection starting.

## 6.8. Supplementary appendices: Tables 6.3, 6.5 & 6.7

**Table 6.3: Goal attainment outcomes for task-specific program vs home program - complete case and per-protocol populations**

| Outcome | 1 week (T1) | | | | | | | | 3 months (T2) | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Complete case analysis | | | | Per-protocol analysis | | | | Complete case analysis | | | | Per-protocol analysis | | | |
| | TSP n = 27 | Comp n = 29 | | | TSP n = 24 | Comp n = 29 | | | TSP n = 24 | Comp n = 21 | | | TSP n = 19 | Comp n = 20 | | |
| | n (%) | n (%) | OR (95% CI) | p-value | n (%) | n (%) | OR (95% CI) | p-value | n (%) | n (%) | OR (95% CI) | p-value | n (%) | n (%) | OR (95% CI) | p-value |
| Goal attainment | 24 (89) | 9 (31) | 17.8 (4.2 – 75.1) | <0.01 | 23 (96) | 9 (31) | 51.6 (6.0 – 446.3) | <0.01 | 20 (83) | 10 (48) | 4.2 (1.1 – 15.6) | 0.03 | 16 (84) | 10 (50) | 5.4 (1.2 – 24.7) | 0.03 |
| *Subgroup analysis – achieved expected goal attainment by GMFCS* | | | | | | | | | | | | | | | | |
| GMFCS I | n = 16 15 (94) | n = 16 7 (44) | 23.7 (2.2 – 252.7) | 0.01 | n = 14 14 (100) | n = 16 4 (44) | TSP predicts success perfectly | - | - | - | - | - | | | - | - |
| GMFCS II | n = 11 9 (81) | n = 13 2 (16) | 23.7 (2.2 – 254.5) | 0.01 | n = 10 9 (90) | n = 13 2 (15) | 46.8 (3.1 – 702.9) | <0.01 | - | - | - | - | | | - | - |

| | | | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| *Sensitivity analysis* | | | | | | | | | | | | | | | | |
| Somewhat more than expected | 13 (48) | 3 (10) | 8.0 (2.0 – 33.1) | <0.01 | 13 (54) | 3 (10) | 10.3 (2.4 – 43.6) | <0.01 | - | - | - | - | | | - | - |
| Much more than expected | 8 (30) | 1 (3) | 11.8 (1.4 – 102.1) | 0.03 | 8 (33) | 1 (3) | 14.0 (1.6 – 122.8) | 0.02 | - | - | - | - | | | - | - |

| | **Mean (SD)** | **Mean (SD)** | **MD (95% CI)** | **p-value** | **Mean (SD)** | **Mean (SD)** | **MD (95% CI)** | **p-value** | **Mean (SD)** | **Mean (SD)** | **MD (95% CI)** | **p-value** | **Mean (SD)** | **Mean (SD)** | **MD (95% CI)** | **p-value** |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| GAS T-score | 50.5 (13.1) | 35.2 (11.4) | 15.4 (10.3 – 20.5) | <0.01 | 52.9 (11.6) | 35.2 (11.4) | 17.7 (13.2 – 22.2) | <0.01 | 54.3 (18.1) | 41.6 (15.2) | 13.7 (5.7 – 21.8) | <0.01 | 56.9 (15.7) | 41.9 (15.5) | 14.6 (8.0 – 23.1) | <0.01 |

Table 6.3 notes: TSP, Task-specific program; Comp, Comparison program;

**Table 6.5: Bike riding related secondary outcomes for task-specific program vs home program – complete case and per-protocol populations**

| Outcome | 1 week (T1) | | | | | | | | 3 months (T2) | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Complete case analysis | | | | Per-protocol analysis | | | | Complete case analysis | | | | Per-protocol analysis | | | |
| | TSP | Comp | | | TSP | Comp | | | TSP | Comp | | | TSP | Comp | | |
| | Mean (SD) | Mean (SD) | MD (95% CI) | p-value | Mean (SD) | Mean (SD) | MD (95% CI) | p-value | Mean (SD) | Mean (SD) | MD (95% CI) | p-value | Mean (SD) | Mean (SD) | MD (95% CI) | p-value |
| Participation in bike riding | | | | | | | | | | | | | | | | |
| Average involvement in home practice | n = 24<br>4.0 (1.1) | n = 23<br>3.6 (1.6) | 0.4<br>(-0.05 - 0.7) | 0.09 | n = 21<br>4.0<br>(0.9) | n = 19<br>3.7<br>(1.5) | 0.3<br>(-0.1 to 0.8) | 0.2 | n = 19<br>4.1 (1.1) | n = 16<br>3.0 (1.4) | 1.1<br>(0.6 – 1.5) | <0.01 | n = 16<br>4.0<br>(1.2) | n = 15<br>3.1<br>(1.5) | 1.0<br>(0.4 – 1.5) | <0.01 |
| Home bike skills practice (hours#) | n = 24<br>0.4 (0.7) | n = 25<br>0.3 (0.2) | 0.1 (-0.1 – 0.3) | 0.4 | n = 22<br>0.5<br>(0.7) | n = 25<br>0.3<br>(0.8) | 0.1<br>(-0.7 - 0.3) | 0.2 | n = 20<br>0.4<br>(0.3) | n = 18<br>0.4<br>(0.4) | 0.03<br>(-0.7 to -0.1) | 0.5 | n = 16<br>0.4<br>(0.2) | n = 17<br>0.4<br>(0.4) | 0.0<br>(-0.1 - 0.2) | 0.6 |
| Bike skills | | | | | | | | | | | | | | | | |
| Cycling skills checklist score | n = 26<br>67.3<br>(16.9) | n = 26<br>60.5<br>(24.6) | 6.8<br>(-0.9 - 14.5) | 0.09 | n = 24<br>68.1<br>(15.5) | n = 26<br>60.5<br>(24.6) | 7.7<br>(-0.3 - 15.7) | 0.06 | n = 23<br>72.0<br>(20.6) | n = 20<br>64.3<br>(27.4) | 6.9<br>(-0.5 - 14.3) | 0.07 | n = 18<br>72.7<br>(19.7) | n = 19<br>65.2<br>(27.8) | 6.8<br>(-2.0 - 15.7) | 0.13 |

| | n (%) | n (%) | OR (95% CI) | p-value | n (%) | n (%) | OR (95% CI) | p-value | n (%) | n (%) | OR (95% CI) | p-value | n (%) | n (%) | OR (95% CI) | p-value |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| PEDI-NL cycling items achieved | n = 27 | n = 28 | | | n = 24 | n = 28 | | | n = 24 | n = 22 | | | n = 18 | n = 20 | | |
| Rides 50m no training wheels, help to start | 13 (48) | 11 (39) | 1.5 (0.5 – 4.3) | 0.5 | 13 (54) | 11 (39) | 1.9 (0.6 – 5.8) | 0.3 | 12 (50) | 11 (50) | 1.0 (0.4 – 3.2) | 1.0 | 9 (50) | 11 (55) | 0.7 (0.2 – 2.6) | 0.6 |
| Rides 100m no training wheels, no help^ | 6 (22) | 8 (29) | 0.7 (0.2 – 2.5) | 0.6 | 6 (25) | 8(29) | 0.9 (0.2 – 3.1) | 0.8 | 9 (39) | 8 (36) | 1.1 (0.3 – 3.7) | 0.9 | 7 (39) | 8 (40) | 0.9 (0.2 – 3.5) | 0.9 |
| High self-perceived bike competence | 23 (85) | 16 (60) | 4.0 (1.1 – 15.4) | 0.04 | 21 (88) | n = 27 16 (60) | 5.1 (1.1 – 22.3) | 0.03 | 19 (80) | 13 (60) | 2.6 (0.7 – 9.5) | 0.16 | n = 19 16 (84) | 13 (65) | 2.8 (0.6 – 13.1) | 0.2 |

Table 6.5 notes: # hours per day (T1) / per week (T2); ^all had also achieved riding 50m without training wheels but can have help to start.

**Table 6.7: Bike riding related secondary outcomes for task-specific program vs home program – complete case and per-protocol populations**

| Outcome | 1 week (T1) | | | | | | | | 3 months (T2) | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| | Complete case analysis | | | | Per-protocol analysis | | | | Complete case analysis | | | | Per-protocol analysis | | | |
| | TSP | Comp | | | TSP | Comp | | | TSP | Comp | | | TSP | Comp | | |
| | Mean (SD) | Mean (SD) | MD (95% CI) | p-value | Mean (SD) | Mean (SD) | MD (95% CI) | p-value | Mean (SD) | Mean (SD) | MD (95% CI) | p-value | Mean (SD) | Mean (SD) | MD (95% CI) | p-value |
| Functional skills | | | | | | | | | | | | | | | | |
| PEDI-CAT scaled scores | n = 26 | n = 29 | | | n = 24 | n = 29 | | | n = 24 | n = 21 | | | n = 19 | n = 20 | | |
| Daily activities | 56.3 (4.2) | 56.8 (4.1) | -0.6 (-1.9 - 0.8) | 0.4 | 56.2 (4.4) | 56.8 (4.1) | -0.7 (-2.2 - 0.9) | 0.4 | 56.5 (3.0) | 58.0 (4.1) | -1.7 (-2.6 to -0.7) | <0.01 | 56.7 (3.1) | 58.1 (4.1) | -1.6 (-2.9 to -0.2) | 0.03 |
| Mobility | 65.5 (3.0) | 64.8 (3.1) | 0.7 (-0.2 - 1.6) | 0.1 | 65.6 (3.1) | 64.8 (3.1) | 0.7 (-0.2 - 1.6) | 0.1 | 65.5 (2.7) | 65.3 (3.4) | 0.05 (-0.8 - 0.8) | 0.9 | 65.7 (2.4) | 65.3 (3.5) | 0.4 (-0.8 - 1.6) | 0.5 |
| Social / cognitive | 67.9 (4.3) | n = 28 67.4 (4.5) | 0.5 (-0.7 - 1.7) | 0.4 | 67.8 (4.5) | n = 28 67.4 (4.5) | 0.3 (-0.9 - 1.6) | 0.6 | n = 23 68.4 (3.7) | n = 20 67.1 (4.9) | 1.4 (0.1 – 2.6) | 0.04 | n = 18 67.9 (4.1) | n = 19 67.3 (5.0) | 0.7 (-1.0 - 2.5) | 0.4 |

| | | | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Responsibility | 60.5 (7.9) | n = 27 50.9 (4.9) | -0.4 (-2.2 - 1.3) | 0.6 | 50.0 (7.9) | n = 27 50.9 (4.9) | -1.0 (-3.7 - 1.6) | 0.4 | n = 23 50.6 (6.2) | n = 19 51.4 (4.3) | -0.8 (-3.3 - 1.8) | 0.5 | n = 18 50.3 (6.9) | n = 18 51.6 (4.4) | -1.5 (-5.3 - 2.2) | 0.4 |
| Physical Activity | | | | | | | | | | | | | | | | |
| Active[†] time per day (hours) | - | - | - | - | - | - | - | - | n = 16 3.1 (1.4) | n = 18 2.9 (0.7) | 0.3 (-0.6 - 1.2) | 0.6 | n = 13 3.3 (1.4) | n = 17 3.0 (0.6) | 0.5 (-0.5 - 1.5) | 0.3 |
| PAQ-C score | - | - | - | - | - | - | - | - | n = 20 2.3 (0.6) | n = 15 2.1 (0.5) | 0.3 (0.02 - 0.5) | 0.04 | n = 16 2.4 (0.6) | n = 14 2.2 (0.4) | 0.3 (-0.04 - 0.6) | 0.09 |
| Self-Perception | | | | | | | | | | | | | | | | |
| SPP domain scores | n = 27 | n = 27 | | | n = 24 | n = 27 | | | n = 24 | n = 22 | | | n = 19 | n = 20 | | |
| Cognitive[#] / scholastic competence | 3.1 (0.6) | 3.0 (0.6) | 0.2 (-0.2 to 0.3) | 0.1 | 3.1 (0.6) | 3.0 (0.6) | 0.1 (-0.1 - 0.4) | 0.2 | 3.1 (0.6) | 3.1 (0.6) | 0.01 (-0.4 - 0.3) | 1.0 | 3.1 (0.6) | 3.1 (0.6) | -0.1 (-0.4 - 0.3) | 0.7 |
| Peer acceptance[#] / social competence | 3.1 (0.8) | 2.9 (0.7) | 0.2 (-0.1 to 0.4) | 0.1 | 3.1 (0.7) | 2.8 (0.7) | 0.3 (-0.1 - 0.6) | 0.1 | 3.2 (0.6) | 3.0 (0.7) | 0.2 (-0.1 - 0.4) | 0.2 | 3.2 (0.6) | 3.0 (0.7) | 0.2 (-0.1 - 0.5) | 0.2 |

| | | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Physical[#] / athletic competence | 2.7 (0.9) | 2.6 (0.8) | 0.1 (-0.3 - 0.5) | 0.6 | 2.7 (0.9) | 2.6 (0.8) | 0.1 (-0.3 - 0.4) | 0.7 | 2.8 (0.7) | 2.7 (0.8) | 0.1 (-0.1 - 0.3) | 0.4 | 2.8 (0.8) | 2.8 (0.8) | -0.05 (-0.3 - 0.2) | 0.7 |
| Global self-worth[~] | n = 18 3.6 (0.4) | n = 15 3.4 (0.6) | 0.2 (0.1 – 0.3) | <0.01 | n = 15 3.5 (0.5) | n = 15 3.4 (0.6) | 0.2 (0.02 – 0.3) | 0.03 | n = 16 3.6 (0.3) | n = 13 3.4 (0.6) | 0.2 (0.1 – 0.3) | <0.01 | n = 11 3.6 (0.3) | n = 12 3.4 (0.7) | 0.2 (0.03 – 0.3) | 0.02 |
| HRQoL | | | | | | | | | | | | | | | | |
| CHU-9D utility score | n = 26 0.9 (0.1) | N = 26 0.9 (0.1) | 0.01 (-0.01 to 0.04) | 0.2 | n = 23 0.9 (0.1) | n = 26 0.9 (0.1) | 0.01 (-0.01 - 0.04) | 0.4 | n = 24 0.9 (0.1) | n = 22 0.9 (0.1) | -0.01 (-0.04 - 0.04) | 0.8 | n = 19 0.9 (0.1) | n = 20 0.9 (0.05) | -0.02 (-0.06 - 0.02) | 0.3 |

Table 6.7 notes:[†]active defined as walking, running or cycling; [#]for children aged 6 -7;[~]for children aged 8 - 15 only;

# CHAPTER 7: GRAND DISCUSSION

*Summary, synthesis and significance of findings, implications, strengths and limitations, future directions, and conclusions*

## 7.1. Summary of findings from each study

The overall aim of this thesis was to develop and test the effectiveness of a task-specific approach to training two-wheel bike skills in ambulant children with CP. Findings from the systematic review of task-specific gross motor skills training (Chapter 3) and practice survey of therapists (Chapter 4) informed the development of the major study – an RCT (Chapters 5 and 6). This trial found that a TST approach was more effective than a parent-led home program for attaining bike-riding goals in this population.

The systematic review of 13 studies identified that low to moderate quality evidence exists for task-specific gross motor skills training to improve activity and participation outcomes in ambulant school-aged children with CP. This evidence supported task-specific approaches for improving performance of specific gross motor skills, functional skills and participation-related outcomes but not for general gross motor function. However, clear recommendations were limited by poor study methodology and heterogeneity in interventions, and there was minimal focus on participation outcomes. Findings of this systematic evaluation indicated that a task-specific approach had the potential to be effective for training bike skills and that any future study testing effectiveness should be rigorously designed and assess the impact on both activity and participation outcomes.

The survey of 96 PTs and OTs from a range of settings across Australia found that goal-directed and activity-focussed approaches to assessment, intervention and evaluation appear common in current two-wheel bike skills training practice for children with CP. However, the variability in intervention characteristics reflects an overall lack of uniform practice. It was identified that, when training bike skills, the focus of eligibility and assessment remains on body structures and functions-related factors and that personal and environmental factors are less frequently considered across each area of practice. Further, besides goal-based tools, published measures to assess and evaluate

activity and participation outcomes related to two-wheel bike riding are rarely used. This study provided valuable information to guide the design of the RCT, particularly related to the interventions being tested and selection of outcome measures.

Sixty-two ambulant children with CP participated in the RCT; the first study to evaluate the effectiveness of a task-specific approach for two-wheel bike skills training in this population. This study found that the TST program was more effective than the comparison, a parent-led home program, for the primary outcome of goal attainment at one-week post-intervention, and that this effect was retained at three months. Results also favoured the task-specific program for some components of the following secondary outcomes: participation in bike riding, physical activity and self-perception - while mixed effects were found for functional skills (favoured the task-specific program for social/cognitive skills but favoured the home program for daily activity skills, both at T2). Little difference between groups was identified for bike skills and health-related quality of life.

## 7.2. Synthesis of findings across the studies

The ICF (World Health Organisation, 2001) was used to guide selection of outcomes in the systematic review and the RCT, and as a framework for analysis and reporting in the practice survey. It can also be used as a tool to synthesise key findings across the studies. As demonstrated in Figure 7.1, this thesis adds new knowledge to each domain of the ICF.

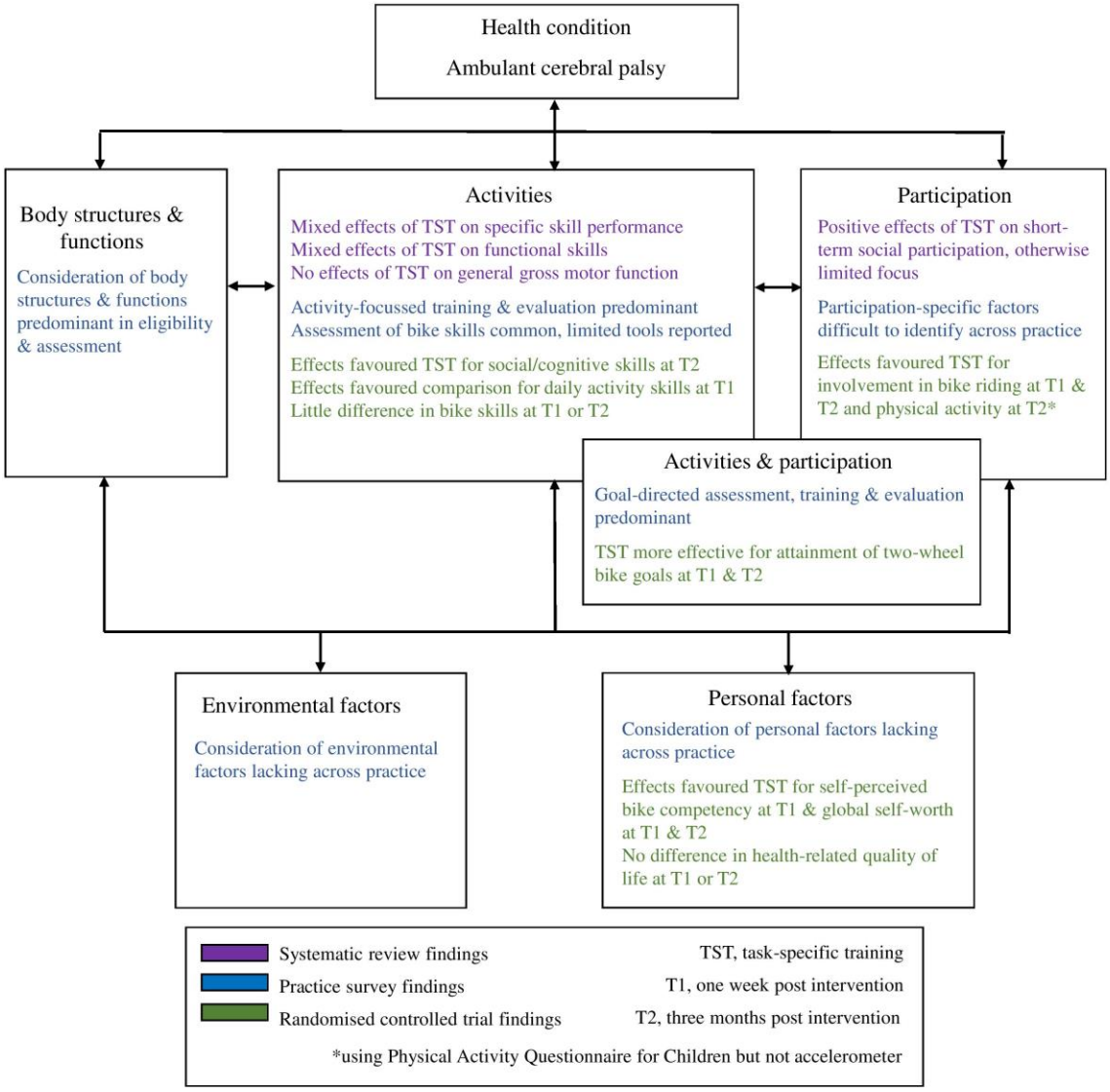



**Figure 7.1** Synthesis of findings within the ICF framework (World Health Organisation, 2001)

Despite the shift towards activity and participation-focused interventions and outcome measurement in contemporary CP literature (Novak et al., 2013), it is apparent that consideration of body structures and functions is a predominant current practice of therapists for determining whether it is appropriate to undertake bike skills training and when conducting an initial assessment. This may be, in part, due to the lack of recognised and reliable tools available that assess bike skill level and participation in bike riding specifically.

Given bike-riding is a physical activity, and that each study was designed with a focus on outcomes that are meaningful for children and families (Damiano, 2006; Imms, Granlund, et al., 2016; Rosenbaum & Gorter, 2012; Vargus-Adams & Martin, 2009), it is not surprising that key thesis findings are largely related to activity and participation. Further, it is not unexpected that current training and evaluation practices of therapists focus on the activity domain. Interestingly, the RCT findings of mixed effects of task-specific bike skills training on functional skills, including social/cognitive and daily activities, reflect the mixed effects of task-specific gross motor skills training found for this outcome in the systematic review.

Attainment of activity and/or participation-related bike riding goals was chosen as the primary outcome of the RCT, not just in the absence of bike skills-specific outcome measures validated in CP, but also because of the predominance of goal-directed practice identified in the therapist survey. This alignment reinforces the clinical relevance of the primary finding of the trial – that a task-specific approach is more effective for goal attainment. Although it was specified that goals set in the RCT could be activity and/or participation-focused, it was difficult to determine if goals, or other outcome measures, reported within the practice survey captured participation-related factors. This may be due to the emerging nature of participation measurement in childhood disability research and clinical practice (Imms, Granlund, et al., 2016). In any case, the positive effects of TST on the additional participation outcomes of involvement in bike riding and self-reported physical activity found in the RCT are congruent with the positive effects of TST on participation found in the systematic review. Both findings demonstrate that activity-level training can influence participation outcomes.

Finally, while there was a lack of consideration of environmental and personal factors across practice areas found in the therapist survey, the trial demonstrated the positive effects of task-specific bike skills training on personal factors including self-perceived bike riding competency and global-self-worth.

Although the ICF has proven a useful framework for this thesis, it is also important to note that it is not without limitations. The ICF has been critiqued in the field of childhood disability as it inherently involves a comparision to "normal." This is particularly notable alongside a pure Social Model of Disability whereby disability is considered to be socially constructed and the result of the interaction between the person with impairment and their social environment, as opposed to the person with impairment themselves (Hemmingson & Jonsson, 2005).

## 7.3. Significance of findings in context of previous work

While each of the studies within this thesis contributes new knowledge to the field of CP research, the focus of the practice survey and RCT on two-wheel bike riding as an intervention is particularly novel. While the attention of interventional research in children with CP has shifted from body structures and functions to activities over the last two decades (Novak et al., 2013; World Health Organisation, 2001), physiotherapeutic interventions targeting activity level outcomes in this population have largely focussed on general gross motor skills whereby walking performance is the most complex level of mobility evaluated (Martin, Baker, & Harvey, 2010; Novak et al., 2013). Findings from the systematic review of TST for gross motor skills within this thesis demonstrate this case in point. However, increasing recognition of goal-directed rehabilitation practice provides impetus to develop and test interventions beyond walking, as has been done in this thesis regarding two-wheel bike riding.

Prior to this thesis, it was known that ambulant children with CP could learn to ride a two-wheel bike. Howver; a significantly lower proportion rode at each age compared to their TD peers, and those who did ride learned to do so later (Toovey, Reid, et al., 2017). However, while several low-quality studies exploring bike skills training in children with ASD (MacDonald et al., 2010), Trisomy 21 (Halayko et al., 2016; Ulrich et al., 2011) or mixed developmental disabilities exist (Dunford et al., 2016; Mishin et

al., 2015), no previous studies have specifically investigated training two-wheel bike skills in children with CP. By acknowledging community bike riding as a realistic goal, the focus of this thesis differs from what has been the emphasis of cycling research in this population – stationary cycling for body structures and functions-related outcomes (Armstrong, Spencer, et al., 2019; Chen et al., 2012; Fowler et al., 2010; Johnston et al., 2008; Mohanty et al., 2015; Siebert, DeMuth, Knutson, & Fowler, 2010).

Motor-learning based interventions, including TST, are increasingly being tested in research settings (Bar-Haim et al., 2010; Bleyenheuft et al., 2015, 2017; Thorpe & Valvano, 2002; Valvano, 2004) and, are becoming core to therapists' skillset when working with children with CP, and motor difficulties more broadly. This focus is reinforced by the predominance of TST of bike skills identified by therapists in the current practice survey. However, while there is strong evidence for UL interventions involving task-specificity in children with CP (Novak et al., 2013; Sakzewski, Ziviani, et al., 2014), until our systematic review, the effectiveness of this approach for gross motor skills had not been evaluated. Further, while reports specific to children with CP regarding overarching practices of therapists in Canada (Anaby et al., 2017) and dosage of therapy in the United States (Palisano et al., 2012) have been published, our practice survey was the first study to focus specifically on training two-wheel bike skills. The systematic review and survey not only informed the RCT in this thesis but have identified gaps in the current evidence base and provided direction for strengthening future research specific to bike skills training, and regarding motor-learning informed therapy more broadly.

As the major study within this thesis, the RCT presents some findings consistent with previous literature and some novel findings. It is the first study to test a task-specific approach for attaining two-wheel bike riding goals in children with CP. Previous studies involving bike riding goals in this population have either included children with CP as a subset of the study population (Dunford et al., 2016), or two-wheel bike riding goals were selected by participants in studies exploring cognitive (Gimeno et al., 2019) or participation-focused approaches for attainment of a broader range of goals (Reedman et al., 2019). The design of the experimental intervention as a high-density burst of group-based TST program led by PTs and involving children's own bikes is also novel, as is consideration of personal factor outcomes (Figure 7.1). While this is not the first

study to indicate that TST approaches can have both positive and negative effects on activity outcomes and positive effects on participation outcomes when compared to other interventions (Bleyenheuft et al., 2015), research exploring participation in CP is an emerging field and challenges remain in engaging children in ongoing participation (Imms, Adair, et al., 2016b; Reedman, Boyd, & Sakzewski, 2017). Hence, longer term follow-up is required to explore whether the short- to medium-term impacts of the task-specific program on involvement in bike riding found in the RCT are maintained. However, the positive effects of a form of task-specific gross motor skills training on physical activity and self-perception found in this study have not been previously reported.

Overall, the three studies that form this thesis make a significant contribution to the literature. Moreover, the systematic review and RCT can contribute to future development of best-practice guidelines regarding interventions that improve activity and participation in CP.

## 7.4. Implications for practice and policy

Findings from this thesis are principally relevant to clinicians who work with children with CP, the children themselves, and their families. In practice, goal-directed motor-based training in children with movement difficulties commonly lies within the disciplines physiotherapy and occupational therapy (Anaby et al., 2017). While primarily conducted in health settings – this thesis draws on theory and practice from health, disability, sport and education sectors (Capio, Sit, Eguia, Abernethy, & Masters, 2015; Imms, Granlund, et al., 2016; Steenbergen, van der Kamp, Verneau, Jongbloed-Pereboom, & Masters, 2010; World Health Organisation, 2001). Thus, findings are relevant to policy makers from these sectors, and can also inform the practice of sport and recreational officers, cycling coaches and teachers.

The main clinical implication of this thesis is that a safe, feasible and effective means of training two-wheel bike skills exists for therapists working with ambulant children with CP who have bike riding goals. Guidance is also available to clinicians relating to current bike skills training practices in Australia and evidence for task-specific gross motor skills training more broadly. This thesis reinforces the notion that it is possible

for ambulant children with CP to ride a two-wheel bike. Prescribing modified bicycles including training wheels or tricycles emerged from the practice survey as a relatively common intervention to address bike riding goals in this population. This is understandable due to the motor challenges intrinsic to CP (Graham et al., 2016) and in the previous absence of literature specific to training two-wheel bike riding goals. Clinicians can now have evidence-based discussions with families around setting realistic two-wheel bike riding goals and implement TST methods to attain them. Furthermore, the sub-group analysis of the primary outcomes by GMFCS within the clinical trial means these discussions can be tailored to functional level with goals more likely to be attained in children with greater motor function (GMFCS I), yet salso attainable for children classified GMFCS II.

Given bike riding is a complex combination of perceptual-motor and cognitive tasks with inherent safety considerations (Wierda & Brookhuis, 1991), and that training bike skills may be extending the scope of many therapists, it is important that education in delivering the task-specific program from the trial is developed. This can optimise safe and evidence-based knowledge translation into practice, especially given that the RCT therapists undertook eight hours of training prior to delivering the task-specific program including provision of the training manual (Appendix XII). Delivering the program in a group context as per the protocol may be feasible for clinicians working in larger practices or where a critical mass of children with bike riding goals can form. However, it may be more challenging for other clinicians, for example, sole practitioners working with small populations of children with CP. As such, it is necessary that future training in the task-specific program caters for clinicians working across a variety of contexts. This can be done by drawing on findings from across the PhD, not just the RCT, including knowledge of current practices of therapists and evidence from task-specific gross motor skills training, as well as literature more broadly. For example, recommentations regarding assessment tools for bike riding-related therapy can be derived from the practice survey findings. Moreover, while commonly used, impairment level outcome measures do not appear necessary for deciding eligbility or guiding progression whereas use of bike-related outcome measures such as the cycling skills checklist (Halayko et al., 2016) has been justified by this thesis. This information could be integrated into the RCT therapist training manual (Appendix XII) to form a

comprehensive guide for future education on training bike skills in children with CP. Given the practical nature and safety considerations of riding a bike, clinician training may be enhanced by being face-to-face, however, online modules could also be developed to enable greater clinician access.

This thesis intentionally focussed on activity and participation outcomes for their clinical relevance and importance to children and families. Goal attainment was chosen as the primary outcome of the trial because of the common use of goal-related tools such as the GAS (Kiresuk & Sherman, 1968) and COPM (Law et al., 2005) to evaluate the effectiveness of interventions in paediatric rehabilitation practice, as highlighted in the therapist survey (Cusick et al., 2006; Krasny-Pacini et al., 2013; Oien et al., 2010; Steenbeek et al., 2007). Clinicians now have an evidence-based example of utilising the GAS to evaluate specific bike riding goals. This is of high clinical utility, especially in the context of the ongoing roll out of the National Disability Insurance Scheme in Australia whereby funding for community-based therapy is influenced by the goals of the child. Several tools that have the potential for greater clinical uptake when training bike skills have also been presented. These measures, including the cycling skills test (Halayko et al., 2016), the Activ8™ accelerometer (Activ8, 2017) and the SPP (Harter, 2012b, 2012a; Harter & Pike, 1983), would strengthen clinical outcome assessment across activity, participation and personal factors. However, algorithms within the accelerometer need further refining to differentiate cycling from running as a distinct type of physical activity (Lankhorst et al., 2019) and validation of the cycling skills test and SPP in the CP population are needed.

The reduced rate of participation in physical and recreational activity seen in children with CP compared with their TD peers is a priority issue across health, sport, disability and education sectors (Bjornson, Zhou, Stevenson, Christakis, & Song, 2014; Capio et al., 2015). Evidence for better outcomes in short to medium-term participation in bike riding and physical activity for children in the PT-led task-specific group compared to the home program was found in the trial. However, we do not yet know if the RCT participants have reached the optimum goal of long-term participation in this physical activity. The implication for these sectors is that they now have an opportunity to support or create more inclusive programs and policies to facilitate ongoing participation in bike riding. Examples of relevant contexts include bike education

delivered in primary schools or traffic schools, cycling programs run by sports and recreation organisations, and advocacy for accessible cycling infrastructure. Physiotherapists can play a key role in achieving this by extending their expertise and scope of practice into these programs and settings and, in doing so, facilitate inclusion of ambulant children with CP.

## 7.5. Strengths and limitations

The strengths and limitations of each study have been described within corresponding chapters. Thus, this section will focus on the PhD as a whole and highlight key strengths and limitations of each study.

### 7.5.1 Strengths

The most notable strength is the clinical relevance of the thesis. Each study explores areas that align with the growing focus on activity and participation-based interventions and outcomes in paediatric rehabilitation (Anaby et al., 2017; Rosenbaum & Gorter, 2012; Vargus-Adams & Martin, 2009). Strong recruitment to the RCT suggests that this study was also of interest to families, while the clinical implications of the thesis detailed in section 7.4 reflect its overall applicability to practice. The significance of the thesis as novel research in the context of current literature, as highlighted in section 7.3, is also a key strength as is the conceptual design of this PhD. The systematic review and current practice survey informed the RCT, and consideration of the ICF within each study enabled synthesis of findings across the thesis in a clinically relevant way.

Standardised methodology and reporting were used within the systematic review to strengthen the quality and clinical utility of the evidence produced. Data were extracted, risk of bias was assessed, and the quality of the evidence was determined based on recommendations of the Cochrane Collaboration (Guyatt et al., 2008; Higgins & Green, 2011). Further, intervention replicability and overall study findings were reported using the TIDiER checklist (Hoffmann et al., 2014) and PRISMA items (Moher, Liberati, Tetzlaff, Altman, & PRISMA Group, 2009) respectively. While data in the practice survey was collected using a customised rather than standardised tool, this enabled a comprehensive assessment across the spectrum of bike skills training practices in children with CP. The recruitment strategy involving invitation via national initiatives

and professional associations, as well as snowball methods, meant the practices of therapists from each state and territory in Australia, and from a variety of clinical settings, were represented.

There are several strengths in the RCT design and conduct that warrant highlighting. First, the trial was designed by a panel of co-authors who have specific expertise in CP, physiotherapy interventions, trial methodology and statistical and economic analyses. Input was also sought from the conceptualisation phase from a parent of a child with CP who had bike-riding goals. Considerable efforts were made to minimise bias (Sterne et al., 2019). Risk of bias arising from the randomisation process was addressed by using computer-generated block randomisation numbers prepared by a statistician not directly involved in the study, while allocation was performed following enrolment and concealed from assessment personnel. Detection bias was reduced through blinding of outcome assessors and the use of other therapies was assessed during the study period and was similar between the intervention groups. Further, risk of bias due to missing outcome data was addressed by performing ITT analyses with multiple imputation, while the use of a valid and reliable primary outcome tool reduced the risk of measurement bias. The trial protocol was prospectively registered and published in a peer-reviewed open access journal (Toovey et al., 2018) while strong recruitment meant adequate power was reached to determine effectiveness. Finally, the study was reported based on standard guidelines (Boutron, Altman, Moher, Schulz, & Ravaud, 2017).

### 7.5.2 Limitations

While a focus on two-wheel bike skills training for children with ambulant CP (GMFCS I-II) allowed for specific clinically-applicable evidence to be generated, this also forms arguably the principal limitation of this thesis. Findings are likely not generalisable to children with greater functional restrictions (GMFCS III-V), or those with cognitive deficits, especially given the motor and safety demands of riding a two-wheel bike. Most children with CP are ambulant (Australian Cerebral Palsy Register, 2018) and the focus of intervention research across CP reflects this (Novak et al., 2013). However, it can be argued that children classified within GMFCS levels III-IV have greater need for intervention given the more substantial impact of CP on their function (Palisano et al., 1997). This thesis does not alleviate this need. In saying that, there is evidence that

intensive training involving task-specificity has positive effects on the function of children classified within GMFCS levels III-IV. Thus, modification of the task-specific approach developed in this thesis to more appropriate activities such as adapted cycling in future may hold promise for this group.

Given the sparse evidence regarding training two-wheel bike skills in ambulant children with CP, the systematic review included studies that targeted gross motor skills more broadly. However, the heterogeneity in interventions and outcomes across included studies precluded meta-analysis and no study involved TST for two-wheel bike riding specifically. Extending the scope of the review to include other conditions involving movement challenges such as Developmental Coordination Disorder or ASD may have yielded more cycling-specific information, which, once systematically evaluated, could have been useful in designing the subsequent studies of the thesis.

The major limitation of the practice survey relates to the generalisability of the findings. Although 95 therapists completed the survey, this represents 51% of the 186 therapists who were deemed eligible. This drop-off may be due to the length, structure or relevance of the survey (Saleh & Bista, 2017). A shorter survey reflecting more focussed study aims may have increased the completion rate, and in turn, improved the representativeness of the findings. Additionally, the proportion of OTs who participated was lower than that of PTs, limiting the generalisability and implications of the findings for this discipline. However, this may also reflect a stronger alignment of bike skills training with physiotherapy practice.

There are some additional limitations of the clinical trial to those discussed in Chapter 6. First, while performance bias was minimised by efforts to conceal the study hypothesis from participants, designing an appropriate, scientifically-sound comparison group that did not deter enrolment was difficult, especially when there is no clear ‘usual care’, as demonstrated in Chapter 4. This is not unique to this study – many rehabilitation trials encounter challenges designing control groups (Levack, Martin, Graham, & Hay-Smith, 2019). However, characteristics of the control, a parent-led home program, compared to PT-led intervention group, in a population group where physiotherapy is often key to management, means that some families may have predicted the study hypothesis. Second, the secondary outcome findings need to be

interpreted with caution. These outcomes were included to generate hypotheses only and the study was not powered to detect superior effectiveness of the task-specific program beyond the primary outcome. Further, while useful to comprehensively explore the effects of bike skills training, psychometric properties of some of the secondary outcome measures specific to the CP population have not been published. These include the cycling skills test (Halayko et al., 2016), cycling item of the Pictorial Scale of Perceived Movement Skills Competence (Barnett et al., 2015, 2016), PAQ-C (Crocker et al., 1997), SPP (Harter, 2012b, 2012a; Harter & Pike, 1983), and CHU-9D (Stevens, 2010). Third, the trial was not powered for analyses to be adjusted for variables other than site. This meant the impact of the imbalance in bike skills at baseline on bike skills outcomes was not explored. Further, the difference between the groups found at either time point did not reach significance despite clear differences over time, particularly in the task-specific group. Finally, the fidelity study and economic evaluation detailed in the trial protocol were out of the scope of the PhD. These studies may yield useful information for greater impact in practice and policy so will be reported following thesis submission.

## 7.5 Future research directions

This thesis builds a foundational evidence base for task-specific bike skills training in children with CP, yet further research is required to comprehensively understand this field of research and ultimately optimise opportunities for children to achieve their bike riding goals. Three key areas are proposed below. With growing evidence to support the involvement of consumers for greater research relevance and impact (Mcintyre, Novak, & Cusick, 2010), it will be important to build on the role of parent advisors in this thesis to involve consumers, including children, in any future efforts.

### 7.5.3 Tailoring intervention to the individual

As discussed in Chapter 6, a drawback of RCT designs is the focus on group differences rather than individual responses to the intervention. There are; however, ways to explore data collected from the trial to enable more tailored recommendations, albeit acknowledging the limitations associated with post-hoc analyses. For example, investigating characteristics of best responders to either program may provide guidance

for clinicians around which families to suggest trialling a parent-led approach in the first instance. This is especially relevant if the task-specific program is only run during school holidays as was done in the trial meaning there may be waiting periods, or at a cost to participants. Previous evidence suggests that successfully learning to ride a two-wheel bike in ambulant children with CP was associated with how important it was to parents that their child rode (Toovey, Reid, et al., 2017). Data collected in the trial also enables future exploration of this relationship. If findings are replicated, the case for educating parents on the benefits of bike riding is strengthened. However, exploration of data only collected from the trial would not take into account those children who had already achieved their bike riding goals and thus may not have considered participating. This limitation also raises the question about whether children younger than six years old (the minimum eligible age for the RCT) are appropriate for two-wheel bike skills training. Given it has been reported that children in the general population typically learn to ride between ages four and six, future research involving younger children is warranted (Toovey, Reid, et al., 2017).

Motor-learning based training, such as the task-specific program in this thesis, is an emergent field in CP research and practice (Bleyenheuft & Gordon, 2014; Morgan et al., 2016). This training has the potential to involve selection of motor learning strategies that are individualised to the child, including type of feedback and structure of practice. However, debate remains around what constitutes a motor-learning approach to therapy and, as described in Chapter 3, poor reporting of motor learning strategies limits evaluation of the relationship of strategies to outcome. To tailor these interventions, future research should explore different comparison interventions and individual responses to strategies, utilise standardised reporting frameworks and comprehensively report fidelity to training. The fidelity study, detailed in the trial protocol but out of the scope of this thesis, uses the Motor Learning Strategies Rating Instrument (Ryan, Levac, & Wright, 2019) to examine therapist fidelity to the task-specific program and may help address these issues.

### 7.5.4 Stronger understanding across ICF domains

The focus and findings of this thesis sit within the activity, participation and personal domains of the ICF (Figure 7.1). To gain a stronger understanding of the factors that

influence, and the impact of bike riding goal attainment in ambulant children with CP, the body functions and structures, and environmental domains need greater attention. Again, data collected at baseline in the RCT such as social risk and parents' own bike-riding competency may prove useful for exploring any relationships with these domains. Further, the economic evaluation of the trial, which is to be reported following thesis submission, will provide greater insight into this environmental consideration. To strengthen the evidence for the effect of bike skills training across the ICF, hypotheses generated by secondary outcome findings from the trial should also be explored. However, to reduce measurement bias, it is important that the validity and reliability of outcome tools are established for children with CP. Establishing the psychometric properties of the Cycling Skills Test, SPP, and refining those of Activ8™ accelerometer, are priorities given the potential clinical utility of these tools.

### 7.5.5 Investigating inclusive interventions and longer-term outcomes

Finally, this PhD provides justification to extend the scope of research on training community cycling skills in children with CP to areas where a dearth of evidence remains – for semi or non-ambulant children and regarding longer-term outcomes. It is promising that an RCT exploring the efficacy of a combined functional electrical stimulation and adapted recreational cycling program for children classified GMFCS II-IV is currently underway (Armstrong, Boyd, et al., 2019). There is also emerging evidence to support environmental modifications (Anaby, Law, Feldman, Majnemer, & Avery, 2018) and behaviour change approaches for performance of participation goals (Reedman et al., 2019) in children across the spectrum of function in CP. Combining task-specific skills training with adapted cycling equipment and a behaviour change approach is an intervention that warrants further investigation for children with greater functional limitations who have cycling goals. Further, this thesis does not address outcomes beyond three months post-intervention. Longer-term follow up of the RCT participants would provide insight into factors associated with ongoing goal attainment and participation in bike riding to inform the development of much needed interventions to address these issues.

## 7.6. Conclusions

This thesis has developed and tested a novel task-specific approach to training bike skills in ambulant children with CP. New knowledge around task-specific gross motor skills training and current practices of therapists when training bike skills in this population has been generated and informed the design of a high-quality RCT. This trial found the task-specific program was more effective than a parent-led home program for attaining two-wheel bike riding goals in ambulant children with CP. The three studies within this thesis each form original contributions to the literature while use of the ICF as a framework enabled clinically relevant findings from each study to synergise, strengthening the contribution of the PhD as a whole. Evidence that ambulant children with CP can attain two-wheel bike riding goals through TST is now available, while this thesis also contributes to emerging research considering activity, participation and personal factor domains in outcome measurement. Findings from this PhD should inform education targeted towards clinicians working with children with CP with two-wheel bike riding goals, for evidence-based translation into practice. Finally, this thesis has implications for policy beyond health and will inform future research regarding children with CP who have cycling goals.

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| Authors | Rachel Toovey, Charmaine Bernie, Adrienne R Harvey, Jennifer L McGinley, Alicia J Spittle | |
| Student's contribution (%) | >70% | |
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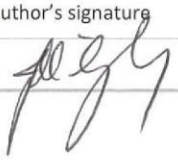
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| Authors | Rachel Toovey, Charmaine Bernie, Adrienne R Harvey, Jennifer L McGinley, Alicia J Spittle | |
| Student's contribution (%) | >70% | |
| Journal or book name | BMJ Paediatrics Open | |
| Volume/page numbers | 1:e000078. DOI: 10.1136/bmjpo-2017-000078 | |
| Status | ☐ Accepted and In-press ☐ In progress ☐ Published | Date accepted/published: Published online 11/8/2017 |

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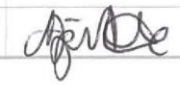
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| Full title | Task-specific gross motor skills training for ambulant school-aged children with cerebral palsy: a systematic review | |
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> 1. Toovey, R., Bernie, C., Harvey, A., McGinley, J., & Spittle, A. (2017). Task-specific gross motor skills training for ambulant school-aged children with cerebral palsy: a systematic review. BMJ Paediatrics Open, 1(1), e000078. https://doi.org/10.1136/bmjpo-2017-000078https://doi.org/10.1136/bmjpo-2017-000078
> 2. Toovey, R., Harvey, A. R., McGinley, J. L., Lee, K. J., Shih, S. T. F., & Spittle, A. J. (2018). Bike skills training for children with cerebral palsy: protocol for a randomised controlled trial. BMJ Open, 8(2), e019898. https://doi.org/10.1136/bmjopen-2017-019898https://doi.org/10.1136/bmjopen-2017-019898
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> Many thanks
> Rachel
>
> Rachel Toovey BPhysio (Hons), MPHTM
> Physiotherapist | PhD candidate | Lecturer
> Neurodisability & Rehabilitation | Murdoch Children's Research Institute
> Department of Physiotherapy | University of Melbourne
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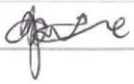
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| | | |
|---|---|---|
| Full title | Training Two-Wheel Bike Skills in Children with Cerebral Palsy: A Practice Survey of Therapists in Australia | |
| Authors | Rachel Toovey, Alicia J Spittle, Anna Nicolaou, Jennifer L McGinley, Adrienne R Harvey | |
| Student's contribution (%) | >70% | |
| Journal or book name | Physical and Occupational Therapy in Pediatrics | |
| Volume/page numbers | DOI: 10.1080/01942638.2019.1585404 | |
| Status | ☐ Accepted and In-press ■ Published ☐ In progress | Date accepted/published: Published online 18/3/2019 |

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| Alicia J Spittle |  | 23/07/19 |

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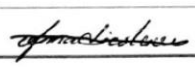
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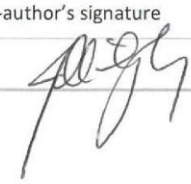
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| Jennifer L McGinley |  | 22/7/19 |

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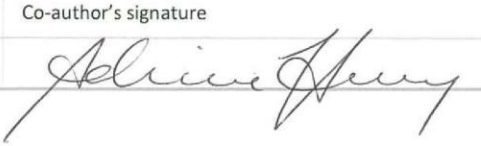
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| Article DOI: | 10.1080/01942638.2019.1585404 |
| Author(s): | Rachel A.M. Toovey, Alicia Spittle, Anna Nicolaou, Jennifer McGinley, Adrienne Harvey |
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# APPENDIX VI Chapter 4 ethics and governance approval certificate

## ETHICS APPROVAL & GOVERNANCE AUTHORISATION

The Royal Children's Hospital Melbourne

22 April 2016

Ms Rachel Toovey
Developmental Disability and Rehabilitation
Murdoch Childrens Research Institute

Dear Ms Toovey

**Project Title:** **Bike skills training for children with cerebral palsy in Australia: what is standard practice?**

---

**RCH HREC Reference Number: 36092A**

I am pleased to advise that the above project has received ethical approval from The Royal Children's Hospital Melbourne Human Research Ethics Committee (HREC).

The HREC confirms that your proposal meets the requirements of the National Statement on Ethical Conduct in Human Research (2007). This HREC is organised and operates in accordance with the National Health and Medical Research Council's (NHRMC) National Statement on Ethical Conduct in Human Research (2007), and all subsequent updates, and in accordance with the Note for Guidance on Good Clinical Practice (CPMP/ICH/135/95), the Health Privacy Principles described in the Health Records Act 2001 (Vic) and Section 95A of the Privacy Act 1988 (and subsequent Guidelines).

The project has also received governance authorisation at the Melbourne Children's Campus (incorporating The Royal Children's Hospital, Murdoch Children's Research Institute and the University of Melbourne Department of Paediatrics).

**HREC Approval Date:** 22 April 2016*
*Please note the HREC are no longer issuing pre-determined approval periods. Ethical approval is now ongoing, subject to the submission of an annual report on the anniversary of approval.*

**Participating Sites**:
Ethical approval for this project applies at the following sites:

| Site Name |
|---|
| • The Royal Children's Hospital, Parkville and Murdoch Childrens Research Institute, Parkville |

**Approved Documents:**
The following documents have been reviewed and approved:

| Document | Version | Date |
|---|---|---|
| Protocol | 1.0 | 18 March 2016 |
| PIS and CF | 1.0 | 4 April 2016 |
| Survey | 1.0 | 4 April 2016 |
| Invitation letter | 1.0 | 4 April 2016 |
| Advertisements | 1.0 | 4 April 2016 |

**Conditions of Ethics Approval:**
- You are required to submit to the HREC:
  - An Annual Progress Report (that covers all sites listed on approval) for the duration of the project. This report is due on the anniversary of HREC approval. Continuation of ethics

approval is contingent on submission of an annual report, due within one month of the approval anniversary. Failure to comply with this requirement may result in suspension of the project by the HREC.
    - A comprehensive Final Report upon completion of the project.
- Submit to the reviewing HREC for approval any proposed amendments to the project including any proposed changes to the Protocol, Participant Information and Consent Form/s and the Investigator Brochure.
- Notify the reviewing HREC of any adverse events that have a material impact on the conduct of the research in accordance with the NHMRC Position Statement: *Monitoring and reporting of safety for clinical trials involving therapeutic products May 2009*.
- Notify the reviewing HREC of your inability to continue as Coordinating Principal Investigator.
- Notify the reviewing HREC of the failure to commence the study within 12 months of the HREC approval date or if a decision is taken to end the study at any of the sites prior to the expected date of completion.
- Notify the reviewing HREC of any matters which may impact the conduct of the project.
- If your project involves radiation, you are legally obliged to conduct your research in accordance with the Australian Radiation Protection and Nuclear Safety Agency Code of Practice 'Exposure of Humans to Ionizing Radiation for Research Purposes' Radiation Protection series Publication No.8 (May 2005)(ARPANSA Code).
- The HREC, authorising institution and/or their delegate/s may conduct an audit of the project at any time.

Yours sincerely



**Emma Land**
Research Ethics and Governance Officer
Research Ethics and Governance
The Royal Children's Hospital Melbourne
Phone : (03) 9345 5044
Email : rch.ethics@rch.org.au
Web : www.rch.org.au

# APPENDIX VII Chapter 4 participant information and consent form

## Information statement and consent form

| | |
|---|---|
| **HREC Project Number:** | 36092A |
| **Research Project Title:** | Bike skills training for children with cerebral palsy in Australia: what is standard practice? |
| **Principal Researcher:** | Rachel Toovey, PhD candidate<br>Developmental Disability & Rehabilitation Research<br>Murdoch Childrens Research Institute<br>Department of Physiotherapy<br>The University of Melbourne |
| **Version Number:** | **1** **Version Date:** 04/04/16 |

You are invited to participate in a research project that is explained below.

**What is an Information Statement?**

The following information tells you about the research project. It explains to you clearly and openly all the steps and procedures of the project. The information is to help you to decide whether or not you would like to take part in the research.

Please read this Information Statement carefully. You can ask us questions about anything in it.

Participation in this research project is voluntary. If you don't want to take part, you don't have to. You can withdraw from the project at any time without explanation and this will not affect your employment, or any other relationship you may have, with The Royal Children's Hospital, Murdoch Childrens Research Institute or the University of Melbourne.

Once you have understood what the project is about, if you would like to take part please check the consent box at the start of the survey. More information about the survey is provided below.

You can print a copy of this information statement to keep.

**What is the research project about?**

This research project aims to determine the current practice of physiotherapists and occupational therapists around training skills related to riding a two-wheel bike to children with cerebral palsy (CP).

On average, children with CP have lower levels of physical activity and participation in recreational activities than their typically developing (TD) peers. Low levels of physical activity in childhood have been linked to the development of chronic disease in adulthood whilst physical activity has been associated with better quality of life and happiness in children with CP. Bike riding is a common activity for children generally and may be a means of increasing physical activity and improving overall health and participation in this population.

In Australian clinical practice, physiotherapists and occupational therapists generally implement therapy related to improving motor skills or task performance such as riding a bike. However, therapists are currently faced with a lack of evidence in terms of approaches for training bike specific skills in children with CP. Prior to researching novel interventions for improving bike riding skills in children with CP, it is important to understand what practice currently involves, or would involve, for Australian physiotherapists and occupational therapists. However, this is not yet known.

We want to find out:

1) What standard practice is, or would be, for occupational therapists and physiotherapists when training skills related to riding a two-wheel bike in children with CP
2) The specific components of this training including considerations, assessment, interventions involved, settings, intensity and evaluation

We hope that physiotherapists and occupational therapists involved in supporting children and young people with CP in Australia will take part in the project. Therapists can take part even if they have not yet provided bike skills training to children with CP.

**Who is funding this research?**

The project is being conducted by a team of researchers, led by PhD candidate Rachel Toovey. Rachel's PhD is funded by an Australian Postgraduate Award scholarship, a Murdoch Childrens Research Institute Postgraduate Health Research top up scholarship and a Centre for Research Excellence in Cerebral Palsy top up scholarship.

**Why am I being asked to be in this research project?**

We are asking you to take part in this project because you

- are a physiotherapist or occupational therapist
- work in Australia
- have experience working with children and young people with CP and their families

**What does participation in this research involve?**

If you consent to take part, we will ask you to complete a short, web-based survey. The survey includes questions about your experience working with children with cerebral palsy, the components of bike skills training you would consider implementing for children with CP and the reasons you choose to implement these components.

It is expected that it will take less than 15 minutes for you to complete the survey.

**What are my alternatives to taking part in this project?**

You do not have to take part if you do not want to. If you choose not to take part your employment, or any other relationship you may have with either The Royal Children's Hospital, Murdoch Childrens Research Institute or the University of Melbourne, will not be affected in any way.

**What are the possible benefits for me?**

There are unlikely to be any direct personal benefits in participating for you.

**What are the benefits to other people in the future?**

We expect the main benefits of this study to be for others in the future. We hope the information we get will help us to develop bike skills programs for children with CP and may help increase participation in physical, recreational and social activities through bike riding.

**What are the possible risks or inconveniences?**

There are no anticipated risks, side effects or discomforts. The main inconvenience of taking part in this project is the time taken to complete the survey. We anticipate that this will be less than 15 minutes.

**What will be done to make sure my information is confidential?**

Any information we collect from you will remain confidential. We will use your information only for this research project. Only the researchers involved in the project and The Royal Children's Hospital Ethics Committee can have access to this information. We can disclose the information only with your permission except as required by law.

The information will be non-identifiable. This means that your responses to the survey will be anonymous. We will not be able to match your survey responses with any identifying information about you.

At the end of the study, results may be presented at conferences or published in medical journals. We are required to keep information for at least 5 years following the last publication of the project. The research information may be destroyed or kept indefinitely in secure storage after this time.

The results of this research will be used by Rachel Toovey, to get a Doctor of Philosophy (PhD) degree.

**Will I be informed of the results when the research project is finished?**

At the end of the survey, you will be given the opportunity to register your name and email address to receive a summary of the study results or to receive information on future research projects in the field of bike riding for children with CP. The research staff will not be able to match your email address with your survey responses, so your responses will remain anonymous.

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If you would like more information about the project or if you need to speak to a member of the research team, please contact:

**Name:** Rachel Toovey **Contact details:** (03) 9936 6088 or rachel.toovey@mcri.edu.au

If you have any concerns and/or complaints about the project, the way it is being conducted or your rights as a research participant, and would like to speak to someone independent of the project, please contact: Director, Research Ethics & Governance, The Royal Children's Hospital Melbourne on telephone: (03) 9345 5044.

***NOTE:*** *This Information Statement may have opened in a new tab in your web browser. To indicate if you provide your consent to participate, and to continue on with the survey, please return to the previous tab.*

*Confidential*



# Bike skills training for children with cerebral palsy

Thanks for your interest in our survey!!

We anticipate that the survey will take less than 15 minutes of your time, and that your responses will help us immensely.

The majority of questions are multiple choice, allowing you to move through the survey quickly. There will also be opportunities to provide more detailed answers to some questions if you would like to do so.

You can pause the survey at any time. Your answers will be saved, and you can come back to complete your survey at a later time.

The survey will remain open until Friday 17 June, 2016.

All of your responses will remain anonymous. The research team have no way of identifying who you are, or matching survey respondents with their answers.

If you do have any questions about the study or would like to do the survey over the phone, you can contact the team at rachel.toovey@mcri.edu.au or (03) 9936 6088.

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Before deciding if you would like to continue, please download and read the information statement attached.

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Participant information statement

[Attachment: "Participant Information Statement v1 2016_04_04 HREC 36092A.pdf"]

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Based on the aims of this project, the survey is open to physiotherapists and occupational therapists in Australia who support children with cerebral palsy and their families.

---

| | |
|---|---|
| Do you work as a physiotherapist or occupational therapist in Australia? | ◯ Yes<br>◯ No |

---

| | |
|---|---|
| In your clinical practice do you support children with cerebral palsy? | ◯ Yes<br>◯ No |

If you answered yes to both of these question, you are eligible to participate.
Do you provide your consent to participate in this survey?

By selecting YES, you are indicating that you:
- have read the information statement version attached above and understand its contents.
- believe that you understand the purpose, extent and possible risks of your involvement in this project.
- voluntarily consent to take part in this research project.
- have had an opportunity to ask questions and are satisfied with the answers you have received.
- understand that this project has been approved by The Royal Children's Hospital Melbourne Human Research Ethics Committee and will be carried out in line with the National Statement on Ethical Conduct in Human Research (2007) - updated December 2013.
- understand that you can print a copy of the Information Statement for your records.

◯ Yes - I consent to participate in the survey ◯ No - I do not wish to participate in the survey

**Questions in the survey refer to the Gross Motor Functioning Classification System (GMFCS) and the Manual Abilities Classification System (MACS). If you are not familiar with these systems you may like to review the below PDFs. You can keep them open whilst you complete the survey.**

1. Gross Motor Function Classification System (GMFCS) descriptors for children with cerebral palsy (CP) aged 6 - 12 years.

[Attachment: "New GMFCS 6-12.pdf"]

2. GMFCS descriptors for children with CP aged 12 - 18 years.

[Attachment: "New GMFCS 12-18.pdf"]

3. Manual Ability Classification System (MACS) descriptors for children with CP aged 4-18 years.

[Attachment: "MACS_2010.pdf"]

| **Your professional background:** | |
|---|---|
| What is your professional background? | ◯ Occupational therapist<br>◯ Physiotherapist<br>(select only one) |
| In addition to your physiotherapy or occupational therapy tertiary qualifications, do you have any relevant additional qualifications? | ◯ Yes<br>◯ No |
| Please describe your relevant additional qualifications | _____ |
| What proportion of your clinical caseload is comprised of supporting children with CP? | ◯ < 25%,<br>◯ 26-50%<br>◯ 51-75%<br>◯ > 75%<br>◯ I do not support children with CP<br>(select only one) |
| What proportion of the children with CP with whom you work are classified as GMFCS levels I-II? | ◯ < 25%,<br>◯ 26 - 50%<br>◯ 51 - 75%<br>◯ > 75%<br>◯ I do not work with children with CP who are classified as GMFCS levels I-II |
| How many years have you been working with children with CP? | ◯ < 1 year<br>◯ 1 - 5 years<br>◯ 6 - 10 years<br>◯ 10 - 15 years<br>◯ > 15 years |
| Which setting best describes where you conduct your primary clinical practice? | ◯ Metropolitan hospital - acute department<br>◯ Metropolitan hospital - rehabilitation department<br>◯ Regional hospital - acute department<br>◯ Regional hospital - rehabilitation department<br>◯ Community - public<br>◯ Community - private<br>◯ Early childhood intervention service<br>◯ School-aged service<br>◯ Special education<br>◯ Other<br>(If more than one, select your primary setting of practice) |
| Please describe this setting. | _____ |

| | |
|---|---|
| In which state or territory do you primarily carry out your therapy practice? | ◯ Tasmania<br>◯ South Australia<br>◯ New South Wales<br>◯ Northern Territory<br>◯ Victoria<br>◯ Queensland<br>◯ Australian Capital Territory<br>◯ Western Australia<br>(If more than one, select your primary state of practice) |

**Expectations**

**Now we would like you to consider your expectations around children with CP and their ability to ride a two wheel bike without training wheels (also known as stabilisers).**

| | |
|---|---|
| Children with CP classified at which GMFCS levels do you think may be able to ride a two-wheel bike without training wheels? | ☐ GMFCS I<br>☐ GMFCS II<br>☐ GMFCS III<br>☐ GMFCS IV<br>☐ GMFCS V<br>(select as many as apply) |
| Children with CP classified at which manual ability classification system (MACS) levels do you think may be able to ride a two-wheel bike without training wheels? | ☐ MACS I<br>☐ MACS II<br>☐ MACS III<br>☐ MACS IV<br>☐ MACS V<br>(select as many as apply) |

**For the purposes of this survey, bike skills training refers to training skills related to riding a two-wheel bike.**

| | |
|---|---|
| Has a child / children with CP been referred to you for bike skills training or a child with CP under your care had goals related to riding a two-wheel bike? | ◯ Yes<br>◯ No |
| Did you provide bike skills training / therapy to this child / children? | ◯ Yes<br>◯ No |
| Instead of providing bike skills training, what did you do? | ☐ Referred the child to another physiotherapist<br>☐ Referred the child to another occupational therapist<br>☐ Referred the child to another professional<br>☐ Referred the child to an established program (including programs coordinated by physiotherapists and/or occupational therapists)<br>☐ Provided guidance or information to the parent/s or carer/s only<br>☐ I did not provide any therapy related to this and did not refer elsewhere<br>☐ Other<br>(select as many as apply) |
| Can you share with us why you referred to another physiotherapist? | _____ |
| Can you share with us why you referred to another occupational therapist? | _____ |
| Can you share with us why you referred the child to another professional? | _____ |
| What was the profession of the other professional you referred the child to? | _____ |
| Can you share with us why you referred the child to an established program? | _____ |
| What was the name of the established program? | _____ |
| Can you describe any other details of the established program?<br><br>This may include setting, intensity of training, professions of the service providers, cost etc. | _____ |
| Please describe the format and content of the guidance you provided to the parent/s or carer/s. | _____ |

| | |
|---|---|
| Can you share with us why you did not provide bike skills training to the child or refer them elsewhere? | |
| Please describe "other." | |
| Are there any key criteria children with CP would have to meet for you to provide bike skills training to them? | |
| Please describe why these are key criteria for you to provide bike skills training to the child | |
| Are there any characteristics or co-morbidities in children with CP that mean you would not provide bike skills training for them? | |
| Please describe why you would not provide bike riding therapy to the child with these characteristics or co-morbidities | |

| **Please indicate how often you would consider each of the following factors as a contraindication to providing two-wheel bike skills training to a child with CP** | | | |
|---|---|---|---|
| | Always | Sometimes | Never |
| GMFCS level I | ○ | ○ | ○ |
| GMFCS II | ○ | ○ | ○ |
| GMFCS III | ○ | ○ | ○ |
| MACS level I | ○ | ○ | ○ |
| MACS II | ○ | ○ | ○ |
| MACS III | ○ | ○ | ○ |
| Good ability to communicate to therapists | ○ | ○ | ○ |
| Poor ability to communicate to therapists | ○ | ○ | ○ |
| Age < 5 years | ○ | ○ | ○ |
| Age 5 - 15 years | ○ | ○ | ○ |
| Age > 15 years | ○ | ○ | ○ |
| Challenging behaviour | ○ | ○ | ○ |
| Mild intellectual disability | ○ | ○ | ○ |
| Moderate intellectual disability | ○ | ○ | ○ |
| Severe intellectual disability | ○ | ○ | ○ |
| Hemiplegic (unilateral) topography | ○ | ○ | ○ |
| Diplegic (bilateral) topography | ○ | ○ | ○ |

If not listed already, are there any additional factors you would consider a contraindication?

_____

If not listed already, are there any additional specific co-morbidities you would see as a contraindication or precaution to providing bike skills training?

Please describe whether each factor is a contraindication or precaution.

_____

If not listed already, are there any specific medications you would see as a contraindication or precaution to providing bike skills training?

Please describe whether each factor is a contraindication or precaution.

_____

**Assessment**

**We'd like to find out more about the assessment you would undertake when providing bike skills training to children with CP:**

What would you measure or assess in a child with CP who you are providing bike skills training to?

This is related in particular to the bike skills training rather than the overall care you may provide.

**Please indicate whether you would include the following components in your assessment of children with cerebral palsy specifically related to bike skills training?**

| | Always | Sometimes | Never |
|---|---|---|---|
| Spasticity | ○ | ○ | ○ |
| Range of movement | ○ | ○ | ○ |
| Strength | ○ | ○ | ○ |
| Balance | ○ | ○ | ○ |
| Coordination | ○ | ○ | ○ |
| Aerobic capacity | ○ | ○ | ○ |
| Motor planning ability | ○ | ○ | ○ |
| Learning style | ○ | ○ | ○ |
| Previous bike riding experience or ability | ○ | ○ | ○ |
| Child and family goals | ○ | ○ | ○ |

Are there any additional factors you would assess?

○ Yes
○ No

Please describe these additional factors.

| | |
|---|---|
| Would you use any outcomes measures to support your assessment? | ◯ Yes<br>◯ No |

Please describe these outcome measures.

**Bike skills training**

**We would now like to find out more about the bike skills training or would provide to children with CP. Just a reminder that for the purposes of this survey, bike skills training refers to training skills related to riding a two-wheel bike.**

Please briefly describe any key intervention approaches involved in the bike skills training you would provide to children with CP. Use of dot points is acceptable.

I.e. Neurodevelopmental therapy or goal directed training

**Of the interventions/approaches you use in your practice, how often would you use the following in the bike skills training you would provide with children with CP. If you do not use a named intervention, do not select a box for that option.**

| | Always | Sometimes |
|---|---|---|
| Provision of a modified bike with training wheels | ○ | ○ |
| Task specific training | ○ | ○ |
| Stationary bike training | ○ | ○ |
| Neurodevelopmental therapy (NDT) | ○ | ○ |
| Conductive education | ○ | ○ |
| Goal directed training | ○ | ○ |
| Electrical stimulation | ○ | ○ |
| Activity focussed training | ○ | ○ |
| Vojta therapy | ○ | ○ |
| Context focussed therapy | ○ | ○ |
| Strength training | ○ | ○ |
| Balance training | ○ | ○ |
| Balance (walker) bike training | ○ | ○ |

Are there any additional intervention approaches you would use not listed above or described already?
○ Yes
○ No

Please describe these additional intervention approaches.

_____

Briefly describe your rationale for choosing this intervention approach or combination of intervention approaches.

_____

| | |
|---|---|
| Describe any physical equipment or materials involved in the bike skills training you would provide (if at all).<br><br>I.e. bicycle, specialised bicycle, information for child/parent/other | |
| Describe any specific strategies involved in the bike skills training you would provide, if at all. (i.e. cognitive strategies, motor learning strategies) | |

**Of the strategies you use in your practice, how often would you use the following in the bike skills training you would provide with children with CP. If you do not use a named strategy, do not select a box for that option.**

| | Always | Sometimes |
|---|---|---|
| Feedback | ○ | ○ |
| Task modification | ○ | ○ |
| Environmental modification | ○ | ○ |
| Dual task learning | ○ | ○ |
| Repetitive practice | ○ | ○ |
| Observational learning | ○ | ○ |
| Movement imagery | ○ | ○ |
| Trial and error learning | ○ | ○ |
| Physical guidance | ○ | ○ |
| Goal setting with child and family | ○ | ○ |
| Cognitive strategies | ○ | ○ |
| Home program | ○ | ○ |

Are they any additional strategies or procedures/activities that would be part of the bike skills training you provide?

○ Yes
○ No

Please describe these additional strategies / processes / activities.

_____

| | |
|---|---|
| Would anyone assist you to deliver the bike skills training to children with CP?<br><br>I.e. therapy assistant, other therapist, parent or carer | ◯ Yes<br>◯ No |
| Who would assist you? | ☐ Parent or carer<br>☐ Therapy assistant<br>☐ Other therapist<br>☐ Other<br>(select as many as apply) |
| Please describe "other." | _____ |

**Delivery of bike skills training**

**We would now like to find out about how you would deliver bike skills training to children with CP.**

| | |
|---|---|
| How would you deliver bike skills training to children with CP? | ☐ Face to face<br>☐ Internet based<br>☐ Over the phone or video-conferening<br>☐ Individual (one child at a time)<br>☐ Group-based<br>(Select ALL that apply) |
| Describe the location or setting where you would deliver the bike skills training. | _____ |

**Please indicate if you would deliver the bike skills training in the following settings routinely, sometimes, occasionally or never**

| | Always | Sometimes | Never |
|---|---|---|---|
| Therapy gym or rooms | ○ | ○ | ○ |
| Child's home | ○ | ○ | ○ |
| Child's school | ○ | ○ | ○ |
| Community setting (e.g. community reserve / paths) | ○ | ○ | ○ |

**Please indicate if you would deliver the bike skills training on the following surfaces routinely, sometimes or never.**

| | Always | Sometimes | Never |
|---|---|---|---|
| Natural grass | ○ | ○ | ○ |
| Astroturf (e.g. hockey pitch) | ○ | ○ | ○ |
| Pavement | ○ | ○ | ○ |
| Flat surface | ○ | ○ | ○ |
| Sloping surface | ○ | ○ | ○ |
| Indoor firm surface (e.g. gym floorboards) | ○ | ○ | ○ |
| Indoor soft surface (e.g. carpet) | ○ | ○ | ○ |

Are there additional types of locations, settings or surfaces you would use?
○ Yes
○ No

Describe these additional features of the training location and setting.

**The following questions relate to the dosage or intensity of the bike skills training you would provide.**

**Just a reminder that for the purposes of this survey, bike skills training refers to training skills related to riding a two-wheel bike.**

| | |
|---|---|
| On average, how many sessions would you provide? | _____ (Type number. Min 1, Max 20) |
| On average, how many minutes would each session be? | _____ (Type number. Min 1, Max 480) |
| On average, over what time period would these sessions occur? Please indicate whether your answer is in days, weeks or months. | _____ |
| Would there be any fees charged to the family for the bike skills training? | ◯ Yes<br>◯ No |
| What would be the fees be (per session)? | _____ |

**Evaluating bike skills training**

**To finish, we'd like to ask about evaluation of the bike skills training you would provide to children with CP.**

Would you evaluate the effectiveness of the bike skills training?

◯ Yes
◯ No

How would you evaluate the effectiveness of the bike skills training?

Briefly describe your rationale for measuring the effectiveness of the bike skills training in this way.

At what time points would you measure the effectiveness of the bike skills training?

Briefly describe your rationale for not measuring the effectiveness of the bike skills training.

When and why would you stop providing bike skills training?

Do you have any final thoughts or comments that you would like to share with us?

◯ Yes - I have some comments or ideas! ◯ No - I have no further comments

You can enter your comments or ideas below:
(Your comments can be as brief or detailed as you wish)

You have now reached the end of the survey.

To finish, please click SUBMIT at the bottom of the page.

You will be redirected to a web form where you can sign up to receive a summary of the results of this study or to be involved in future research in this field.

Please note if you sign up your details will not be connected to your survey response. If you do not wish to sign up, just exit the form when it comes up.

Thank you for your time.

## Co-author authorisation form

THE UNIVERSITY OF MELBOURNE

All co-authors must complete this form. By signing below co-authors agree to the listed publication being included in the student's thesis and that the student contributed greater than 50% of the content of the publication and is the "primary author" ie. the student was responsible primarily for the planning, execution and preparation of the work for publication.

In cases where all members of a large consortium are listed as authors of a publication, only those that actively collaborated with the student on material contained within the thesis should complete this form. This form is to be used in conjunction with the *Declaration for a thesis with publication form*.

Students must submit this form, along with the *Declaration for thesis with publication form*, when the thesis is submitted to the Thesis Examination System: *https://tes.app.unimelb.edu.au/*

Further information on this policy and the requirements is available at:
*gradresearch.unimelb.edu.au/preparing-my-thesis/thesis-with-publication*

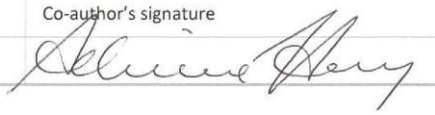
**A. PUBLICATION DETAILS** *(to be completed by the student)*

| | | |
|---|---|---|
| Full title | Bike skills training for children with cerebral palsy: protocol for a randomised controlled trial | |
| Authors | Rachel Toovey, Adrienne R Harvey, Jennifer L McGinley, Katherine J Lee, Sophy TF Shih, Alicia J Spittle | |
| Student's contribution (%) | 70% | |
| Journal or book name | BMJ Open | |
| Volume/page numbers | 8:e019898 DOI: 10.1136/bmjopen-2017-019898 | |
| Status | ☐ Accepted and In-press ■ Published ☐ In progress | Date accepted/published: Published 3/2/2018 |

**B. CO-AUTHOR'S DECLARATION** *(to be completed by the collaborator)*

I authorise the inclusion of this publication in the student's thesis and certify that:

- the declaration made by the student on the *Declaration for a thesis with publication form* correctly reflects the extent of the student's contribution to this work;
- the student contributed greater than 50% of the content of the publication and is the "primary author" ie. the student was responsible primarily for the planning, execution and preparation of the work for publication.

| Co-author's name | Co-author's signature | Date *(dd/mm/yy)* |
|---|---|---|
| Adrienne R Harvey |  | 24/7/19 |

# Co-author authorisation form

THE UNIVERSITY OF MELBOURNE

All co-authors must complete this form. By signing below co-authors agree to the listed publication being included in the student's thesis and that the student contributed greater than 50% of the content of the publication and is the "primary author" ie. the student was responsible primarily for the planning, execution and preparation of the work for publication.

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Students must submit this form, along with the *Declaration for thesis with publication form*, when the thesis is submitted to the Thesis Examination System: *https://tes.app.unimelb.edu.au/*

Further information on this policy and the requirements is available at:
*gradresearch.unimelb.edu.au/preparing-my-thesis/thesis-with-publication*

**A. PUBLICATION DETAILS** *(to be completed by the student)*

| | | |
|---|---|---|
| Full title | Bike skills training for children with cerebral palsy: protocol for a randomised controlled trial | |
| Authors | Rachel Toovey, Adrienne R Harvey, Jennifer L McGinley, Katherine J Lee, Sophy TF Shih, Alicia J Spittle | |
| Student's contribution (%) | 70% | |
| Journal or book name | BMJ Open | |
| Volume/page numbers | 8:e019898 DOI: 10.1136/bmjopen-2017-019898 | |
| Status | ☐ Accepted and In-press ■ Published<br>☐ In progress | Date accepted/published<br>Published 3/2/2018 |

**B. CO-AUTHOR'S DECLARATION** *(to be completed by the collaborator)*

I authorise the inclusion of this publication in the student's thesis and certify that:

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| Co-author's name | Co-author's signature | Date *(dd/mm/yy)* |
|---|---|---|
| Jennifer L McGinley |  | 23/7/19 |

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|---|---|
| Full title | Bike skills training for children with cerebral palsy: protocol for a randomised controlled trial |
| Authors | Rachel Toovey, Adrienne R Harvey, Jennifer L McGinley, Katherine J Lee, Sophy TF Shih, Alicia J Spittle |
| Student's contribution (%) | >70% |
| Journal or book name | BMJ Open |
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| Status | ☐ Accepted and In-press ☒ Published ☐ In progress |
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| Co-author's name | Co-author's signature | Date *(dd/mm/yy)* |
|---|---|---|
| Katherine J Lee | KJLee | 24/4/19 |

# Co-author authorisation form

THE UNIVERSITY OF MELBOURNE

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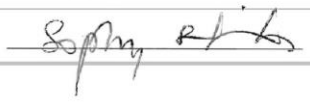
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| Co-author's name | Co-author's signature | Date *(dd/mm/yy)* |
|---|---|---|
| Sophy TF Shih |  | 23/07/19 |

# Co-author authorisation form

THE UNIVERSITY OF MELBOURNE

All co-authors must complete this form. By signing below co-authors agree to the listed publication being included in the student's thesis and that the student contributed greater than 50% of the content of the publication and is the "primary author" ie. the student was responsible primarily for the planning, execution and preparation of the work for publication.

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| Co-author's name | Co-author's signature | Date *(dd/mm/yy)* |
|---|---|---|
| Alicia J Spittle |  | 23/07/19 |

# APPENDIX X Chapters 5 and 6 ethics and governance approval certificates

## ETHICS APPROVAL

26 October 2016 (amended 07 November 2016)

Ms Rachel Toovey
Developmental Disability and Rehabilitation Research
Murdoch Childrens Research Institute

Dear Ms Toovey

**Project Title:** **Bike skills training for children with cerebral palsy: a randomised controlled trial**

---

**HREC Reference Number:** **HREC/16/RCHM/116**
**RCH HREC Reference Number:** **36209B**

I am pleased to advise that the below modification has received ethical approval from The Royal Children's Hospital Melbourne Human Research Ethics Committee (HREC).

The HREC confirms that your proposal meets the requirements of the National Statement on Ethical Conduct in Human Research (2007). This HREC is organised and operates in accordance with the National Health and Medical Research Council's (NHRMC) National Statement on Ethical Conduct in Human Research (2007), and all subsequent updates, and in accordance with the Note for Guidance on Good Clinical Practice (CPMP/ICH/135/95), the Health Privacy Principles described in the Health Records Act 2001 (Vic) and Section 95A of the Privacy Act 1988 (and subsequent Guidelines).

**HREC Approval Date:** 26 October 2016

**Participating Sites**:
Ethical approval for this project applies at the following sites:

| Site Name |
|---|
| • Melbourne Children's Campus (incorporating The Royal Children's Hospital, Murdoch Childrens Research Institute and University of Melbourne Department of Paediatrics) |
| • Monash Health |

**Approved Documents:**
The following documents have been reviewed and approved:

| Document | Version | Date |
|---|---|---|
| Protocol | 4.0 | 18 October 2016 |
| Parent/Guardian Information Statement and Consent form | 4.0 | 18 October 2016 |
| Information for child | 3.0 | 18 October 2016 |

**Site Specific Assessment**:
Site-specific governance authorisation must be obtained by each participating site before the study can commence at that site.

You are required to provide a copy of this HREC approval letter to the principal investigator at each site covered by this ethics approval to assist each site PI with obtaining governance approval to commence the project at that site.

**Conditions of Ethics Approval:**
- You are required to submit to the HREC:
  - An Annual Progress Report (that covers all sites listed on approval) for the duration of the project. This report is due on the anniversary of HREC approval. Continuation of ethics

approval is contingent on submission of an annual report, due within one month of the approval anniversary. Failure to comply with this requirement may result in suspension of the project by the HREC.
    - A comprehensive Final Report upon completion of the project.
- Submit to the reviewing HREC for approval any proposed amendments to the project including any proposed changes to the Protocol, Participant Information and Consent Form/s and the Investigator Brochure.
- Notify the reviewing HREC of any adverse events that have a material impact on the conduct of the research in accordance with the NHMRC Position Statement: *Monitoring and reporting of safety for clinical trials involving therapeutic products May 2009*.
- Notify the reviewing HREC of your inability to continue as Coordinating Principal Investigator.
- Notify the reviewing HREC of the failure to commence the study within 12 months of the HREC approval date or if a decision is taken to end the study at any of the sites prior to the expected date of completion.
- Notify the reviewing HREC of any matters which may impact the conduct of the project.
- If your project involves radiation, you are legally obliged to conduct your research in accordance with the Australian Radiation Protection and Nuclear Safety Agency Code of Practice 'Exposure of Humans to Ionizing Radiation for Research Purposes' Radiation Protection series Publication No.8 (May 2005)(ARPANSA Code).
- The HREC, authorising institution and/or their delegate/s may conduct an audit of the project at any time.

Yours sincerely



**Emma Land**
Research Ethics and Governance Officer
Research Ethics and Governance
The Royal Children's Hospital Melbourne
Phone : (03) 9345 5044
Email : rch.ethics@rch.org.au
Web : www.rch.org.au

# GOVERNANCE AUTHORISATION

7 November 2016

Ms Rachel Toovey
Developmental Disability and Rehabilitation Research
Murdoch Childrens Research Institute

Dear Ms Toovey

**Project Title:** **Bike skills training for children with cerebral palsy: a randomised controlled trial**

---

**HREC Reference Number:** **HREC/16/RCHM/116**
**SSA Reference Number:** **SSA/16/RCHM/120**
**RCH HREC Reference Number:** **36209A**

I am pleased to advise that the below modification has received governance authorisation at the Melbourne Children's Campus (incorporating The Royal Children's Hospital, Murdoch Childrens Research Institute and the University of Melbourne Department of Paediatrics).

**Governance Authorisation Date:** 7 November 2016

**Authorised Documents:**
In addition to the documents listed on the HREC approval letter, the following documents have been authorised for use at the Melbourne Children's Campus:

*No additional documents for noting.*

**Conditions of Governance Authorisation**

As Principal Investigator, you are required to:

1. Comply with the Investigator's responsibilities as outlined in the *Note for Guidance on Good Clinical Practice (CPMP/ICH/135/95).*
2. Submit a copy of this letter to the person responsible for radiation safety at this site. **This condition only applies if** the project involves exposure to ionising radiation that exceeds dose constraints, and the Medical Physicist's report has advised that the project needs to be added to the site's *Licence for Research Involving Human Volunteers* issued by the Department of Health Radiation Safety Section (for more information, visit http://www.health.vic.gov.au/radiation/). *Note: If the Medical Physicist's report has advised that the project needs to be added to the site's licence, the project cannot commence at site until you have confirmed that the project has been added to the site's licence.*
3. Notify the RGO of:
    - The actual start date of the project.
    - Any amendments to the project after these have been approved by the reviewing HREC.
    - Any adverse events involving patients at this site, in accordance with the NHMRC Position Statement: *Monitoring and reporting of safety for clinical trials involving therapeutic products May 2009*.
    - Any changes to the indemnity, insurance arrangements or Clinical Trial Research Agreement for this project. This includes changes to the project budget or other changes which may have financial or other resource implications at this site.
    - Your inability to continue as Principal Investigator or any other change in research personnel involved in this project.
    - Failure to commence the study within 12 months of the Reviewing HREC approval date or if a decision is taken to end the study at this site.

- Any other unforeseen events.
- Any other matters which may impact the conduct of the project at this site.

4. Ensure that HREC approval remains current for the entire duration of the project. Investigators undertaking projects without current Reviewing HREC approval risk their indemnity, funding and publication rights.
5. Submit an annual progress report every 12 months for the duration of the project. This report is due on the anniversary of HREC approval. Continued Governance Authorisation is contingent on receipt of an annual report by the RGO. In addition, a comprehensive final report should be submitted to the RGO upon completion of the project.

You must also abide by the following requirements:

1. Where applicable, ensure that the CTN has been electronically lodged to the TGA by the sponsor.
2. For clinical trials where the site is the Sponsor, you are required to contact MCTC to organise submission of the electronic Clinical Trial Notification (e-CTN) to the TGA. This must be completed before commencement of your project.
3. It is the Principal Investigator's responsibility to ensure that copies of the complete submitted e-CTN and TGA issued acknowledgement are included in the study Site File for the project at this site.
4. Ensure that the Clinical Trial Research Agreement (CTRA) and Indemnities (or other research agreements as applicable) are fully executed, i.e. signed by all parties; and an original version (or copy) placed in the study file.

The RGO may conduct an audit of the project at any time.

If you have any matters that arise regarding conduct of the research at this site, please ensure you contact the Research Governance Manager on 03 9345 5044.

I wish you and your colleagues every success in your research.

Yours sincerely



**Emma Land**
Research Ethics and Governance Officer
Research Ethics and Governance
The Royal Children's Hospital Melbourne
Phone : (03) 9345 5044
Email : rch.ethics@rch.org.au
Web : www.rch.org.au

MonashHealth

Research Support Services
Monash Health
Monash Medical Centre
246 Clayton Road
Clayton Victoria 3168

Level 2 I Block
Australia

Tel (03) 9594 4611
Fax (03) 9594 6306

14 November 2016

Ms Rachel Toovey
Paediatric Rehabilitation Physiotherapist
Monash Health
246 Clayton Road
Clayton VIC 3168

Dear Ms Toovey,

**Study title: Bike skills training for children with cerebral palsy: a randomised controlled trial**

**NMA Reference Number: HREC/16/RCHM/116**
**SSA Reference Number: SSA/16/MonH/379**
**Monash Health HREC Ref: RES-16-0000555X**
**Protocol Number: 36209B Version 4.0 dated 18/10/2016**

Thank you for submitting a Site Specific Assessment Form for authorisation of the above project at Monash Health.

I am pleased to inform you that authorisation has been granted for this project to be conducted at the Monash Children's Hospital Clayton campus of Monash Health.

The following conditions apply to this research project at your site. These conditions are additional to those imposed by the Human Research Ethics Committee that granted ethical approval:

The Principal Investigator is required to notify Research Support Services, Monash Health of the following:

1. Any change in protocol and the reason for that change together with an indication of ethical implications (if any)
2. Serious or unexpected adverse effects of project on subjects and steps taken to deal with them
3. Any unforeseen events that might affect continued ethical acceptability of the project
4. Any expiry of the insurance coverage provided in respect of sponsored trials
5. Discontinuation of the project before the expected date of completion, giving reasons
6. Any change in personnel involved in the research project including any study member resigning from Monash Health &/or the study team.

At the conclusion of the project or every twelve months if the project continues, the Principal Investigator is required to complete and forward an annual report to Research Support Services.

**List of Approved Documents:**

| *Document* | *Version* | *Date* |
|---|---|---|
| Monash Health Parent/Guardian Information Statement and Consent Form based on Parent/Guardian Information Statement and Consent Form Version 4.0 dated 18/10/2016 | 1.0 | 27/10/2016 |
| Monash Health Information for Child Sheet based on Information for the child Version 3.0 dated 18/10/2016 | 1.0 | 27/10/2016 |

| | | |
|---|---|---|
| Monash Health VPRS Recruitment Letter/Email based on VPRS Recruitment Letter/Email Version 2.0 dated 18/08/2016 | 1.0 | 27/10/2016 |
| Monash Health Text for CRE=CP website and enews based on Text for CRE=CP website and enews Version 2.0 dated 18/08/2016 | 1.0 | 27/10/2016 |
| Monash Health Telephone Script based on Telephone Script Version 2.0 dated 18/08/2016 | 1.0 | 27/10/2016 |

If you should have any queries about your project please contact Mr Michael Kios on 9594 4606 or via email michael.kios@monashhealth.org or Ms Deborah Dell on 9594 4605 or via email deborah.dell@monashhealth.org

Research Support Services wishes you and your colleagues every success in your research.

Yours sincerely

**Dr James Doery**
**Deputy HREC Chair**

*Attachments:*
*2 x fully executed Collaborative Group CTRA documents*

Cc: Health Information Services, Monash Health

# APPENDIX XI Chapters 5 and 6 participant information and consent form

Murdoch Childrens Research Institute
Healthier Kids. Healthier Future.

The Royal Children's Hospital Melbourne

## Information statement and consent form

| | |
|---|---|
| **HREC Project Number:** | 36209B |
| **Research Project Title:** | Bike skills training for children with cerebral palsy: a randomized controlled trial |
| **Principal Investigator:** | Rachel Toovey, PhD candidate<br>Developmental Disability and Rehabilitation Research<br>Murdoch Childrens Research Institute<br>Department of Physiotherapy, University of Melbourne |
| **Version Number:** | 4 **Version Date:** 18/10/16 |

### 1 Introduction

This is an invitation for your child to take part in this research project because they have cerebral palsy. The research project is comparing two different bike-riding training skills programs.

This Participant Information Sheet/Consent Form tells you about the research project. It explains the tests and treatments involved. Knowing what is involved will help you decide if you want your child to take part in the research. There is also an information sheet about this study for your child. We encourage you to either give this information to your child to read or for you to explain the research project to them.

Please read this information carefully. Ask questions about anything that you don't understand or want to know more about. Before deciding whether or not your child can take part, you might want to talk about it with a relative, friend or the child's local doctor.

Participation in this research is voluntary. If you do not wish for your child to take part, they do not have to. Your child will receive the best possible care whether or not they take part.

If you decide you want your child to take part in the research project, you will be asked to sign the consent section. By signing it you are telling us that you:
- Understand what you have read
- Consent to your child taking part in the research project
- Consent for your child to have the tests and treatments that are described
- Consent to the use of your child's personal and health information as described.

You will be given a copy of this Participant Information and Consent Form to keep.

### 2 What is the purpose of this research?

In general, children with CP do less recreational and physical activities than children without CP. Not doing enough physical activity in childhood can lead to chronic diseases as an adult. Bike riding is a good physical activity for children to improve health. It is also one of the most common ways to be active, a great way to get around and can be done with friends and family. We know from past research that

some children with CP do learn to ride a two-wheel bike. However, fewer children with CP can ride a bike compared to children the same age without CP. Specific training of bike riding skills may be a way to help children with CP learn to ride and get active.

Some physiotherapists and occupational therapists teach bike skills to children with CP. Other children with CP learn these skills at home with their families. Overall, there hasn't been any research to test different approaches to see if they work, or if one way is better than another. The aim of this study is to test two different bike skills training programs in children with CP to determine which method is more effective.

We hope up to 60 families will take part in this project.

This project is funded by postgraduate grants from the Murdoch Childrens Research Institute, The University of Melbourne and the National Health and Medical Research Council.

## 3 What does participation in this research involve?

Participation in this research will last for 13-19 weeks. During this time, your child will need to:

- Have a bike and helmet of their own or have access to an appropriately sized bike and helmet
- complete three research study visits at either The Royal Children's Hospital or Monash Children's Hospital
- complete a one week bike-riding skills training program
- practice bike-riding skills for 12 weeks after the training program

Study Visit 1

This is done to confirm that your child is suitable to take part in the study and collect some information before they are placed in a bike skills training program group. This visit will take up to 90 minutes.

At this visit, a physiotherapist will collect information about your child's: age, gender, residential address, medical conditions, mobility, hand function and areas of the body that are affected. We will also check that your child has an appropriate bike and helmet to train on. You will be asked questions about your child's past bike experience and your own interest in bike riding, and to complete a short questionnaire about your family including employment, education and family structure.

At this visit, your child will be given an activity monitor. This is a small device that records physical activity that is taped to the thigh with a waterproof hypoallergenic dressing. Your child will need to wear this during the day and night (including during bathing) for one week. If your child has known allergies to dressings, you will be provided with a strap for your child to wear it around their thigh. You will be provided with extra dressings (or the strap) for the activity monitor and a prepaid registered envelope to return the monitor to the research team at the end of the week.

Randomisation

Your child will be randomly placed (like flipping a coin) into one of two bike riding training skills programs so we can compare which program works best. The two groups are:

- Centre-based program
- Home-based caregiver-led program

Both training programs will take place during one week of a school holiday period, involve the child and at least one parent or caregiver, and are designed for beginner bike riding skills, rather than road safety.

Centre-based program

The centre-based program will run for 2 hours per day for 3 consecutive days at either The Royal Children's Hospital or the Monash Children's Hospital or in the parklands near to the hospitals. You and your child will join 4–6 other children and parent/caregivers and take part in the program as a group. The group will be led by a physiotherapist and an allied health assistant. Each day, children will work on bike-riding skills on their own bike. Photos or video may be taken of your child so the research team can see how well the therapists are able to deliver to the training program. You will also be given a short home program to practice skills on other days during the training week and for the following 12 weeks.

Caregiver-led program

Families in the caregiver-led program will receive written information about how to train bike skills. This information should be used to work with the child towards their bike skills goals for at least 30 - 45 minutes a day for up to 7 days. Halfway through the training week, a physiotherapist will call you to suggest different strategies and to plan for the following 12 weeks.

Study Diary

You will be asked to keep a diary of the bike riding practice your child does during the training week and for the following 12 weeks. The diary is expected to take less than 5 minutes to complete each time. You will also be asked to keep brief notes about other therapies your child receives but you will be asked not to start any new bike skills specific therapy in the follow up periods.

Study Visit 2 and 3

The second study visit will take place within one week after the training program, and the third and final study visit will be 12 weeks later. The second and third visits are expected to take 60-70 minutes.

At each visit, you will be asked about your child's bike riding goals, functional skills, self-perception, physical activity behaviour and health-related quality of life. The child's current bike skills will also be assessed. At the initial and final assessments, your child will be given the activity monitor for the child to wear during the day for another week. Again, you will be provided with a prepaid registered envelope to return the monitor to the research team at the end of the week.

There are no costs associated with participating in this research project, nor will you or the participant be paid. Car parking for all visits, including for the program if you are allocated to the centre-based group, will be funded or free parking will be provided.

If you decide that your child can participate in this research project, the researcher will contact your child's doctor and get clearance from them for your child to participate.

## 4 Does the child have to take part in this research project?

Participation in any research project is voluntary. If you do not wish for your child to take part, they do not have to. If you decide that they can take part and later change your mind, you are free to withdraw your child from the project at any stage.

Your decision that your child can or cannot take part, or that they can take part and then be withdrawn, will not affect their routine treatment, relationship with those treating them, or their relationship with the Royal Children's Hospital or the Monash Children's Hospital.

## 5 What are the possible benefits of taking part?

We cannot promise that the child will receive any benefits from this research; however, possible benefits may include achieving bike riding goals and improved bike riding skills, physical activity and participation in school, family and peer bike riding activities.

## 6 What are the possible risks and disadvantages of taking part?

It is possible your child may experience the following while participating in this study:

Injuries
It is unlikely that your child will suffer an injury, but it is a possibility if, for example, they fall off their bike. If a child injures themselves it is likely to be a mild injury, such as a graze or bruise from falling.

Discomfort
Occasionally when doing physical activity such as bike riding, people will report some discomfort during and after the activity. It is usually related to muscle strengthening. Muscle soreness during and after physical activity is usually mild to moderate. It is usually worst in the first 3 days after physical activity and then it resolves.

Inconvenience
Attending three assessments and participating in either training program does present an inconvenience to families. Both training programs require parent participation. The centre-based group bike skills training program involves travel to a centre to participate whilst the parent-led home program involves practice at home or in the community.

If you or your child become upset or distressed as a result of participation in the research, the researcher will be able to arrange for counselling or other appropriate support. Any counselling or support will be provided by qualified staff who are not members of the research project team. This counselling will be provided free of charge.

## 7 Can the child have other treatments during this research project?

Whilst your child is participating in this research project, they should not do other physiotherapy or occupational therapy related to training bike skills. It is important to tell us about any treatments or medications your child may be taking, including other physiotherapy, occupational therapy or Botulinum Toxin-A injections. You should also tell us about any changes to these during the child's participation in the research project.

## 8 What if I withdraw the child from this research project?

If you decide to withdraw your child from the project, please notify a member of the research team before you withdraw them. This notice will allow that research team to further discuss any health risks or special requirements linked to withdrawing.

If you do withdraw the participant during the research project, personal information already collected will be retained to ensure that the results of the research project can be measured properly and to comply with law. If you do not want them to do this, you must tell them before your child joins the research project.

## 9 What happens when the research project ends?

When the research project ends, the results will be analysed and reported. We will send you a summary of the results of the whole study. You may like to share these results with your child.

## 10 What will happen to information about the child?

By signing the consent form you consent to the collection and use of personal information about your child for the research project. Any information obtained in connection with this research project that can identify your child will remain confidential. Your child's information will only be used for the purpose of this research project and it will only be disclosed with your permission, except as required by law. All information will be stored securely in the Developmental Disability and Rehabilitation Research Group at the Murdoch Childrens Research Institute.

Only the research team involved in the project and the Ethics Committees at the Royal Children's Hospital, Monash Health, the University of Melbourne and Deakin University can have access to this information.

The stored information will be re-identifiable. This means that we will remove identifying information such as you and your child's name and give the information a code name. Only the research team can match you and your child's name to their code name, if it is necessary to do.

We are required to keep information collected as part of this research project for a certain length of time. Data will be kept until the 25th birthday of the youngest participant. Following this time, it may be confidentially destroyed.

In accordance with relevant Australian and/or Victorian privacy and other relevant laws, you have the right to request access to the participant's information collected and stored by the study team. You also have the right to request that any information with which you disagree be corrected. Please contact the study team member named at the end of this document if you would like to access the participant's information.

It is anticipated that the results of this research project will be published and or presented in a variety of forums. In any publication and/or presentation, information will be provided in such a way that the child cannot be identified, except with your permission. The results of this research will be used by Rachel Toovey to get a PhD degree.

## 11 Who has reviewed the research project?

All research in Australia involving humans is reviewed by an independent group of people called a Human Research Ethics Committee (HREC). The ethical aspects of this research project have been approved by the HREC of The Royal Children's Hospital and Monash Health.

This project will be carried out according to the National Statement on Ethical Conduct in Human Research (2007). This statement has been developed to protect the interests of people who agree to participate in human research studies.

## 12 Further information and who to contact

The person you may need to contact will depend on the nature of your query. If you want any further information concerning this project:

*Information contact person:*

| | | | |
|---|---|---|---|
| **Name** | Rachel Toovey | **Position** | Principal Investigator |

| | | | |
|---|---|---|---|
| **Telephone** | (03) 9936 6088 | **Email** | Rachel.toovey@mcri.edu.au |

If the participant has any medical problems which may be related to their involvement in the project (for example, any side effects), you can contact:

*During intervention contact person:*

| | | | |
|---|---|---|---|
| **Name** | Dr Adrienne Harvey | **Position** | Associate Investigator |
| **Telephone** | (03) 9345 5522 ext 57540 | **Email** | Adrienne.harvey@rch.org.au |

If you have any complaints about any aspect of the project, the way it is being conducted or any questions about being a research participant in general, then you may contact:

Reviewing HREC approving this research and HREC Executive Officer details

| | |
|---|---|
| **Reviewing HREC name** | The Royal Children's Hospital |
| **HREC Executive Officer** | Alexandra Robertson, Director |
| **Telephone** | (03) 9345 2044 |
| **Email** | Rch.ethics@rch.org.au |

## CONSENT FORM – Parent/Guardian

| | |
|---|---|
| **Research Project Title:** | Bike skills training for children with cerebral palsy: a randomized controlled trial |
| **HREC Project Number:** | 36209B |
| **Principal Investigator** | Rachel Toovey |
| **Location** | Royal Children's Hospital [or Monash Children's Hospital] |
| **Version Number:** | 3 **Version Date:** 13/09/16 |

- I have read, or someone has read to me in a language that I understand, the information statement version listed above and I understand its contents.
- I believe I understand the purpose, extent and possible risks of my child's involvement in this project.
- I voluntarily consent for my child to take part in this research project.
- I have had an opportunity to ask questions and I am satisfied with the answers I have received.
- I understand that this project has been approved by The Royal Children's Hospital Melbourne Human Research Ethics Committee and the Monash Health Ethics Committee and will be carried out in line with the National Statement on Ethical Conduct in Human Research (2007) – including all updates.
- I freely agree to the child participating in this research project as described and understand that I am free to withdraw them at any time during the research project without affecting their future health care.
- I understand I will receive a signed copy of this document to keep.
- I understand that, if I decide to discontinue the child's study treatment, a request may be made for them to attend follow-up visits to allow collection of information regarding their health status.

OPTIONAL CONSENT

| | | |
|---|---|---|
| ☐ I do | ☐ I do not | Consent to the storage of my child's information for use in **future ethically approved** research related to cerebral palsy |

Child's Name

| Parent/Guardian Name | Parent/Guardian Signature | Date |
|---|---|---|
| Name of Witness to Parent/Guardian's Signature | Witness Signature | Date |

Declaration by researcher: I have given a verbal explanation of the research project, its procedures and risks and I believe that the parent/guardian has understood that explanation.

| Research Team Member Name | Research Team Member Signature | Date |
|---|---|---|

Note: All parties signing the Consent Form must date their own signature.

# APPENDIX XII Chapters 5 and 6 therapist training manual (relevant sections)

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## CP-BIKE RCT

## Bike skills training for children with CP: a randomised controlled trial

## HREC/16/RCHM/116
## RCH HREC #36209

## Therapist training manual

Version 1, 27 October 2016

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**CONFIDENTIAL**

This document is confidential and the property of the Murdoch Childrens Research Institute and The University of Melbourne. No part of it may be transmitted, reproduced, published, or used without prior written authorisation from the institution.

**Statement of Compliance**

This document is a training manual for a research project. This study will be conducted in compliance with all stipulation of the study protocol, the conditions of the ethics committee approval, the NHMRC National Statement on ethical Conduct in Human Research (2007) and the Note for Guidance on Good Clinical Practice (CPMP/ICH-135/95).

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information (Appendix v) either via email or express post. If received prior to the intervention period families will be explicitly asked not to open the information until the start of the one week period of training during the school holidays. The AI (Harvey or Spittle) will then inform the trained VPRS therapists of the allocation of each participant as well as the following details

- Child's name
- Child's age
- Parent's name
- Contact details (address and contact phone)
- GMFCS, MACS
- Highest level of bike riding skill
- Previous bike riding practice
- Bike-specific goal
- Any other clinically relevant detail

Following randomisation the participants will be asked to contact the VPRS therapists in case of issues (e.g. non-attendance) between randomisation and the intervention period and during the intervention period. During the follow up period participants will be asked to contact the AI in case of issues (e.g. adverse events).

## II. The intervention group

### i. Overview of the novel task-specific program

Participants randomised to the intervention group will participate in a novel bike skills training program. The intervention has been informed by dynamic systems theory (14), motor learning principles (13) and research and evaluation specific to training bike skills conducted by the research team (including the systemic review and case series (12, 15) The intervention involves seven key components: group-based, therapist-led, intensive, task-specific, goal-directed, parent involvement and ecological setting.

1. **Task-specific**: The motor learning approach will be task-specific (21) and informed by the dynamic systems theory and principles of motor learning. The dynamic systems theory considers the dynamic interaction between systems including the task, the child and the environment to achieve motor skills in a task-specific context (14) . Each of these systems is considered at each stage of the motor learning process. Initially new motor tasks are scaffolded, so that the participant will always actively complete at least part of the task. As performance improves, the motor challenge is increased by altering the task or environment to encourage problem solving. This may include modifying the bike (E.g. seat height, location of the brakes, straps for hand or feet) in order to achieve each progression of the skill/s. In the case any modifications are made to the bike, therapists will document details of this on the therapist's attendance log (Therapists will keep a log per participant). Once a motor skill is acquired, variability of practice in terms of task difficulty and environmental challenge is introduced to increase the complexity and generalisability of the skill. Amount and type of feedback will be guided by participant preference but at a minimum verbal feedback, either

knowledge of results or knowledge of performance will be given one in every four trials of a new skill (22), for example getting on and off the bike.

2. **Group-based**: the intervention will be delivered to groups of four to six participants. There is evidence to suggest group-based rehabilitation programs improve functional skills, self-perceived performance and cost-effectiveness of treatment as much, or more than individual therapy (23).
3. **Therapist-led:** In Australian clinical practice, physiotherapists and occupational therapists generally implement therapy to improve motor skills or daily task performance such as learning to ride a two-wheel bike for children with CP. Each program will be conducted by at least one physiotherapist and one other therapist (physiotherapist, occupational therapist or allied health assistant). There will be a minimum ratio of one therapist to three children participants in each group. All therapists will be employed by VPRS and trained in the intervention protocol as outlined in section 6.2.1 and the same therapists will lead the 3 consecutive days program of each.
4. **Intensive:** Each program will run for two hours per day over three consecutive days during one week of the school holiday period. This intensity is supported by the motor learning literature in particular the benefits of repetitive practice in the skill acquisition phase (24). This intensity allows for repetitive practice (25), including repetitive practice in the home environment following the program and has been supported by parent evaluation of the intensive program delivered as part of the pilot case series (15). Breaks from physical activity will be offered at least every 30 minutes and families can request additional rests. Participants will also be given a home program of one to three bike skills practice exercises following each session and encouraged to practice these up to 30 minutes per day during the weeklong intervention period and three to five bike skills exercises to practice when able in the twelve week follow up period.
5. **Goal-directed:** The Goal Attainment Scale (GAS) will be used as an outcome measure and as a process for setting goals related to bike skills training. The therapists delivering the intervention will be aware of each participant and family's goals and individualise components of their program to work towards these goals. There is evidence to suggest interventions that are goal-directed improve gross motor function more than those that are not (26). Goal setting is a key component of paediatric rehabilitation and been well established in the literature (17).
6. **Parent or caregiver involvement:** At least one parent or caregiver will be required to attend each session of the program for each participant. Parent involvement and education is recognised as a key component in family-centred practice (27). It facilitates a partnership between the therapist and parent or carer towards achieving a common goal; that of the child. Parents will be coached by the therapists regarding approaches to motor learning, including gradually increasing the difficulty of the task whilst ensuring this intersects with success. Parents will be provided verbal guidance regarding strategies and safety of practice in the home environment. Parents will be asked to bring along water and a snack for their child.
7. **Ecological setting**: When possible the program will be conducted in outdoor recreation or community reserves at or in close proximity to the MCH and RCH. This aligns with dynamic systems theory and task-specific training in terms of the role the environment has in promoting motor learning. Different surfaces and gradients will be available to individualise

the environment based on each participant's stage of motor learning and to promote successful problem solving. All program settings will be closed to cars and quiet in terms of other people using the space. Participants will be encouraged to practice outside of the program in similar environments and advised to avoid practice on roads, busy bike paths or other risky environments (e.g. very steep hills) during the intervention of follow up periods.

### ii. Specific components of the novel task-specific program

**Task-specific progressions**

Skill progression should be tailored to the individual's skill level but may follow the following structure for a child who has very little experience or low skills level on the bike:

1. Bike fit and adjustments
   a. Remove the training wheels (if on) and pedals – this can be done with a spanner where possible (occasionally pedals get rusty and are difficult to remove)
   b. Helmet on and appropriately fitted
   c. Setting the seat height– start off with the seat at a height where the child's feet can comfortably touch the ground flat, generally this is the groin to the heel measurement
   d. Handle bar height and angle – start with what is easiest for the child's UL function and to allow ease of turning.

2. Familiarity with the bike and basic handling skills
   a. Locate brakes
   b. Picking bike up off flat ground and putting it down
   c. Walking next to the bike
   d. Getting on and off the bike

3. Basic balance skills (generally pedals still off first and then repeat all steps with pedals on)
   a. Walking on bike
   b. Scooting on bike
   c. Coasting or gliding on bike

4. Pedalling
   a. Reattach the pedals if they were removed.
   b. Have child put foot on one pedal only and repeat the coasting as above. Do for both sides.
   c. Now have the child put one foot on each pedal and coast (no actual pedalling yet)
   d. Practice pedalling whilst already coasting
   e. Once child becomes more comfortable with pedalling, gradually increase the height of the seat to the appropriate height for the child.
   f. Introduce gentle braking as the bike starts to slow down.

5. Riding in a straight line
   a. Therapist/parent pushes off through the seat, and then instructs the child to pedal.
   b. Encourage child to look straight ahead
   c. Don't turn head or shoulders

6. Stopping
   a. Encourage child to apply brakes gently, and at the same time (if bike has front and rear brakes)

b. If child has difficulty co-ordinating 2 brakes, encourage the back brake as the main brake to focus on using

7. Starting
   a. Often is one of the last skills to learn
   b. To start from a standstill, have one pedal at 2 o'clock position
   c. Assistant to support through the seat
   d. Other foot on the ground and then just above until balanced and going.

8. Turning
   a. Encourage child to slow down before turning
   b. Turning is a combination of steering and leaning

9. Using gears
   a. If applicable, should be left to last, and only for higher level/more competent riders
   b. Lower gears are best for going uphill, whereas higher gears for downhill riding

**Motor learning strategies**

Underlined strategies should be used more often in the skill acquisition phase.

*What the therapist says*

1. Instructions
   - Direct attention to object/environment
   - Direct attention to body
2. Feedback
   - Ask vs tell
   - Feedback related to movement performance
   - Feedback related to movement results
   - Frequency of feedback (minimum 1 in every 4 trials)
   - Self-controlled vs constant or no control
   - Feedback related to what was done well vs what was done poorly
3. Analogy learning – Linking to another task or setting (e.g. it's like riding a horse)
4. Encourage mental practice
5. Cognitive approaches – Goal, Plan, Do , Check + key words

*What the therapist does*

1. Observational learning – this involves the demonstration of the task by the therapist, another child or a parent to aid the child undertaking the task
2. Physical guidance
3. Error reduced vs Trial and error approached
4. Task modification
5. Environmental modification
6. Dual task learning
7. Home program prescribed – 1-3 exercises per participant (see below)

*Practice*

1. Repetitive
2. Whole practice vs part practice
3. Varied vs constant practice
4. Random vs blocked
5. Progressive (as above)

**Breaks**

In moderate whether (i.e. 10 – 30 degrees Celsius) breaks from physical activity will be offered at least every 30 minutes and families can request additional rests. See section 3 Risk identification and management (p 20) and Appendix viii Risk Identification Checklist for excessive weather condition plan.

**Home practice**

Participants in the intervention group will also be given one to three bike skills practice exercises by the VPRS therapists following each session and encouraged to practice these up to 30 minutes per day during the weeklong intervention period and three to five bike skills exercises to practice when able in the 12 week follow up period. They will document this practice in Appendix vi Intervention group participant diary.

**Equipment required**

- Cones – small and large if possible
- Tool box - spanners and Allen keys (various sizes)
- Felt strips, scissors and Velcro straps to make hand and foot straps
- Labels and textas / pens
- Sunscreen
- First aid kit*
- Mobile phone*
- Clipboard
- Attendance and adverse events log (Appendices viii and ix)

*If going off RCH or Monash grounds

**Basic modifications**

Foot straps

Hand straps

**Relevant appendices**

Appendix vi Intervention group participant diary

Appendix vii Intervention group attendance log

## III. The comparison group

### i. Overview of the parent-led home program

Preliminary analysis of data collected through a national online survey of over 150 physiotherapists and occupational therapists who work with children with CP indicates that frequency, intensity or type of bike skills training is not uniform in typical practice. Further, other research conducted by members of the research team suggests that whilst the majority of ambulant children with CP (GMFCS I-II) are currently not able to ride a two-wheel bike, many of those who can ride learnt in informal settings with their families (8). There also is good evidence to support home-based therapy programs involving parent education for goal attainment in children with CP (5, 16).

Participants randomized to the comparison group will undertake a parent-led home bike skills training program. Following baseline assessment and randomisation parents of these participants will receive written general information (Appendix v) on training bike skills either in person or via email dependent on consenting and baseline assessment location. Families will receive this information at the start of the one-week period of training during the school holidays. Parents will be encouraged to work with their child on two-wheel bike skills goals guided by the written information. This information involves:

1. **Intensity**: Families will be encouraged to practice at least 30 - 45 minutes on each of the 7 days of the one-week period.
2. **Safety**: Families will be encouraged to practice in settings that are closed to cars and quiet in terms of other people using the space. They will also be advised to perform a risk assessment of the location prior to practice in a location and avoid practice on roads, busy bike paths or risky environments (e.g. very steep hills). Other factors such as weather and adequate hydration are also included.
3. **Appropriate bike and helmet fit:** Information regarding fitting the bike and helmet to the child for skill development, safety and potentially useful modifications

Trained VPRS therapists will also telephone families in the comparison group between 3-5 days into the one-week home program period. This phone call will involve asking the parents how the home program is going and providing general advice regarding practice for the remaining 2-4 days of the program period.

**Relevant appendices:**

Appendix v Information for families in comparison group

Appendix vii Comparison group participant diary

Appendix x Comparison group telephone log

### ii. Specific components of the parent-led home based program

All families randomised to the comparison group will receive a phone call from a trained VPRS physiotherapist on the Wednesday or the Thursday of the training week. In the case the family does not answer on the first attempt, the VPRS therapist will attempt to call each family a maximum of three times. The telephone discussion should be general in nature and approx. 10-15 mins in length. It may involve the following questions

1. How is the child going with the home based program?
2. How much bike riding practice have they done so far?
3. Where are you practicing bike riding? I.e. park, home, path?
4. What are the challenges you are facing?
5. What are you finding works well?

**Dos** - The advice given can involve the following aspects:

- Intensity of training – duration of each session and breaks provided
- Location of training – safety aspects
- Learning environment – encourage a positive learning environment and allow the child time to process and problem solve

**Don'ts** - The advice should not include:

- Specific instructions around skill progressions (i.e. take pedals off)
- Specific instructions about types of feedback
- Specific instructions about task or environmental modifications or other motor learning strategies

Trained therapists should document the length of time and the content of the phone call for each participant on Appendix x Comparison group telephone call log

# 3 Risk identification and management

## I. Risk identification

Risks should be identified at the commencement of the group-based program each day. This can be done by completing Appendix viii Risk Identification Checklist. If any risks are identified, the following management strategies should be utilised.

## II. Risk management

Manual handling

- 2 persons to provide assistance where able
- Safe assisting postures including core support and using lower limbs
- Basic manual handling education to parents
- Safe transport of equipment

Avoiding occupational violence

- Supportive and positive learning environment
- Providing positive feedback and encouragement
- Discuss behaviour with child and caregiver
- Have regular breaks in particular if child or caregiver is frustrated with progress

Biological exposure

- In the case snakes, large amounts of bugs or pollutants are present in the training environment, the therapist should relocate the program to an area well away from the risk

Physical conditions

- In the case of cold (5 – 10 degrees Celsius), wet or hot (> 31 - 35 degrees Celsius) weather, at least 1 hour of that day's session should be conducted in an indoor therapy space.
- If the temperature is below 5 degrees Celsius or exceeds 35 degrees Celsius the entire 2 hour session should be conducted in an indoor therapy space.
- Sunscreen will be included in the therapist first aid kit and families will be advised to bring adequate water and sunscreen to the programs.
- Therapists should also provide information regarding safe weather conditions for practicing at home to parents.

Stress conditions

- If a participant becomes upset or distressed as a results of participation, counselling or other appropriate support will be arranged through the Victorian Paediatric Rehabilitation Service.
- The therapist should advise Associate Investigator Adrienne Harvey and instigate an internal referral to the VPRS site psychologist.

Mechanical hazards

- Therapists delivering bike skills training in parklands area proximate to the RCH or MCH should conduct an assessment of any stable mechanical hazards (walls, seats, ditches, poles, steep hills etc.)
- Ensure the training is conducted in open spaces where able and ensure adequate space is available for activities to avoid dynamic hard issues (other participants, the public)

Slip, trip and fall

- For components of the training involving pedalling without training wheels, at least one parent or therapist should be close by to each participant.
- Therapists should involve falls minimisation strategies in the training including safe progressions and assistance

Dealing with the public

- Therapists delivering the program should inform any other members of the public using the parklands area of the program and the space requirements.
- Therapists should remove themselves from the situation if they feel threatened and call the Police (000) in the case they fear for their own or the participants' safety (once they have removed themselves from the situation).

Medical concerns

- Regular breaks to prevent physical stress should be provided in the intervention group
- In the case of an adverse event on study site, follow site specific medical emergency procedures
- In the case of an adverse event off study site, therapists should call 000 or advise the child and family to attend the emergency department or GP (whichever is more appropriate)

**Therapists should also follow any site-specific procedures if leaving the hospital grounds (i.e. notify administration staff or write names of staff on board, carry mobile or Ascom phone and First Aid Kit)**

# 4 Adverse events

## I. Definitions

*Unanticipated Problems:* Unanticipated problems involving risks to participants or others to include, in general, any incident, experience, or outcome that meets all of the following criteria:

- unexpected in terms of nature, severity, or frequency given (a) the research procedures that are described in the protocol-related documents, and (b) the characteristics of the participant population being studied;
- related or possibly related to participation in the research; and
- suggests that the research places participants or others at a greater risk of harm (including physical, psychological, economic, or social harm) than was previously known or recognised."

*Adverse Event (AE):* Any untoward medical occurrence in a patient enrolled into this study regardless of its causal relationship to study/interventions. I.e. any event the therapist deems requires medical assistance.

*Serious Adverse Event (SAE):* An SAE is defined as any AE that:

- results in death; or
- is immediately life threatening; or
- requires inpatient hospitalisation; or
- results in persistent or significant disability/incapacity; or

Important medical events will be considered an SAE when, based upon appropriate medical judgement, they may jeopardise the patient and may require medical or surgical intervention to prevent one of the outcomes listed in this definition.

## II. Reporting procedure

Therapists should document any unanticipated problems or AEs on Appendix vii Intervention group attendance log or Appendix x Comparison group telephone call log. AEs that occur during the intervention period (in the intervention group or reported over the phone in the comparison program) should be reported to Al Harvey at the earliest possible occasion. Any serious adverse events (SAEs) need to be reported to Al Harvey within 24 hours of the event or notification.

The description of each AE reported to Al Harvey should include:

- A description of the AE;
- The onset date, duration, date of resolution;
- Severity (mild, moderate or severe);
- Seriousness (i.e. is it an SAE?);
- Any action taken, (e.g. treatment, follow-up tests);
- The outcome (recovery, death, continuing, worsening);
- The likelihood of the relationship of the AE to the study intervention (Unrelated, Possible, Probable, Definite).

# APPENDIX XIII Chapters 5 and 6 comparison group written information for parents

**Parent-led home bike skills training program**

You and your child have been randomly placed in the home-based caregiver-led training program. This information will help guide you through the training week, with pointers around safety, good spots to practice and bike fit.

You will have set goals for the training week with the researcher at your baseline assessment. To work towards these goals, we encourage your child to use their bike for at least 30 - 45 minutes a day for up to 7 days. You have also been given a 'diary' to document practice done each day of the training week and each week during the 12-week follow-up period.

A physiotherapist from the Victorian Paediatric Rehabilitation Service will telephone you in the middle of the training week. If you have not heard from them by Thursday, please call Lauren Thompson-Glennen on (03) 9594 4001.

## Before getting on the bike

**1. Safety first!**

- Finding the right location

  There are a few things to consider when finding a good location for practicing bike riding. If possible, find somewhere away from cars that is not too busy with other people. A variety of ground surfaces (grass, gravel, footpath) may help build confidence and then skills. When you arrive at a location, take a quick look around to check for hazards such as benches, other people, steep hills, long grass and animals/bugs. Make sure you have a clear place for practicing and enough space to get some speed up on the bike! We advise you stay away from practicing on roads but if you do practice on footpaths keep the following information from VicRoads in mind:

  "You can ride on a footpath if you are: a child under the age of 12, an adult supervising a child under the age of 12 or you have a medical certificate that says your child has a disability that makes it difficult to ride on the road"

- Weather and water

  Bike riding is a physical activity and can be tiring for both you and your child, especially in inclement weather. In warm weather, avoid peak heat times (10am – 3pm) and if the temperature is over 35 or under 5 degrees Celsius, avoid practicing outdoors. Some bike handling skills may still be practiced indoors. If it is over 30 degrees we advise no longer than 30 minutes at time practicing outdoors. Whatever the weather, make sure you and your child take regular breaks, have adequate water and wear sun protection.

- Safe manual handling

  Teaching a child to ride a bike often involves providing physical assistance. This can be in the form of steadying the bike or providing a push off to get up enough speed. Where possible, avoid bending from your back – instead bend your knees and keep your body stable. As much as possible

let your child handle the bike. This includes picking the bike up, moving it around and steering. This not only gives you a break but the practice is great for their skills.

- Minimising risk of falls

  Many of us will have had a few falls from our bike especially when we were learning. There are ways to minimise the risk of falls.

  1. Pick a good spot to practice - an open space with as few distractions as possible. Practice on soft surfaces such as grass to begin, with as it can help build up your child's confidence.
  2. How much help you give (if required) can make a difference. Stay close, but give your child enough room to get on and off the bike safely. If you hold the bike from the seat, rather than the handle bars, this can also make the bike feel more balanced for a child. At the beginning stages of learning, or for older kids and adolescents, it can be useful to have two people available to provide assistance.
  3. Start by practicing skills your child can achieve. Gradually building their skills also helps to make sure they are not trying something that is too hard for them.

- Avoiding other injuries and physical or emotional stress

Bike riding is like any exercise - it can cause some muscle soreness and tiredness. To make sure this doesn't affect your child's ability to practice, plan your sessions. Take regular breaks (at least every half hour) and avoid long periods of practice on the bike in one go (no more than a few hours in one day). Teaching and learning skills can be a stressful experience. This can be avoided by providing positive feedback.

**2. Fitting the bike and helmet to the child**

Most bikes require just a couple of simple tools to get the right fit – all you will need is an allen key and/or a spanner.

- Look at seat height – measure the length of your child's leg from inside groin to heel to get indication of height of seat. It can be best to start with the seat at a height that means they can touch both feet flat on the ground.
- Handle bar height and angle – start with what is easiest for your child's arm function and to allow ease of turning. For children with affected arm/s this may mean adjusting the angle so the handle bars are closer to the body.
- Helmet fit – you may need to adjust your child's helmet to get a good fit. A good fit is when the helmet sits snuggly on your child's head and they can get two fingers between the strap and their chin.

## Time to get riding!

**Don't forget that your child should always wear a helmet!**

**Things to discuss with the physiotherapist**

- It may be helpful to keep a list of challenges you and your child are facing.
- You can discuss these with the physiotherapist on the telephone in the middle of week.
- The physiotherapist may also provide you with a few activities for you to do with your child to do for the 12 week follow up period. .

If your child experiences any falls, stress, injury or discomfort during the training week or the 12 week follow up period, whether or not it is related to the bike riding, please contact*: Dr Adrienne Harvey, Ph: (03) 9345 5522 ext 57540, E: Adrienne.harvey@rch.org.au*