You Can Ride Too! An Exploration of the Guided Discovery of Two-wheeled Cycling Skills by Youth with Intellectual Disabilities

by

Janine Halayko

A thesis submitted in partial fulfillment of the requirements for the degree of

Master of Science

in

Rehabilitation Science- Physical Therapy

Rehabilitation Medicine

University of Alberta

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Abstract

Learning to ride a bike is an important milestone in the life of a child, yet many children with disabilities never have the opportunity to experience this success. Few studies have examined how to teach this skill to individuals with intellectual disabilities, with techniques that do not use specialized bicycles. The main purpose of this thesis was to explore the applicability of Cognitive Orientation to daily Occupational Performance (CO-OP) for teaching cycling to individuals with intellectual disabilities. Two studies were completed: a single case multiple baseline design across seven participants (ages 10-19), and an in-depth analysis of one of the cases. In the first study the seven youth—six with a diagnosis of Down syndrome, one with Smith-Lemli-Opitz syndrome-were videotaped over the course of the baseline, intervention and follow-up phases. Cycling skills and the maximum distance and time ridden were assessed and analyzed using the non-overlap of all pairs (NAP) method. Completion of the CO-OP intervention coincided with significant improvements in all measures for six of the seven participants. At follow-up, cycling distances for these 6 youth ranged from 31-1756m, time cycled extended from 11 seconds to over 9 minutes, and an average of over 10 of 20 independent cycling skills were gained. One participant did not learn how to ride and one did not participate in two-wheeled cycling after follow-up. In the second study involving the oldest participant with Down syndrome, the youth's trajectory of learning and cycling skill acquisition was described along with the key features of CO-OP used to facilitate his learning. The results of these studies provide proof of principle that CO-OP may be an effective way to teach cycling to individuals with intellectual disabilities. Further evaluation is warranted to determine its potential use with others with intellectual disabilities or when teaching other skills.

Preface

This thesis is an original work by Janine Halayko. The research project, of which this thesis is a part, received research ethics approval from the University of Alberta Research Ethics Board, Project Name "Learning to Ride a Two-wheeled Bicycle", No. Pro00036251, April 10, 2013. No part of this thesis has been previously published.

Dedication

This thesis is dedicated to my family: My incredible husband Jason who has believed in me and supported me every step of the way; my wonderful children Cole, Ashton and Alexa, whose hugs have kept me going; my mother who continues to inspire me to make the world a better place; my father who is always there when it is important; and my sisters Michele and Laura who have done numerous things to help make this journey a little easier. I cannot thank you enough.

'Life is like riding a bicycle. To keep your balance, you must keep moving.'

—Albert Einstein

Acknowledgements

I am deeply indebted to all three of my amazing supervisors, whose support has been invaluable. Dr. Helene Polatajko, thank you for motivating me all those years ago and for guiding me to discover how much the children we teach can contribute to the process. I have appreciated your flexibility with this unconventional arrangement, our discussions over Skype and the encouragement to see the bigger picture. Dr. Veronica Smith, I am grateful for your positive reinforcement that contributed to my decision to return to school, and for your expertise in single case research. Finally, I would like to thank Dr. Joyce Magill-Evans, for helping me narrow down my focus, inspiring me to find balance between home and work and for being such a calming and supportive influence on my chaotic life. For this and all you did to make things just that much easier-sharing your research assistant time, recruiting students to help with filming, modeling your editing wizardry, presenting this research in Japan, and being a positive support— I cannot thank you enough. I would also like to acknowledge the Edmonton Public School Board (EPSB) who provided financial assistance and the time off I needed to complete my coursework, and the Women and Children's Health Research Institute for their summer studentship and immersion into the world of community-based research. Finally, I would like to thank the amazing families and children that I have met through the study and through my work with You Can Ride 2. I am sure that I learned as much or more from them as they did from me, and I hope others wishing to teach their children to ride will hear their voices through this work and the knowledge translation that follows.

Table of Contents

List of Tables	viii
List of Figures	ix
List of Appendices	x
Chapter 1: Introduction	1
Purpose and Direction of Thesis	3
Background	
Cycling Programs	5
CO-OP: An Alternate Teaching Technique	
Motor Learning Theory	
The Impact of Cognition on Learning and Motor Performance	
The Potential Use of CO-OP for Individuals with Intellectual Disabilities	
Table 1 Summary of Studies Using iCan Bike Equipment	19
Chapter 2: "Look Ma, I did it!": Enabling Two-wheeled Cycling for Youth with	Intellectual
Disabilities Through Strategy Use	36
Abstract	36
Methods	40
Participants and Settings	
Experimental Design	
Materials	
Dependent Measures and Interobserver Agreement	
Intervention Description and Treatment Fidelity	
Results	47
Time and Distance Cycled	
Cycling Skills	
Parent Ratings	
Discussion	50
Applicability of the approach	51
Contributions	52
Limitations and Future Directions	54
Table 2 Participant Characteristics	55
Figure 1. COPM Parent Pre and Post Ratings of Performance and Satisfaction	55
Figure 2. Distance and Time Cycled	56
Figure 3. Independent Sub-skills at Baseline and Follow-up	57
Figure 4. Skill Checklist Scores at Baseline and Follow-up	57
References	58
Appendix A Cycling Skill Checklist	63

syndrome: An in-depth case study	
Abstract	
Method	70
Study Design	
Participant and Settings	
Procedure	
Data Collection and Analysis	
Results	74
Learning Trajectory at Baseline	74
Learning Trajectory During Intervention	74
Learning Trajectory at Follow-up	75
Key Features of Learning with CO-OP	
Discussion	79
Conclusion	81
Table 3 Ratings of Performance and Satisfaction	83
Table 4 Focus and Guided Discoveries in CO-OP Intervention	83
Figure 5. Cycling Skill Progression.	84
Figure 6. Distance and Time Cycled (Pre and Post Intervention)	84
Table 5 Domain Specific Strategies Used	85
References	86
Appendix B Domain Specific Strategy Descriptions	90
Chapter 4: On a roll: Discussion, Implications and Future Direction	- 01
, , ,	S
I Can Bike: Advantages and Limitations	s91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative	91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings	91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective	91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths	91 91 93 93 101 104
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation	91 93 93 93 93 101 104 105
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research	s91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research Recommendations	s91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research Recommendations Conclusion	s91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research Recommendations Conclusion References	s91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research Recommendations Conclusion References Bibliography	s91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research Recommendations Conclusion References Bibliography Appendices	s91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research Recommendations Conclusion References Bibliography Appendices Appendices	s
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research Recommendations Conclusion References Bibliography Appendices Appendix C Ethics Approval Appendix D Pre-Study Parent Questionnaire	s91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research Recommendations Conclusion References Bibliography Appendices Appendix C Ethics Approval	s
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative	s91
I Can Bike: Advantages and Limitations CO-OP: a Feasible Alternative Summary of Research Findings An ICF Perspective Limitations and Strengths Knowledge Translation Future Research Recommendations Conclusion References Bibliography Appendices Appendix C Ethics Approval Appendix D Pre-Study Parent Questionnaire Appendix E Pre-Study Youth Questionnaire Appendix F Post-Intervention Parent Questionnaire Appendix G Post Follow-up Parent Questionnaire	s

Chapter 3: Guiding discovery of two wheeled cycling strategies, for a youth with Down

List of Tables

Table 1:	Summary of studies using iCan Bike equipment	.19
Table 2:	Participant Characteristics	55
Table 3:	Ratings of Performance and Satisfaction	.82
Table 4:	Focus and Guided Discoveries in CO-OP Intervention	.82
Table 5:	Domain Specific Strategies Used	.84

List of Figures

Figure 1:	COPM Parent Pre and Post Ratings of Performance and Satisfaction	55
Figure 2:	Distance and Time Cycled	.56
Figure 3:	Skill Checklist Scores at Baseline and Follow-up	.57
Figure 4:	Independent Sub-skills at Baseline and Follow-up	.57
Figure 5:	Cycling Skill Progression	.83
Figure 6:	Distance and Time Cycled (Pre and Post Intervention)	.83

List of Appendices

Appendix A:	Cycling Skill Checklist	63
Appendix B:	Domain Specific Strategy Descriptions	89
Appendix C:	Ethics Approval	131
Appendix D:	Pre-Study Parent Questionnaire	132
Appendix E:	Pre-Study Youth Questionnaire	133
Appendix F:	Post-Intervention Parent Questionnaire	134
Appendix G:	Post Follow-Up Parent Questionnaire	135
Appendix H:	One Month Follow-Up Parent Questionnaire	136

Chapter 1: Introduction

Most children enjoy riding bicycles. In Canada, an estimated 88% of children own a bicycle and over 75% consider cycling as among their favorite recreational activities (Cragg, Cameron, & Craig, 2006; Craig, Cameron, Russell, & Beaulieu, 2001). However, there is a significant discrepancy in cycling participation between children with and without disabilities. Steele (1996) found that only 16% of Canadian youth with a physical disability cycled at least weekly compared to 53% of youth without a disability. In some cases this discrepancy may be due to a lack of access to a suitable bicycle. Adapted bicycles allow for cycling participation for almost all children, regardless of their disability, but they are often cost prohibitive (Pickering, Horrocks, Visser, & Todd, 2013), costing upwards of \$2000. In addition, many families and children report that riding an adapted bicycle is less desirable than riding a conventional bicycle. In contrast, two-wheeled cycling is seen as both a skill that can increase social and community participation opportunities and one that has numerous health benefits (MacDonald, Jaszewski, Esposito, & Ulrich, 2011; Menear, 2007).

The discrepancy in cycling participation between those with and without disabilities lies in the challenge of mastering a two-wheeler. While most children learn to ride a bicycle around the age of 5 or 6 (Hansen, Eide, Omenaas, Engasaeter, & Viste, 2005; Klein, McHugh, Harrington, Davis, & Lieberman, 2005), children with disabilities take much longer to achieve mastery (MacDonald et al., 2012), if this comes at all (Ulrich, Burghardt, Lloyd, Tiernan, & Hornyak, 2011). Local data in the Edmonton area confirms this, where we see large demand and waiting lists for learn-to-ride programs such as Free2BMe and You Can Ride 2 geared specifically to children over the age of eight with physical and intellectual disabilities (A. Ebert, personal communication, April 12, 2013). You Can Ride 2 is a free learn to ride program in Edmonton, Alberta that was conceptualized by the author of this thesis and has been serving children with disabilities since 2003 (Halayko, 2014). At the onset, You Can Ride 2 was geared towards children with developmental coordination disorder, and boasted a 100% success rate. Over the years, the program evolved to support the increasingly diverse learners that were enrolling. The goal was for all children to experience mastery in a variety of tasks before balance was challenged, and for the sessions to allow for significant variability and practice.

Most learn-to-ride courses use either direct skill training or adapted bicycles to teach; cycling skills are broken down and taught in a specific sequence or the bikes engineered to allow skills to progress. In You Can Ride 2, a second grouping was added that aimed to accommodate the specific needs of individuals with intellectual disabilities with the goal of improving their success. This group progressed at a slower pace with decreased verbal instruction and increased physical support. This proved to be largely unsuccessful. While the children with developmental coordination disorder, autism spectrum disorder, cerebral palsy and brain injury continued to experience an 80-100% success rate, the children with intellectual disabilities experienced failure and frustration far more frequently; of the 11 children with Down syndrome that have participated in You Can Ride 2, none learned to ride. Free2BMe, a local program run out of the University of Alberta has fared better, though success rates are still under 50% and a number of children with Down syndrome that can balance do not learn starting or braking (A. Ebert, personal communication, August 11, 2014). The goal of this study was to see if a different approach, one based on guided discovery and strategy use, would be more effective with this population.

Purpose and Direction of Thesis

This is a paper-based thesis comprised of 4 chapters. Chapters 2 and 3 are papers in preparation for submission to *Physical Therapy* and *Physical and Occupational Therapy in Pediatrics* respectively; this will be done in joint authorship with the supervisory committee involved. This thesis reports on a single case study whose overall purpose was to explore the applicability of Cognitive Orientation to daily Occupational Performance (CO-OP) for teaching cycling with individuals with intellectual disabilities, and to obtain a better understanding of the trajectory of cycling skill acquisition. It is hoped that this research will contribute to the ultimate goal of better understanding how individuals with Down syndrome and other intellectual disabilities can acquire cycling skills, how best to instruct them in attaining mastery and how to decrease the barriers between activity and participation.

The thesis starts with a presentation of the background for this study (Chapter 1). The first paper (Chapter 2), "Look Ma, I Did It!": Enabling Two-wheeled Cycling for Youth with Intellectual Disabilities Through Strategy Use", presents the findings of a study evaluating the effectiveness of a modified CO-OP approach with individuals with a moderate intellectual disability. The second paper, "Guided Discovery and Two-wheeled Cycling by a Youth With Down Syndrome: An In-depth Case Study" (Chapter 3), is a closer look at the learning trajectory of a youth who was unsuccessful in a learn-to-ride program (You Can Ride 2) but who learned to ride using CO-OP. Chapter 3 also looks at the key features of CO-OP that were used in the intervention, with the purpose of determining which of these may have more utility for individuals with Down syndrome. The final chapter explores the limitations of current methods of instruction, as well as pulling together the findings, and laying out a plan for future research (Chapter 4).

Background

The International Classification of Functioning, Disability and Health (ICF) model of human function and disability (World Health Organization, 2001) provides a helpful framework when considering the impact of existing cycling programs. It is increasingly being used to augment physiotherapy practice (Jelsma & Scott, 2011). The ICF conceptualizes a bidirectional relationship between an individual's health condition, personal factors and environmental factors on three components: body functions and structures, activities, and participation (World Health Organization, 2001). Aside from considering multiple influences that enhance activity or participation, the ICF also provides a universal standard language to describe health (Skelton & Rosenbaum, 2010) and to measure disability (World Health Organization, 2001). It does not seek to define the nature of or the relationship between the various components (Francescutti, Gongolo, Simoncello, & Frattura, 2011; Holt, Wagenaar, & Saltzman, 2010).

A better understanding of the relationships between personal and environmental factors, and activity and participation can lead to more effective interventions for those with movement challenges (Holt et al., 2010), or those who struggle to participate in an activity such as cycling. Several theorists in the fields of ecological and developmental psychology have explored this person-task-environment relationship. In his theory of affordance, Gibson (1977) postulated that the qualities and conditions of objects in the environment (or the environment itself) encourage certain interactions, as illustrated by the affordance a ball has to being thrown or a den has to being used as a shelter. While Gibson's theory considered mainly how a person (or animal) perceives their environment, Newell's theory of movement constraints extended these ideas to also consider the influence of the task (Newell, 1991). For example, one's ability to throw a ball is influenced by the size of one's hand in comparison to the ball, just as the distance one wishes to throw it (or the task goal) will influence the type of throwing pattern one chooses to adopt. This also forms the basis of the ecological task analysis approach (Davis & Burton, 1991; Davis & Broadhead, 2007). Similarly, the dynamic systems theory suggests that patterns of movement emerge from the relationships between the person, the task and the environment (Thelen & Smith, 1994; Thelen, 1989). As such, it is necessary to consider performer variables relative to each task dimension and within the relevant environmental context. While factors within the person, the task, or the environment might impede learning, manipulation of these variables can also lead to mastery.

Cycling Programs

Learning to ride a bicycle may be influenced by factors within the individual, the task or the environment. Within the individual, failure may be due to poor endurance, lack of focus, or behavioral challenges (Klein et al., 2005), or as a result of decreased motivation or fear (Witter, 2013). Success may be impacted by task related factors including steering, balancing, pedaling and braking (Ducheyne, De Bourdeaudhuij, Lenoir, Spittaels, & Cardon, 2012), and considerations such as bicycle fit or the cycling environment (Witter, 2013).

The task and the environment are manipulated in a cycling program called iCan Bike (www.icanshine.org), formerly "Lose the Training Wheels" (Burt, Porretta, & Klein, 2007; Klein et al., 2005). This five-day camp begins teaching cycling indoors (typically in a school gym), using adapted bicycles that alter the tasks of balancing and pedaling (Klein et al., 2005; MacDonald et al., 2011; Ulrich et al., 2011). Of these modifications, Klein explains, "We don't teach anyone to ride per se; instead, the adapted bikes do the teaching" (Klein et al., 2005). In iCan Bike, children generally begin on one of the most stable bicycles where both wheels have been replaced by rollers that ensure stability but retain the dynamic properties of a two-wheeler (Klein et al., 2005). As stated by Roberts (1995), "it is the acquisition of the necessary habits of steering, with their delicate adjustment of timing, that constitutes the principal task involved in learning to ride a bicycle" (p. 223). The bicycle's design facilitates implicit learning of these appropriate balance reactions, which consequently shapes the skill by allowing the child to practice and encode effective motor responses on progressively less stable bicycles (Klein et al., 2005). This approach is thought to be effective since few people are aware of either the mechanisms of balance or their contributions to the steering process as they are riding (Davis & Broadhead, 2007). In the progression of bikes, the tire configuration changes from stable rollers to those with more contour and less stability, as well as from more to less friction which forces the child to participate more in balance reactions and allows for more speed (Burt, 2002). Additionally, explicit feedback is given to the child on factors such as maintaining a forward visual focus, pedaling continuously and initiating the handlebar steering actions, skills considered by Klein to be the foundational skills of cycling (Burt et al., 2007; Klein et al., 2005).

Most of the research on teaching two-wheeled cycling to children with disabilities is affiliated with the iCan Bike program, which reports a success rate of around 70-80% (Klein et al., 2005). A summary of the studies using the adapted bicycles of iCan Bike is presented in Table 1. Using the ICF framework, this success is likely measured at the activity level rather than at the participation level. ICan Bike studies show that between 56% and 100% of children are able to ride 9-30 meters on a two-wheeled bicycle after completing the program (Burt et al., 2007; MacDonald et al., 2012; Ulrich et al., 2011). However, as these numbers do not necessarily reflect the ability to ride in an outdoor setting, self-start, navigate, or brake, they overestimate the level of success. For example, while 22 (73.3%) children with Down syndrome were able to ride a distance of over 30 meters, only 5 (16.7%) were able to launch the bike on

their own (MacDonald et al., 2012). Similarly, Burt (2002; 2007) reported a 100% success rate in learning to ride; however, success was defined as being able to ride a straight distance of 12 meters after being helped with starting, and less than half of participants were able to navigate obstacles. In many of the iCan Bike studies, the equipment used and the environment ridden in are different from what would be encountered in the community. In the case of Burt's studies, children used iCan Bike's cruiser style of bicycle that allowed them to sit relatively upright in the saddle rather than on their own bike and rode in a gym. The transfer of cycling skills to an outdoor environment was not reported.

In itself, skill mastery in the outdoor setting is not a guarantee that cycling can be generalized or transferred. Outdoor riding with iCan Bike is typically assessed in a parking lot or a similar flat, controlled environment (MacDonald et al., 2012). As this is not necessarily representative of what one would face riding in the community it is not unexpected that success rates in the community would be less than those reported in the course. Witter (2012) surveyed 11 families who went through the iCan Bike camp and found that while 9 were riding independently or with a spotter at the end of the program, only 7 maintained the skill in the community. Similarly, Witter's 2013 survey found that though 9 of 10 children learned to ride in the camp, only 5 were riding at follow-up. The generalizability of cycling skills also appears to be dependent on disability. In these two studies, participants with autism were most likely to learn and retain the skill, while those with intellectual disabilities and developmental coordination disorder were least likely. MacDonald and her colleagues (2012) found a similar trend; the parents of 93% of children with autism spectrum disorder reported their children continued to ride at home after experiencing success in the class, compared with only 61% of children with Down syndrome.

Just as factors within the person, the task or the environment can influence cycling mastery, so too can these factors influence cycling participation. The child's fear of the unknown, their difficulty mastering certain skills, poor weather conditions, decreased accessibility of the child's riding environment and decreased parental knowledge or support may all contribute to limited participation in community cycling (Witter, 2013). Though iCan Bike offers several advantages, not least of which is the immediate success when riding an adapted "two-wheeled" bicycle and the resultant decrease in fear associated with the activity (V. Temple, personal communication, August 20, 2014), challenges remain with transferring the activity learned in the camp to participating in the home environment.

CO-OP: An Alternate Teaching Technique

Cognitive Orientation to daily Occupational Performance (CO-OP) is a teaching technique that may provide an alternative to programs that do not address skill transfer to everyday activities. The objective of CO-OP is to promote skill acquisition through strategy use, which in turn leads to generalization and transfer of the skills (Polatajko et al., 2001; Polatajko & Mandich, 2004). It is an individualized, task-oriented approach that uses cognitive strategies to integrate motivational, learning and movement science theories (Polatajko & Mandich, 2004). To date, CO-OP has not been tested with individuals with intellectual disabilities; however, it has been used successfully to teach a variety of skills (including cycling) to individuals with a diagnosis of developmental coordination disorder (Bernie & Rodger, 2004; Mandich, Polatajko, & Rodger, 2003; Miller, Polatajko, Missiuna, Mandich, & Macnab, 2001; Ward & Rodger, 2004), autism spectrum disorder (Phelan, Steinke, & Mandich, 2009), stroke (McEwen, Polatajko, Huijbregts, & Ryan, 2009; McEwen, 2009), and acquired brain injury (Dawson et al., 2009). There are seven key features to CO-OP: 1. client-chosen goals, 2. a dynamic performance analysis, 3. strategy use, 4. guided discovery, 5. enabling principles, 6. intervention format, and 7. parent/caregiver involvement (Polatajko & Mandich, 2004). Each of these features is described below.

CO-OP intervention begins with client goal selection (Key feature 1) using completion of the Canadian Occupational Performance Measure (Law et al., 1990) to identify 3 performance goals (Polatajko & Mandich, 2004). Several benefits exist from self-selecting goals. Deci, Vallerand, Pelletier and Ryan (1991) suggest that "when a behavior is self-determined, the regulatory process is choice, but when it is controlled, the regulatory process is compliance (or in some cases defiance)" (p 327). A self-selected motor goal is therefore more likely to result in engagement and ultimately, learning. It is also a positive predictor for motivation (Larin, 1998), which drives a person to attempt to master a challenging task in his or her environment (Thelen, 2005). This can have the added effect of increasing self-efficacy once the goal is achieved (Heller, Hsieh, & Rimmer, 2004). Bandura defines self-efficacy as the belief in one's capability to organize and execute the action required to achieve a specific goal (Haibach, Reid, & Collier, 2011). In a study by Witter (2013), self-efficacy relating to cycling improved following successful completion of a 5-day bike riding camp. Changes were seen in both those who learned to ride independently, and those who improved in their skills but did not achieve their goals, suggesting even small successes can have positive effects on self-efficacy.

Bandura's self-efficacy theory (1977) proposes that a motivated action satisfies the individual's need to be competent within a skill, and affects both initiation and persistence of behavior. Individuals with lower self-efficacy tend to approach difficult learning tasks as threats to be avoided rather than challenges that offer the reward of skill mastery (Bandura, 1986). This results in decreased motivation to practice motor skills, thus resulting in lower levels of physical

activity (Cairney et al., 2005), which becomes more pronounced with age (Cairney, Hay, Faught, Corna, & Flouris, 2006). Conversely, higher self-efficacy is one of the strongest predictors of physical activity in several populations, including individuals with intellectual disabilities (Hutzler & Korsensky, 2010; Peterson et al., 2008). This also seems true for cycling; Ulrich et al. (2011) found that individuals with Down syndrome who learned to ride decreased their sedentary behavior by 75 minutes per day.

Once goals have been set, performance problems and breakdown points are identified by a process called dynamic performance analysis (Key feature 2) (Hyland & Polatajko, 2012; Polatajko, Mandich, & Martini, 2000; Polatajko & Mandich, 2004). This client-centered, iterative analysis explores the impact of the person, the task and the environment on the child's performance (Hyland & Polatajko, 2012). Subsequently, the global problem solving strategy of *goal-plan-do-check* is taught (Key feature 3). This frames the CO-OP intervention sessions. With a goal in mind, the knowledge gained through the dynamic performance analysis, and a process called guided discovery (Key feature 4), children are assisted to set a plan and identify strategies that will assist them in achieving their goals (Mandich, Polatajko, Missiuna, & Miller, 2001; Polatajko & Mandich, 2004).

Having children participate in the cognitive process of learning a new skill fits with the first step in the three-stage model of the process of motor learning proposed by Fitts and Posner (1967) and the two-stage models proposed by Gentile (1972). The models include both the development of an understanding of the task dynamics (Key feature 2), and the acquisition of strategies (Key feature 3) that can be used to carry out the task either independently or with scaffolding (Shumway-Cook & Woollacott, 2007). Younger children have decreased awareness of the strategies that might be effective and therefore needed external guidance (Key feature 4) to

assist them to regulate their learning (Cooper & Corpus, 2008); the same holds true for individuals with intellectual disabilities.

Key feature 5 of CO-OP is enabling principles. Vygotsky (1978) suggests that cognitive functions are developed during interactions between the child and a more knowledgeable other as they engage in a goal-focused joint activity (Greenberg Lyons, 1984). This scaffolding "enables a child or novice to carry out a task or achieve a goal, which would be beyond his (or her) unassisted efforts" (Wood, Bruner, & Ross, 1976). The difference between what a child is able to do independently and what they can do under adult guidance, or in collaboration with more capable peers is defined as the "zone of proximal development," and was introduced in Vygotsky's social development theory (1978). Forms of scaffolding include simple instructions such as hints, directions, or reminders and facilitative scaffolding which encompasses prompting with questions, enhancing the proportion of the task the learner is able to do by breaking a task down into component parts, or providing help with part of the task so the child can do more (Page & Ross, 2004). The concepts of zone of proximal development and scaffolding can likewise be applied to the process of learning motor skills (Exner, 1990), and may be extremely helpful for children who cannot figure out how to learn to ride a bike on their own. Within CO-OP, the strategies used to help scaffold skills (e.g., modeling, prompting, shaping) are encompassed in the enabling principle of promoting learning. Three other enabling principles are also used: make it fun, work towards independence and promote generalization and transfer.

While the global strategy of CO-OP provides a general framework for the intervention session, domain specific strategies are specific to each task and are introduced only to solve specific performance issues (Polatajko & Mandich, 2004) (Key feature 3). These include discussion regarding the specifics of the task (task specification), cueing attention to body

position or feeling the movement, talking through the steps of a motor sequence (verbal rote script), prompting attention to doing the task, guiding motor performance by labeling task components (motor mnemonic), and supplementing task knowledge. These strategies are used to promote skill acquisition over the intervention period, which typically runs for 10-12 sessions (Key feature 6). Parents and caregivers are encouraged to be an active part of the entire CO-OP process (Key feature 7), particularly to facilitate the generalization and transfer of the skills to the home environment (Polatajko & Mandich, 2004).

Motor Learning Theory

The ultimate focus of most interventions, including CO-OP, is to promote motor learning; that is, to effect relatively permanent changes in skilled movement through practice or experience (Schmidt & Lee, 2011). Currently, there is no one prevailing theory that explains motor skill acquisition in its entirety; rather, cognitive, developmental, behavioral, and learning theories are used in combination to guide intervention (Zwicker & Harris, 2009). Motor learning strategies are the practical application of these theories, and can be used to structure the presentation of the task and the learning environment during intervention (Levac, Wishart, Missiuna, & Wright, 2009). Three main strategies are appropriate for physiotherapy interventions: giving verbal instructions; organizing the amount, structure and schedule of practice; and providing feedback about task performance or outcome (Levac et al., 2009). In CO-OP, the key features of guided discovery and domain specific strategies fit under the first motor learning strategy which encompasses providing information about relevant task information or directing the learners' attention towards different aspects of the task. The other strategies can easily be incorporated into intervention sessions.

Motor learning in children with intellectual disabilities. Ample practice time is required to promote mastery of motor skills for children with disabilities (Bouffard, 1990). In addition to the number of practice sessions, the structure of practice sessions also needs to be considered (Schmidt & Lee, 2011). Studies show that most individuals are better able to retain and transfer motor skills when the following conditions are met: a) several different but related conditions are practiced (Magill & Hall, 1990; Shea & Morgan, 1979), b) one skill is learned in variable conditions (Shea & Morgan, 1979; Shumway-Cook & Woollacott, 2007), and c) skills are practiced in a random versus a blocked schedule (Edwards, Elliot, & Lee, 1986; Lee & Magill, 1983). With respect to cycling, the following examples might apply: a) practicing on a conventional bike, scooter and spin bike, b) practicing cycling on a hill, in a parking lot, and on a trail, and c) if different components of cycling (e.g., pedaling and starting) were practiced in a random order. Battig (1979) termed these phenomena "contextual interference," and postulated that to practice in high contextual interference the learner must process the information with greater depth, thus creating a more resilient memory of the motor skills (Lee & Swinnen, 1994; Nearingburg, 1989). Though the initial acquisition rates with high contextual interference are slower, both learning retention and transfer is enhanced (Shea & Morgan, 1979). This paradox also appears to hold true for individuals with Down syndrome (Edwards et al., 1986) and other individuals with intellectual disability (Porretta & O'Brien, 1991).

A similar pattern of slow acquisition but improved retention has been found with the third motor learning strategy, when knowledge of results or extrinsic feedback is provided relating to success in achieving a motor goal (Wu et al., 2011). However, in a situation where the learner is acquiring whole body actions—such as is the case with learning to ride a bike—the learner also requires knowledge of performance, or information about the dynamics of the movement in

addition to knowledge of the outcome (Newell, 1991). Both knowledge of performance and knowledge of results are frequently incorporated into CO-OP interventions as children are supported in checking to see if their plans were effective.

The Impact of Cognition on Learning and Motor Performance

Cognitive functioning can have a significant influence on motor learning (Danielsson, Henry, Messer, & Ronnberg, 2012)because of the problem solving and decision-making involved (Shuell, 1986). Intellectual disability is defined as "a group of developmental conditions, characterized by significant impairment in cognitive functions, which are associated with limitations of learning, adaptive behavior and skills" (Salvador-Carulla et al., 2011). Psycho-educational testing is widely used to measure cognition, though it only determines the unaided ability to answer specific questions or solve certain types of problems, which are largely used to determine school programming (Campione & Brown, 1985; Elliott, 2003). While cognition is generally reported to be in the domain of body functions, a higher intelligence quotient (IQ) is also associated with higher performance in the activity and participation categories of the ICF (Carr, 1994; Rihtman et al., 2010).

In 25-50% of cases of intellectual disability there is a genetic cause (McLaren & Bryson, 1987), the most common being Down syndrome, which impacts 1 in 800 Canadians (Canadian Down Syndrome Society, 2014). Down syndrome is a multisystem genetic condition that affects growth and development as well as participation in daily activities (Rihtman et al., 2010). Because of the prevalence of Down syndrome and the fact that there are multiple other challenges faced by this population as compared with others with intellectual disabilities, this group is discussed in greater detail. Considered from the perspective of the ICF framework, in the areas of body functions and structures, children with Down syndrome tend to present with a specific behavioral phenotype compared with other children with intellectual disabilities (Chen, Ringenbach, Albert, & Semken, 2014; Daunhauer & Fidler, 2011; Patterson, Rapsey, & Glue, 2013). In the areas of behavior, cognition, and language, relative strengths are seen with non-verbal memory, visualspatial and visual perceptual abilities, and social behavior (Baddeley & Jarrold, 2007; Daunhauer & Fidler, 2011), whereas relative challenges are evident in the areas of verbal memory, expressive language and goal directed behavior (Baddeley & Jarrold, 2007). In the physical and motor domain, individuals with Down syndrome also have more challenges with balance and hand function (Chen et al., 2014; Jobling, 1999; Lahtinen, Rintala, & Malin, 2007).

While memory abilities generally develop with cognitive age, individuals with Down syndrome have a much easier time recalling information presented visually (e.g., modeling and printed text) than they do remembering information presented verbally (Jarrold & Baddeley, 2001; Laws, MacDonald, Buckley, & Broadley, 1995). Compared with others with intellectual disabilities, individuals with Down syndrome are often more effective imitators of visual models (Edwards et al., 1986; Maraj, Li, Hillman, Jeansonne, & Ringenbach, 2003). The use of hand gestures or other forms of visual support is also beneficial (Wang, Bernas, & Eberhard, 2001).

If scaffolding involves verbal instruction, one must keep in mind that the language challenges experienced by individuals with Down syndrome are broad, encompassing all aspects of speech and language development, and go beyond the intellectual disability associated with the syndrome (Næss, Lyster, Hulme, & Melby-Lervåg, 2011). The one exception is receptive vocabulary, which is largely in line with cognitive age (Næss et al., 2011). While this may be a relative strength—at least related to other language domains—Morgan, Moni and Jobling (2009)

suggest it may still be necessary to pre-teach the meaning of question words to facilitate learning. Wishart (2001) also describes a profile of avoidance and reluctance to take initiative when individuals with Down syndrome are faced with learning new skills. While this may, as the author describes, be a result of individuals with Down syndrome generally having challenges with motivation, it may also be that they do not have the self-efficacy to attempt the task (Clarke & Faragher, 2014).

In addition to memory and language challenges, individuals with Down syndrome exhibit lower tone, increased flexibility and differences in motor reaction and performance as compared with those without the diagnosis (Daunhauer & Fidler, 2011). These differences, as reviewed by Virji-Babul, Lloyd and Van Gyn (2003), include longer reaction times, longer overall movement times and increased variability of performance, particularly when it relates to a complex movement sequence, such as riding a bicycle. Differences in motor timing (as well as balance and coordination) in individuals with Down syndrome may be related to changes in the cerebellum (M. F. Morgan et al., 2009; Pinter, Eliez, Schmitt, Capone, & Reiss, 2001); however, other factors such as attention and concentration may also have an impact (Almeida et al., 2000).

Bouffard (1990) concludes that with individuals with intellectual disabilities, cognitive difficulties with movement may be due to several factors including inadequate motivation, lack of executive control or self-regulation, deficiencies in the knowledge base, inadequate metacognitive knowledge and understanding, inability to use strategies spontaneously, and decreased practice. Each of these factors as well as language, verbal memory and motor skills must be considered when using a CO-OP intervention.

The Potential Use of CO-OP for Individuals with Intellectual Disabilities

In CO-OP, a certain level of cognitive skills, behavioral responsiveness and language fluency is considered to be essential to benefit from the verbally based approach (Polatajko & Mandich, 2004). Consequently, the cognitive and language challenges of individuals with intellectual disabilities would suggest that CO-OP may not be an effective intervention. This assumption is supported by other studies, at least with respect to the applicability of a metacognitive strategy. Yang and Porretta (1999) explored the effectiveness of a four-step strategy for teaching throwing skills with individuals with mild intellectual disabilities. While the steps *ready*, *look*, *do* and *score* effectively cued participants to focus on certain aspects of the task, their ability to retain and generalize these steps was questionable. Similar concerns exist with the global "problem solving process in movement situations" strategy described by Bouffard (1990). This process involved five steps: 1. identifying that a problem exists; 2. defining or representing the problem; 3. retrieving or constructing a plan; 4. executing the plan; and 5. evaluating progress. As described in Bouffard, individuals with intellectual disabilities have difficulty in each of the above steps: they are less likely to notice that there is a problem and are often unable to represent it, and they cannot independently generate, execute or evaluate appropriate plans.

There has been at least one study reporting an individual being excluded for not having enough cognitive ability to be able to work with the therapist using CO-OP, though in this case the goals chosen (i.e., improving spelling and reading) were academic in nature (Missiuna et al., 2010). Polatajko and Mandich (2004) clarify that "sufficient cognitive ability does not translate into a specific level of ability on any specific measure; rather, it means that the child can attend and understand sufficiently to interact with the therapist around strategies" (p. 50). Likewise, the

authors do not specify a set level of behavioral responsiveness, or receptive or expressive language ability, though they suggest the child should want to do the activity (generally assumed with self-selected goals) and should be able to engage and communicate with the therapist. While no studies have yet looked at the effectiveness of CO-OP with individuals with intellectual disabilities, studies have looked at the applicability of the technique with other populations with cognitive deficits. These include adults with a stroke or brain injury that had no premorbid cognitive challenges (Dawson et al., 2009; McEwen, Polatajko, Davis, Huijbregts, & Ryan, 2010; Skidmore et al., 2011), and children with a traumatic brain injury for whom pre-injury function was not reported (Missiuna et al., 2010). Since therapists have the ability to adjust their language level and intervention approach to meet the child's abilities, and can use several enabling principles to promote learning, it is likely that at least certain aspects of CO-OP will be effective in teaching children with intellectual disabilities to ride two-wheeled bicycles.

The following chapters describing the program of research are designed:

- To explore the applicability of Cognitive Orientation to daily Occupational Performance (CO-OP) for teaching cycling to individuals with intellectual disabilities
- To evaluate the use and utility of the components of CO-OP as they apply to individuals with intellectual disabilities and cycling
- 3. To describe the learning trajectory and cycling skill acquisition of a youth with Down syndrome participating in a CO-OP intervention
- 4. To pull together the findings and to lay out future directions

Table 1

Study	Age	N	Bike skill achieved				
	Environment ¹	Diagnosis ²	% Riding	% stopping	% Self-start	Navigation	Maintenance/
	Bike		(meters)		(launch)		generalization
Burt (2002, 2007) ³	7-11 years	Total (10)	100% (12 m)	NR^4	NR	3/10 able to	6/10 maintained skill 2
	indoors					navigate cones	days post camp (indoors)
	camp bikes used	ASD (4)	100% (12 m)	NR	NR	3/4 navigating	3/4 maintained
		DS (3)	100% (12 m)	NR	NR	0/3 navigating	1/3 maintained
		Other (3) (DD, CP, dyspraxia)	100% (12 m)	NR	NR	0/3 navigating	2/3 maintained
Macdonald et al. (2012)	9-18 years	Total (71)	80.3% (30 m)	74.6%	47.9%	NR	38/47 riding at home
	outdoors		0.5 40/	02.00/	70.70/	ND	11 did not respond
	camp bikes used	ASD (41)	83.4%	82.9%	/0./%	NK	7 unknown
		DS (30)	73.3%	63.3%	16.7%	NR	11/18 riding at home; 4 unknown
Ulrich et al. (2011)	8-15 years	DS (34)	56% (9 m)	NR	NR	NR	NR- sedentary
	environment& bike used unclear	(control group 27)	(0%-control)				behavior decreased by 75 min/ day vs control

¹ environment refers to location (indoors or outdoors) in which skills were assessed ² *Note.* ADHD, attention deficit hyperactivity disorder; ASD, autism spectrum disorder; CP, cerebral palsy; DCD, developmental coordination disorder;

DD, developmental delay; DS, Down syndrome; GDD, global developmental delay; GM delay, gross motor delay; ID, intellectual disability;

OCD, obsessive compulsive disorder; TS, Tourette's syndrome

³ Burt (2007) is comprised of same population as Burt (2002) minus the three participants with Down syndrome ⁴ NR, not reported

Table 1 (continued)

Summary of Studies Using iCan Bike Equipment

Study	Age	Ν	% Riding	%	% Self- start	Navigation	Maintenance/
	Environment	Diagnosis	(meters)	stopping	(launch)		generalization
	Bike						
Witter et al.	7-11 years	Total (11)	45% independent,	45%	NR	NR	64% riding at home
$(2012)^3$	outdoorg		36% with spotter	36%			3-4 months post
	outdoors		(distance NR)				camp
	bike used unclear	ADHD (1)	100%	100%	NR	NR	1/1 riding at home
		ASD (4)	50% (50% with spotter)	50%	NR	NR	4/4 riding at home
		DCD (4)	25% (25% with spotter)	25%	NR	NR	1/4 riding at home
		DS (1)	(100% with spotter)	0%	NR	NR	0/1 riding at home
		ID (1)	100%	100%	NR	NR	1/1 riding at home
Witter (2013)	7-18 years	Total $(25)^6$	76% (30m)	76%	76%	NR	5/10 riding at home
			(20% with spotter)				15 unknown
	outdoors	DS (2)	50%	50%	50%	NR	0/2 riding at home
		ASD (3)	100%	100%	100%	NR	2/3 riding at home
	various bikes	ID (1)	100%	100%	100%	NR	0/1 riding at home,
	(4/10 used						1 had setback ⁷
	own bicycle	CP, GDD,	100%	100%	100%	NR	3/4 riding at home,
	with handle attached)	GM delay & TS/OCD/DCD					1 had setback

⁵ based on information and table shared by V. Temple on August 20, 2014
 ⁶ only 10 of 25 participants who agreed to participate completed follow-up. While riding success is known, follow-up data is based on n=10
 ⁷ setback refers to something external that negatively influenced cycling (e.g., a fall, surgery)

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Chapter 2: "Look Ma, I did it!": Enabling Two-wheeled Cycling for Youth with Intellectual Disabilities Through Strategy Use

[in preparation for submission to *Physical Therapy*]

Abstract

Learning to ride a bicycle can be a significant milestone in the life of a child. Children with disabilities frequently struggle with mastering this skill, limiting their participation in this important childhood leisure activity. Cognitive Orientation to Occupational Performance (CO-OP) is an approach that has been used to teach motor skills to children with average cognition, but has not vet been studied with children with intellectual disabilities. A multiple baseline single case design with pre-post measures was implemented in a community setting to test the applicability of the CO-OP approach for enabling children with intellectual abilities to ride a bike independently. Primary outcome measures were distance and time cycled, and secondary measures were cycling skills mastered and performance and satisfaction measures (by parent report) using the Canadian Occupational Performance Measure. Seven youth with a moderate intellectual disability (IQ 40-53) aged 10-19 and unable to ride a two-wheeler were included. Intervention sessions consisted of up to eight (M=7.4) sessions provided over a range of 27 to 44 days (M= 35). Following intervention, six of the participants rode more than 100 meters (range 103m-1756m) and demonstrated improvements in all biking skills and measures of parent satisfaction. The seventh child did not learn to ride. The positive results across measures and participants are higher than is typically reported for the amount of intervention given. Results suggest CO-OP is effective for teaching two-wheeled cycling to children with intellectual disabilities. This is helpful as we move towards determining best practice for teaching cycling to children with disabilities and expand the populations for whom a CO-OP approach may be beneficial.

Introduction

Learning to ride a two-wheeled bicycle can be a significant milestone in the life of a child. On average, children learn to ride a bike at the age of six (Blondis, 1999; Klein et al., 2005), though this can range from age three to eight (Hansen et al., 2005). For children with intellectual disabilities, specifically those with concomitant motor challenges such as Down syndrome, learning to ride a bike can be extremely challenging, if not elusive. Only between 9 and 36% of children with Down syndrome ever master the skill, and many families report having given up after years of failed attempts (Buckley, Bird, Sacks, & Archer, 2002; Ulrich et al., 2011). Those that do learn report several positive outcomes including decreased time spent in sedentary activities, increased motivation to try other physical and sports activities, increased self-esteem and positive peer relationships (Klein et al., 2005; MacDonald et al., 2011; Ulrich et al., 2011). As many children with intellectual disabilities struggle in one or more of these areas (Duvdevany & Arar, 2004; Esposito, MacDonald, Hornyak, & Ulrich, 2012; Hutzler & Korsensky, 2010), the value of cycling as both a social and a fitness opportunity cannot be understated.

The World Health Organization's International Classification of Functioning, Disability and Health (ICF) is a recognized framework that can be used to describe the impact of health conditions on functioning (World Health Organization, 2001). The components of the ICF (body functions and structures, activities, participation and contextual factors) are also important to consider when developing physiotherapy interventions and evaluating the effectiveness of a particular intervention (Darrah, 2008; Goldstein, Cohn, & Coster, 2004). When looking at setting a child up for success on a two-wheeled bicycle, research suggests that increased age and strength (Ulrich et al., 2011), social motivation to ride with peers (MacDonald et al., 2012), and adapted bicycles (Klein et al., 2005), may each play a role; however, which factors are most

important and how best to teach the skill remains unknown. Existing studies on teaching cycling to children with disabilities look at modifying or gradually increasing the task demands, adapting the equipment and environment or using some combination of these.

The iCan Bike program, formerly "Lose the Training Wheels", is arguably the most widely researched cycling instruction method used for children with disabilities. This technique is based on dynamic systems theory and focuses mainly on adapting the equipment and environment to promote success (Klein et al., 2005). The course is organized in a camp format with cycling instruction provided over 75 minutes for five consecutive days (MacDonald et al., 2012; Ulrich et al., 2011). As a child becomes more confident and capable of maintaining appropriate speed, body position, and balance, they progress through a series of increasingly tapered roller wheels allowing for the balance demands of cycling to be gradually increased (Burt et al., 2007; Klein et al., 2005). According to Klein et al. (2005, p. 53), the primary goals of iCan Bike are to "maintain a forward visual focus, pedal continuously, initiate handlebar steering actions, and consequently remain upright." Other skills such as self-starting, braking and navigation are considered secondary. Still other skills such as riding in the community are never attempted. Using the teaching methods and equipment of iCan Bike, the majority of children with intellectual and physical disabilities (56-100%) learn to ride over straight distances of up to 30 meters (Burt et al., 2007; MacDonald et al., 2012; Ulrich et al., 2011). However, these numbers do not reflect independence in all of the skills required to be successful with cycling in the community; if self-starting is considered, less than 17% of children with Down syndrome master the skill (MacDonald et al., 2012).

Cognitive Orientation to daily Occupational Performance (CO-OP), described in detail in Polatajko and Mandich (2004), combines motor learning principles with behavioral and learning

theories, and emphasizes the role of goal setting, problem solving and other cognitive processes on the development of movement skills. CO-OP is delivered in a specific format and includes six additional key features: client chosen goals, dynamic performance analysis, cognitive strategy use, guided discovery, enabling principles, and parent involvement. Studies have demonstrated its success in teaching a variety of skills including cycling to individuals with developmental coordination disorder (Bernie & Rodger, 2004; Miller et al., 2001; Ward & Rodger, 2004), autism spectrum disorder (Phelan et al., 2009), brain injury (Dawson et al., 2009), and stroke (McEwen et al., 2009), but no studies have yet been reported using it with individuals with intellectual disabilities.

Once a client has identified a goal (e.g., riding a bike), the therapist leading the CO-OP intervention begins the iterative process of determining where in their client's performance the breakdown is occurring (Polatajko & Mandich, 2004). This process of "dynamic performance analysis" takes into consideration a client's understanding of the activity, their desire and ability to carry out each aspect of the task, and the occupational and environmental demands of the task, and allows for additional teaching, motivational strategies or environmental modifications as required (Polatajko et al., 2000). Knowledge gained during the dynamic performance analysis also helps the therapist focus their "guided discovery" or the teaching process used to help clients self-identify solutions for success.

Intervention sessions are framed by the four step global cognitive strategy of *goal-plando-check*. Clients are guided to discover the plan of how they are going to reach their goal (e.g., pointing the wheel at a target to ride in a straight line) and to apply other more domain specific strategies aimed at facilitating success (e.g., learning the verbal motor mnemonic of "strong arms" to help remind them to keep their wheel pointing straight ahead). Finally, the client

performs the activity (do) using their discovered strategies and checks if the plan was implemented and if so if it was effective. If it was not, the client is guided to amend the plan and try again. Throughout all sessions, enabling principles are used to ensure the client remains engaged, is having fun, and continues to work towards independence. Parent involvement is a vital part of the process and ensures skills are generalized and transferred to their natural environment (Polatajko & Mandich, 2004).

Many consider learning to ride a bike to be a rite of passage of childhood. For children with intellectual disabilities, using modified bicycles is the predominant strategy for learning in the literature (Burt et al., 2007; Klein et al., 2005; MacDonald et al., 2012; MacDonald et al., 2011; Ulrich et al., 2011), but little if any research has focused on other methods of instruction such as CO-OP. This study aimed to determine the applicability of the CO-OP approach in teaching children with intellectual disabilities to ride a conventional two-wheeled bicycle.

Methods

Participants and Settings

Participants between the ages of 8 and 19 were recruited through the You Can Ride Two¹ website, word of mouth, and through an information bulletin put out by a local Down syndrome society. Inclusion criteria were a) the inability to ride a two-wheeled bicycle; b) mild to moderate intellectual disability (IQ of 40-70), c) the ability to verbalize choices, d) access to a suitable bicycle and e) willingness to learn to ride. Exclusion criteria included neuromusculoskeletal conditions, health concerns or behaviors that might impact participation

¹ You Can Ride Two is a volunteer-run program offered through the Edmonton Bicycle Commuters Society that aims to improve access to cycling to individuals with special needs.

(by parent report). The University of Alberta Research Ethics Board approved all procedures. Written informed consent from parents and verbal assent from each youth was obtained.

Participant characteristics. Thirteen families expressed interest in taking part in the study. Potential participants (8 male, 5 female) ranged in age from 8-19. Five youth did not meet inclusion criteria (two had IQ scores over the cutoff of 70, one was non-verbal, one refused to participate, and one had musculoskeletal surgery scheduled). One female, age 9, was accepted into the study but learned to ride during baseline, so was not included. Ultimately, seven youth (3 males, 4 females, M_{age}= 13.7, age range: 10-19 years) participated; six had a diagnosis of Down syndrome, one a diagnosis of Smith-Lemli-Opitz syndrome². Intelligence quotient (IQ) scores were obtained from the most recent psycho-educational assessment provided by parents and indicated that six of the children had a moderate cognitive delay (IQ= 40-53, M= 46). One participant had never received psycho-educational testing, though his parents felt his IQ fell within this range. Participants were also asked whether or not they wanted to learn to ride a two-wheeler; three youth said no despite assenting to participate in the study. Characteristics of the participants, and whether or not they wanted to learn to ride are presented in Table 2.

Setting. All baseline and intervention sessions as well as at least two follow-up sessions occurred in the parking lot and adjacent shallow hill and public bicycle trail system of a city park. Follow-up sessions were held at a location parents chose, typically in their community or on a trail system they were interested in exploring as a family.

Experimental Design

A single case multiple baseline design across participants was used. As required in this design, the start date of the intervention was staggered so that participants could serve as both

² Smith-Lemli-Opitz syndrome is a genetic condition frequently associated with intellectual disability, behavior challenges and multiple congenital malformations (Koenig, Scahill, & Teague, 2002).

their own controls and as controls for others (Kratochwill et al., 2010). However, as none of the children could cycle nor perform many of the sub skills associated with cycling, repeated assessment during the intervention would be meaningless and add unnecessary stress and frustration. Rather, a multiple probe technique was employed (D. Morgan & Morgan, 2009); probes in the intervention phase were done immediately prior to the intervention sessions. Pre and post measures were also used to better determine the clinical validity of the intervention. Inter-rater reliability analyses using the weighted kappa statistic and the interclass correlation statistic were performed to determine consistency among raters (Advanced Analytics, 2013).

Materials

Each participant brought his or her own bicycle to the sessions, which was inspected and tuned up by a certified mechanic. All participants were required to wear a helmet when on their bicycle. Optional safety equipment included kneepads, elbow pads, cycling gloves and a wingman support harness. In one case (participant 4), a support handle was used instead of the harness; this was designed and installed by his father.

Dependent Measures and Interobserver Agreement

Time and distance cycled. The primary dependent variables were distance and time cycled. Both were measured from where the participant put both feet on their pedals and/or external support was removed (whichever came last) to the point when one foot contacted the ground and/ or external support was provided (whichever came first). Two 7-meter lines taped to the parking lot were marked at 1-meter intervals to facilitate measurement; all cycling distances less than 7 meters and the corresponding riding times were measured by video analysis. Distances over 7 meters were measured using a 4-inch metric measuring wheel (SKU: 8351488, Princess Auto). Within each baseline and follow-up session, the longest distance was measured,

and it was noted whether riding was achieved by stationary launch, dynamic launch or without assistance. The longest distances were timed by observation of the video.

Cycling skills. A cycling skill checklist developed by the primary investigator (Appendix A) was used to give more specific information relating to the secondary dependent variable of skills mastered. The skill checklist consisted of twenty cycling tasks divided into four sections: bike manipulation and stationary skills, pre-pedaling dynamic skills, and beginning and advanced riding skills. Each task was given a score of 0 to 5, with 0 representing a refusal or inability to complete the skill, and 5 representing independence with the skill. An observational score out of 100 was obtained. The skill checklist was created in consultation with two experienced cycling instructors with adapted physical education backgrounds, employed by an unrelated agency offering cycling classes for children with disabilities. Its validity has not been formally assessed.

Parent ratings. Prior to beginning the study, using the format of the Canadian Occupational Performance Measure (Law et al., 1990), all parents were asked how important it was to them that their child learns to ride, how they viewed their child's performance and how satisfied they were with their child's cycling ability. Importance placed on cycling is presented in Table 2 and the pre and post intervention parent ratings of performance and satisfaction are presented in Figure 2.

Interobserver agreement. Skill checklist ratings, time measurements and distance measurements less than seven meters were determined by video observation. Forty percent of baseline and follow-up sessions for each participant were randomly selected and rated by a second rater blinded to the phase of the study. Inter-rater reliability for the skill checklist was

very good (κ_w = 0.835 (p<0.001), 95% CI [0.797, 0.873]), and the reliability for both distance and time measurements was excellent (ICC (3,1)= 1.0 (p<0.001), 95% CI [0.999,1]).

Intervention Description and Treatment Fidelity

Baseline. The primary dependent variables of distance and time cycled were measured on five occasions as per the What Works Clearinghouse quality standards for single subject designs (Kratochwill et al., 2010). Each participant completed the five baseline probes over 3 to 5 separate days. Where two sessions occurred on the same day (for 5 of the participants), an effort was made to separate the sessions by a period of at least 10 minutes. This occurred for all but participant 7, who waited an average of 5 minutes between rides. Because of the multiple baseline design, participants remained in the baseline phase for between 6 and 42 days (Mdn= 14, M= 17.7). All but one family (participant 4) reported not practicing during this phase.

Both a stationary launch and a dynamic launch were attempted at baseline for all participants, as none were able to start riding independently. For a stationary launch, the rider was stationary with one or both feet on the pedals. The person supporting the rider took up to three steps before letting go. Stationary launch was attempted three times and the longest distance ridden after letting go was measured. For a dynamic launch, external support was given for more than three steps and was only removed if the child was relying very little on the instructor to remain balanced; the longest distance ridden without support within the session was measured. For safety reasons, if too much external support was required to help with balance while riding, the dynamic launch was not attempted and this was noted in each case. The secondary dependent variable of skills mastered was also measured at baseline. This included the following skills: getting on and off of the bike, balance on the bike, moving while on the bike (feet on ground propelling bike forward), pedaling in a straight line with support for balance, steering and braking.

Intervention. Each participant attended between 5 and 8 separate intervention sessions (Mdn= 8, M= 7) over a span of 21 to 47 days (Mdn= 31, M= 34.6). This variability was due to parent scheduling preferences over the summer months. The length of intervention sessions was also variable, though typically averaged 40-50 minutes. While 60 minutes was allotted for each session, late arrivals, unsuitable weather conditions, and lack of participant engagement sometimes caused the sessions to start late or to end early. Video recording errors also occurred, resulting in portions of the intervention not being captured. The shortest intervention session (session 7, participant 4) lasted 9 minutes; following a successful ride the youth declared he was "all done." Following this session, he moved on to the follow-up phase as he was able to start and stop independently, could turn and maneuver the bicycle and could ride for at least 1 minute; participant 2 also moved on early (after session 5). These skills corresponded with a score of 4 or 5 out of 5 on at least 17 of the tasks of the skill checklist, for a total score of 85 or higher. All other youths moved on to the follow-up phase when they had completed 8 sessions.

The global CO-OP strategy of goal-plan-do-check was taught in the first intervention session and reinforced or re-introduced as necessary each subsequent intervention session. At the beginning of each session, participants were prompted to choose a sub-goal or focus (e.g., participant 1 consistently asked to work on "starting" and participant 6 "balancing") and were asked where they wanted to practice (e.g., parking lot or hill). When a youth could not independently come up with a sub-goal, they were given choices of skills based on the dynamic performance analysis of where their cycling performance was breaking down. Participants were then guided to discover plans to overcome their performance problems (e.g., for riding in a straight line, they were asked if they would like to try the plan of pedaling *fast* or *slow*), and their choice was practiced before checking if the plan chosen was effective. Visual cues and reinforcements (e.g., use of pictures depicting different activities, getting the tires wet to show the trajectory of the bike at different speeds, review of videos, drawing pictures) were used as necessary to help participants decide on a goal or plan, or to check their plan's effectiveness. In this way, the youth were involved in all aspects of the decision making process.

Follow-up. Participants attended 5 follow-up sessions over 11 to 28 days (Mdn= 14, M= 16.7). Distance and time measurement procedures were the same as in baseline, though when independent launch had been achieved, only this method was measured. As most participants had progressed to independent cycling by follow-up, more advanced cycling skills were also assessed including managing different terrains (including hills), riding on narrower paths, self-starting, and navigation skills.

One month or more after the last follow-up session, a social validity questionnaire was completed to obtain feedback from parents about cycling in the community, barriers to cycling (if any), and the form of cycling (e.g., conventional or adapted bicycle) seen as most functional for the participant. Parent ratings of satisfaction with their child's cycling abilities and ratings of their child's performance were also obtained and parents were asked if they had used the strategies of CO-OP in other situations.

CO-OP modifications. The intervention was modified a priori with respect to clientchosen goal, and involvement of significant others. Regarding the former, only one goal cycling—was addressed and that goal was given to the youth rather than being chosen. Instead, the youth were given the opportunity to choose sub-goals. Regarding the latter, though parent involvement was encouraged throughout the intervention sessions and most parents stayed to

watch and/ or participate, this was not a requirement of the study, nor was there any plan that the parents would be asked to practice with the child outside the sessions. In addition, it was anticipated that additional modifications may have to be made to the approach, thus a log was kept of all modifications made and analyzed at the completion of the study. With these specific exceptions, all of the other principles of CO-OP were followed.

Treatment training and fidelity. Sessions were led by a pediatric physiotherapist with 12 years of experience teaching cycling and with training in the use of CO-OP from one of the developers of the approach. One intervention session for each child was selected and fidelity determined by video analysis by a second rater blinded to the intervention session. Intervention sessions were randomly stratified to allow rating of 7 different sessions (13% of sessions). Fidelity to the principles of CO-OP intervention was 90%; without considering discussion of homework, fidelity was 98.6%.

Results

The primary outcome measures of distance and time cycled were plotted on a graph and analyzed for trend, level and variability (Figure 2). The Nonoverlap of All Pairs (NAP) method (Parker & Vannest, 2009), was also used to compare the differences between baseline and follow-up measures and an online calculator used to confirm calculations (Vannest, Parker & Gonnen, 2011). The secondary outcome measures of skills mastered, and performance and satisfaction ratings were compared using the Wilcoxin matched pairs test. All secondary and pre-post measures were analyzed using SPSS (version 20).

Time and Distance Cycled

None of the participants were able to ride more than 1 meter entering the study. For all but one youth, the data demonstrated stability at baseline, with 6 participants cycling only

between 0-2 meters throughout the five baseline sessions. Participant 4 showed an upward trend, riding a distance of 30 meters on his fifth baseline session with a dynamic launch. On follow-up, six participants were able to ride a minimum distance of 31 meters (11 seconds), and rode over 100 meters (35 seconds) at least once. One youth (participant 7) remained unable to ride. The NAP indices were calculated at 1.0 for the 6 riders and 0.74 for the non-rider. Combined, this yielded an index of 0.96 (p<0.001), 90% CI [0.725, 1.201], corresponding to a large effect size in multiple baseline research (Petersen-Brown, Karich, & Symons, 2012). This significant effect is also reflected in the obvious positive changes in trend and levels for all but participant 7.

Cycling speed at follow-up for participants 1-6 ranged from 2.5 m/s to 4.5 m/s. On Figure 3 where distances and time correspond exactly (e.g., most of the data points for participant 3), the speed cycled is 3 m/s. Where the data points for the distance cycled are above the data points for the time cycled (e.g., all of data points for participant 4), the speed cycled is higher than 3 m/s.

Cycling Skills

Inevitably, with exposure to activities on the bicycle most participants improved in their skills over the five baseline measurements even though these improvements did not translate to increased distance cycled. The exception was participant 4 who by the end of the 5 sessions was able to cycle a distance of 30 meters; this was the participant who had been practicing daily with his father. Unlike the participant who withdrew from the study because she had mastered all 20 of the skills, participant 4 remained unable to turn, start or stop his bicycle. His final score on the skills checklist was 41/100 and the total number of skills he demonstrated independence in was 8/20. It was therefore decided that he would remain in the study.

Total scores (/100) for all participants on the skills checklist ranged from 24 to 47 (Mdn= 35, M= 36.5) for baseline sessions and 38 to 99 (Mdn= 84, M= 76.4) on follow-up. The trajectory of skills (/20) that participants were able to do without physical assistance (i.e., a score of 3-5 on the skill checklist) is presented in Figure 3 and ranged from 4 to 9 (Mdn= 7, M = 7) for baseline sessions and 7 to 20 (Mdn= 18, M= 16) on follow-up. Skills varied both within and between sessions, and were dependent on the environment (e.g., weather and length of trail) and the participant (e.g., feeling tired, in a good mood). A significant effect was evident between the average baseline and follow-up measures of cycling skills (Z= -2.37, p<0.05). The scores from the skills checklist at baseline and follow-up are presented in Figure 4.

Parent Ratings

Before beginning the study, five of the seven parents rated both their child's cycling performance on the COPM as 1/10 (M= 1.4) and their satisfaction with this performance as 1/10 (M= 2.6). None of the seven youth were able to ride without significant encouragement and physical support. Following intervention, the parental ratings ranged from 1-9 for performance (Mdn= 7, M= 6.4) and from 1-10 for satisfaction (Mdn= 10, M= 8.6). These changes were statistically significant (Z= -2.37, p<0.05; Z= -2.23, p<0.05). The parents of participant 4 indicated that they had practiced cycling regularly from the onset of the study and the parents of participants 2, 3, 5 and 6 reported that they began practicing once their children began demonstrating some independence with cycling skills.

At the one month follow-up, the parents of five of the seven youth (participants 2, 3, 4, 5 & 6) indicated that their child would continue to use a two-wheeled bicycle and that they had participated in one or more family bike rides since the conclusion of the study. The parents of participants 1 and 7 reported that they rarely if ever practiced (half hour or less) and that they

were uncertain whether two-wheeled cycling would be used in the future; future cycling would depend on if their children acquired further cycling skills. Participant 7 was unable to ride and while participant 1 was cycling, her mother reported that 2-wheeled cycling was not feasible for several reasons: the road was too busy to ride on safely, her daughter refused to ride on a sidewalk not bordered by grass on both sides (not available in her community), they had no easy trails around their house, and she could not transport her 6 children and all of their bicycles in their vehicle.

Discussion

This study demonstrated that a modified CO-OP approach is effective when teaching two-wheeled cycling to youth with a moderate intellectual disability. At the activity level of the ICF, results across participants indicated significant improvements in distance and time cycled as well as in all subjective measures (skills checklist, COPM parent ratings). At the participation level of the ICF, four of the seven youth were able to ride independently in the community (with supervision for safety) following intervention, with two other youth requiring closer supervision. Five parents reported that their children's cycling skills were maintained after the study concluded.

Parent support and the cycling environment appear to be key factors in the success within both the activity and participation levels of the ICF. Both positive and negative influences on success are demonstrated in four cases; 1. participant 4, whose father practiced with him on an almost daily basis, was able to ride for short distances even before intervention was implemented; 2. participant 1, whose mother rated cycling as minimally important (3/10) and had practiced a total of 30 minutes, was able to ride but did not continue cycling in the community; 3. participant 5, whose parents continued practicing with her after the follow-up and

provided videos of her continued skill improvement and, 4. participant 7, who had not practiced at all, did not learn how to ride.

Applicability of the approach

In addition to the a priori adaptations made to the approach, other aspects (specifically, use of the global strategy and the enabling principles) had to be modified or proved to work differently than generally reported in the literature. Most participants did not appear to remember or understand the meaning of the words "goal" "plan" "do" and "check" in the global strategy, and only one could recall them at the conclusion of the study. Regarding the latter, a number of enabling principles promoting learning and behavioral strategies not typically reported in CO-OP studies were implemented. These included direct teaching and prompting using visual supports (e.g., use of pictures and demonstrations), shaping the skill using environmental supports (e.g., use of knee pads and wingman postural support harness to inspire confidence) and positive behavioral reinforcement (e.g., happy faces earned, rewards following successful participation in the session).

For most participants, the guided discovery, domain specific strategies and enabling principles were particularly effective. Despite the cognitive delays of the participants, most were able to generate sub-goals and come up with appropriate plans given adequate guidance. Combining dynamic performance analysis and guided discovery resulted in the intervention progressing differently for each participant; the focus depended not only on where the performance was breaking down but also on what strategies the youth discovered. This also allowed participants to come up with their own ideas, though much of the time these were not verbalized. Participant 1 combined the idea of pre-positioning the pedals for starting with two foot coasting for generating speed and ended up launching the bike independently for the first time. This strategy for launching the bike was also the preferred method of starting for three other participants in the study. Conversely, participants 4 and 7, while being generally willing to try certain activities, were minimally engaged in the guided discovery process (e.g., often refused or were unable to make consistent choices). Because of the regular home practice of participant 4, it was difficult to determine how much impact the CO-OP approach had on his learning; at the very least, his ability to balance on his bicycle was learned through the practice he did with his father. As participant 7 had not had a psychological assessment, it was difficult to determine the impact of his cognition.

In all cases, the applicability of goal-plan-do-check as a global strategy was questionable with this population. Most of the youth did not seem to understand the meaning of the four words and could not remember them from week to week; similar findings were reported in a study using CO-OP with children with acquired brain injury (Missiuna et al., 2010), and in a study using the global strategy of ready, look, do and score with children with mild cognitive delays (Yang & Porretta, 1999).

Contributions

This study provides several contributions to the field of pediatric physiotherapy. First, it shows that individuals with intellectual disabilities including Down syndrome can learn to ride a two-wheeled bicycle with a specialized approach to teaching that does not require adapted bicycles. While the iCan Bike program is effective at targeting cycling at the activity level, its impact at the level of participation is less evident. Using CO-OP within a single case design, it was possible to assess how the interaction between each child's skills and the contextual factors impacted their participation in community cycling. The variability of skills seen within sessions, between sessions and between locations further reinforces the importance of considering the

child, the task and the environment when teaching cycling. In pediatric physiotherapy, CO-OP is not yet highly utilized; however, the positive results seen in terms of participant engagement in the intervention, in the outcomes at both the activity and participation levels and the ease of generalizing the strategies to the community make it an extremely powerful intervention method.

Second, the innovative plans generated by participants (e.g., how to launch the bike independently) highlight the feasibility and importance of soliciting the input of individuals with intellectual disabilities in the intervention process. This involvement was ensured by modifying certain aspects of CO-OP to enhance understanding; visual strategies were used to supplement guided discovery and behavioral reinforcements were used to increase engagement. These same strategies are helpful when using CO-OP with children with autism spectrum disorder (Phelan et al., 2009). In pediatric physiotherapy the results serve as a powerful reminder of the importance of involving the clients in the intervention process.

Finally, the fact that 86% of participants in this study were able to cycle a distance of over 30 meters (83% of participants with Down syndrome), and 71% of participants (67% of participants with Down syndrome) were able to generalize and transfer the skills to their communities highlight the efficacy of a cognitive approach compared to traditional approaches of teaching cycling to this population. As this is the first study investigating both the use of conventional bicycles and the use of CO-OP with this population, further research is required to confirm these results. However, since learning to ride can have a significant positive effect on decreasing sedentary behavior (Ulrich et al., 2011), these results hold promise for the pediatric physiotherapist.

Limitations and Future Directions

A limitation of the study is that parent involvement and practice was not considered closely; the parents of the child who did not learn to ride and the one who did not continue riding were the only ones who reported little if any practice at home. Although the results of this study indicate a strong effect of the intervention within a single-case research design, more complex study designs are necessary in order to continue building support for CO-OP when teaching two-wheeled cycling or other skills to children and youth with intellectual disabilities. A study that more closely adheres to the key features of CO-OP (e.g., child chosen goals, suggested intervention format) would be beneficial for determining its use with this population, as would a study examining the use of specific training in problem solving as a first step for using the CO-OP approach with this population. Future research should include group studies with larger numbers, a greater diversity of participants and a control group. Comparing the effectiveness of a CO-OP intervention against other cycling teaching techniques (e.g., iCan Bike) would also be beneficial.

Table 2

Participant Characteristics

ID	age	sex	Diagnosis *	IQ	Test/ year	Height (in cm)	Weight (in kg)	Desire to learn	Parent importance placed on cycling (/10)
1	12	F	DS	50	SB-5 (2011)	134	37.1	no	3
2	10	F	SLO	52	WISC-4 (2012)	136	31.5	yes	10
3	13	F	DS	40	WISC-4 (2013)	139	49.5	yes	10
4	13	М	DS	40	SB-5 (2013)	138	36.7	no	9
5	14	F	DS	41	WISC-4 (2013)	147	53.1	yes	8
6	19	М	DS	53	SB-5 (2013)	152	76.7	no	8
7	15	М	DS	NT	-	163	72.6	yes	8

* Note. DS, Down syndrome; SLO, Smith-Lemli-Opitz syndrome



Figure 1. COPM Parent Pre and Post Ratings of Performance and Satisfaction



Figure 2. Distance and Time Cycled



Figure 3. Independent Sub-skills at Baseline and Follow-up



Figure 4. Skill Checklist Scores at Baseline and Follow-up

Note: In Figures 3 and 4, B1 for participant 1 (P1) only tested 4 skills. As this differed from the skills presented in all of the other sessions and to all of the other participants, it was excluded from the analysis.

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| | r | C | yenng | 5 SKIII | Chee | KIISt | | |
|------------------------------|-----------------------|---------------|---------------|--------------------------|--------|----------|------------------|--|
| NAME: | Level Success/Support | | | | | | COMMENTS | |
| | | | | | | | | |
| | | | | | | | | |
| Skill | Succ | ess | Prom | npt | Phy | vsical | | |
| | | | | | Ass | istanc | e | |
| | t I | 2 - | ~ | 9 - | | | | |
| | pende | ls a fer
/ | aal/ Vi | o/ clos
Iby
rvisio | mal | imal | /
ole 0 | |
| | Inde
5 | Need | Vert
cue 4 | Dem
stane
supe | 2 Mini | Max
1 | DNA
unat
0 | |
| BIKE MANIPULATION A | ND ST | CATIO | ONAR | Y SKI | LLS | • | • | |
| 1.Picks up bike or | | | | | | | | Choose one of bike up OR kickstand up
5. able to do proficiently |
| kickstand up | | | | | | | | 4. verbal prompt on HOW to do it
3. demonstration |
| 2 Cata and ff hiles | | | | | | | | 2/1 physical cueing
5. gets on and off proficiently |
| 2. Gets on/off blke | | | | | | | | movement is not fluid but it is safe requires close standby or may fall |
| | | | | | | | | child tripped or fell or requires bike to be stabilized
on occasion |
| | | | | | | | | 1. requires full support to get on bike |
| 3. Puts pedals into position | | | | | | | | 5. consistently able to position pedals (or position of pedals unimportant for starting) |
| for starting riding | | | | | | | | 4. positions pedais occasionally or when both feet on pedals |
| | | | | | | | | can copy demonstration needs min physical prompt to move pedals pedals positioned for child |
| 4. Balances with one foot | | | | | | | | confident with balance can balance but requires more than one attempt; no |
| on pedal, one on ground | | | | | | | | risk of falling
3. child requires very close supervision due to |
| | | | | | | | | possibility of falling
2. child tripped or fell or needs bike to be stabilized |
| (5 S) | | | | | | | | 1. unable to balance without support |
| PRE-PEDALING DYNAM | IC SK | ILLS | <u> </u> | 11 | | 1 | <u> </u> | |
| 5. Moves bike with feet; | | | | | | | | skilled at coasting beginning to coast/glide with 2 feet; able to coast for |
| period of coasting | | | | | | | | fair distance but often falls to side/ is unstable
3. child walking bike; no period of double foot glide |
| | | | | | | | | child needs bike to be stabilized on occasion unable to walk bike without constant support |
| BEGINNING TO RIDE | 1 | 1 | 1 | 1 1 | | 1 | 1 | |
| 6. Stops | | | | | | | | uses brakes consistently can stop but often uses other method/ needs cue |
| | | | | | | | | requires demonstration as to how brakes work requires physical cueing to stop; cannot stop in safe |
| | | | | | | | | distance
1. requires significant physical cueing to stop |
| | | | | | | | | 0. does not put feet down when bike is stopped for
them. |
| 7. Pedals bike 5 | | | | | | | | can pedal 5 times but not fluidly able to pedal at least 3 times, may not be continuous
and may nood eminding |
| continuous revolutions | | | | | | | | 2. rarely pedals / pedaling is very intermittent
1 physical cueing is given to pedal
0. does not pedal |
| 8. Pedals bike 15 | | | | | | | | Note: this is looking strictly at pedaling not balance.
4. able to pedal consecutively but cannot quite do 15 |
| continuous revolutions | | | | | | | | (maybe due to space), may not be fluid but only needs intermittent cues |
| | | | | | | | | 3. able to pedal at least 10 times; may not be continuous
and may need reminding |
| | | | | | | | | 2. rarely pedals / pedaling is very intermittent
1. physical cueing is given to pedal |
| | | | | | | | | 0. does not pedal |

Appendix A Cycling Skill Checklist

NAME:	Level of Support					COMMENTS		
Skill	Success		Prompt Phy		nysical			
	Success		Trompt		Assistance		e	
		2						
	dent	few trie	Visual	lose ion 3		_	lable	
	ndepen	Veeds a inskiled	/erbal/` ue 4	Jemo/ cl tandby upervis	Minimal	Maxima	INA/ un	
BEGINNING TO RIDE (co	ntinue	ed)			E A	-		
9. Steers in generally								Note: this is strictly looking at trajectory (with/ without support)
straight path								5. rides straight 4. deviations in path over short distance
								S. significant deviations in path but going from A-B eds some physical cues to get to destination l. needs significant physical support to get to
								destination 0. unable to get to destination despite physical support
10. Can descend hills								 able to descend hills during regular rides able to descend but may go a bit fast, can take small
								grades but not larger ones3. requires standby supervision for safety; can ride but is only barely under control; feet not on pedals
								2. intermittent physical contact required (e.g., to slow down)
								0. does not attempt any hills
MASTERING RIDING								
11. sufficient speed/power								 Can ride independently and proficiently able to ride but looks down or has deviations in path able to ride but close standby supervision is required
to ride 10 meters								2. can power bike for 10 meters, person assisting can let go for short periods
								1. constant support for riding 0. cannot ride 10 meters
12. Can ride 30 meters in								 5. Can ride independently and proficiently 4. able to ride but looks down or has significant
parking lot								a solutions in path; consistent with balancea solution is required as balance is precarious
								2. can power bike for 10 meters, person assisting can let go for short periods
								0. Cannot ride 30 meters in wide space
13. Can ride 30 meters on								 independent on path veers off of path/ onto wrong side but self-corrects needs close standby assistance for safety
bike path/ road								 intermittent contact with bike or safety harness, falls can ride 30 m on path but only with physical support
14 Starts from stationary								5. able to start consistently and independently
position (and continues								4. several false starts but able to start independently without help (and continue riding for 30+ m)
riding)								5. may need encouragement of token support (e.g., finger on back, knowledge that someone is there) 2. needs stabilization but not assistance with speed
								 requires help with speed and stabilization in order to get started (helper needs to walk/ run with bike) to launch
								0. unable to start independently
15. Turns								 can grade turns appropriately able to turn, but unskilled (e.g., looks down, keeps head rigid, lots of correction); better at one direction
								 close standby supervision is required for turns; can only take very shallow turns safely intermittent contact with child, bike or barness
								required during even shallow turns 1. constant contact with bike required on turns
								0. unable to turn/ turns not observed

NAME:	Level of Support						COMMENTS	
		n			DI	<u> </u>		
Skill		Pro	mpt		Physical			
					Assi	stance	9	
	Independent 5	Needs a few tries/ unskiled	Verbal/ Visual cue 4	Demo/ close standby supervision 3	Minimal 2	Maximal 1	DNA/ unable 0	
MASTERING RIDING (con	ntinue	d)						
16. Manages different terrains (e.g., bumps)								 5. manages variety of terrains 4. no terrain changes observed, but child is riding proficiently; some trepidation with changes 3. encouragement or close supervision on different terrains 2. minimal support is required on rough terrains 1. child able to ride but needs full support on other terrains 0. different terrains not observed, child not able to ride and not likely to be able to manage
17. Can ascend small hills								5.able to climb hills 4. can handle small grades with encouragement or deviation in path 3. requires standby supervision as may veer significantly 2. needs minimal assistance for power to go up hills 1. full assistance required
18. Steers between/ around obstacles								 sable to manage obstacles can manage slalom with encouragement; may occasionally bump into obstacles (and keep riding) requires close standby supervision when maneuvering falls when attempting to go between obstacle or requires intermittent support constant support required to manage obstacles/ steering unable to steer between obstacles/ unaware of environment
19. Rides on narrow path								 rides on sidewalk or one side of path able to manage sidewalk but will occasionally veer off close supervision as veering may compromise safety intermittent support required to remain on path/safe maximal support required for child to feel comfortable riding on sidewalk unable or unwilling to ride on narrow path
20. Safety								 child is starting to master higher level skills (e.g., shoulder checks) and consistently follows safety rules child needs reminding to follow rules of riding (e.g., right side, stopping at corners) child is able to ride but may do things that are unsafe (e.g., overestimate abilities, inconsistently follows "stop" command, poor awareness of environment). Can be redirected. needs intermittent physical cueing to be safe needs consistent physical cueing to be safe o. child does things that are deliberately unsafe.

Chapter 3: Guiding discovery of two wheeled cycling strategies

for a youth with Down syndrome: An in-depth case study

[in preparation for submission to Physical and Occupational Therapy in Pediatrics]

Abstract

Over 60% of children with Down syndrome are unable to ride a bike, limiting their participation in one of the most popular activities of childhood (Buckley et al., 2002). Current studies suggest modified bicycles may be the only way to facilitate success, but no studies have yet investigated teaching methods that are not reliant on adapted equipment. As part of a larger study, twowheeled cycling was taught to a 19-year-old youth with Down syndrome using key features of the Cognitive Orientation to Occupational Performance (CO-OP) approach. An in-depth case analysis is presented here to describe how this youth mastered the activity of riding and the CO-OP strategies that supported the skill acquisition. The positive results provide proof of principle that CO-OP can be an effective way to teach cycling to individuals with Down syndrome using standard equipment and warrants further evaluation to determine its potential use with others with intellectual disabilities.

Keywords: cycling, CO-OP, Down syndrome, strategy use

Riding a bike is an excellent opportunity for facilitating community participation and social interaction, with 82% of Canadians cycling for leisure or recreation (Cragg et al., 2006; Menear, 2007). Most children learn to ride a two-wheeled bicycle around the age of 6 (Hansen et al., 2005), though there can be a significant range in age, particularly for children with physical or intellectual disabilities (Klein et al., 2005). Little research has been done on how to teach cycling skills, with even fewer studies including children with Down syndrome; however, the potential to positively impact these children is significant. Only 9-36% of individuals with Down syndrome ever learn to ride a two-wheeler, with many families giving up after years of failed attempts (Buckley et al., 2002; Ulrich et al., 2011).

Down syndrome is a genetic condition that affects growth, development and participation in daily activities, and is one of the leading causes of intellectual disability (Daunhauer & Fidler, 2011). Most individuals with Down syndrome have intelligent quotient (IQ) scores that fall in the moderate range of cognitive impairment, though these scores can vary by 60 points (Carr, 1994); a significant positive correlation exists between IQ and function (Rihtman et al., 2010). Compared with children with other intellectual disabilities, children with Down syndrome demonstrate relative strengths in visual-spatial processing, non-verbal memory, and receptive vocabulary and relative challenges in balance, working memory capacity, verbal short-term memory and expressive language skills (Daunhauer & Fidler, 2011; Næss et al., 2011).

Of the programs that teach cycling to individuals with Down syndrome, many use adapted equipment. The most widely reported of these programs is "Lose the training wheels," now iCan Bike (iCan Shine, 2014). ICan Bike uses a dynamic systems perspective by facilitating cycling success by modifying the task and the environment (Burt et al., 2007). Children begin riding indoors on a modified bicycle with a large flat wheel, and progress through

bikes with wheels of increasing curvature and decreasing stability (Klein et al., 2005). While the majority of children with Down syndrome (56-73.3%) who participate in iCan Bike interventions learn to ride 9- 30 meters (MacDonald et al., 2012; Ulrich et al., 2011), most do not tend to learn such aspects of bike riding as self-starting and braking. When these skills are considered, only 16.7% achieve mastery (MacDonald et al., 2012). As such, the generalizability and transferability of skills to community riding may be limited.

The Cognitive Orientation to daily Occupational Performance (CO-OP) approach has been used to teach a variety of motor skills including cycling to individuals with developmental coordination disorder (Bernie & Rodger, 2004; Miller et al., 2001; Ward & Rodger, 2004), autism spectrum disorder (Phelan et al., 2009), and stroke (McEwen et al., 2009). Adapted equipment is not typically used in CO-OP, though it may be brought in temporarily to enable learning (Polatajko & Mandich, 2004). CO-OP has a specific protocol and several key features, described in detail by Polatajko and Mandich (2004). It draws upon behavioral, cognitive and motor learning theories and focuses on guiding children to discover cognitive strategies to solve motor performance problems. Following client selection of 2-3 goals, the global problemsolving strategy of *goal-plan-do-check* is introduced. For each of the goals selected, the client is guided iteratively to come up with plans of what needs to be done to perform the skill. After the plan is implemented, the client is guided to check first if the plan was done, and if so, if it was effective. Throughout the CO-OP sessions, the therapist conducts a dynamic performance analysis, which involves determining the client's motivation and task knowledge as well as identifying areas of task performance breakdown (Polatajko & Mandich, 2004). The analysis allows the therapist (and ultimately, the clients themselves) to identify domain specific strategies to solve the performance problems (Hyland & Polatajko, 2012). When it is clear the client does

not understand the task, guided discovery may involve the therapist supplementing the client's task knowledge, or breaking down the skill into subtasks. When the client has the idea of what to do but needs support, strategies might involve calling attention to the client's body position or movement sequence, or helping a client visualize a certain movement or sequence by use of a mnemonic or rote script (Polatajko & Mandich, 2004). Several other principles that enable learning may be used throughout the sessions including direct teaching, modeling, chaining, prompting and fading. In addition, each of the 10 sessions is structured to promote learning in a fun way, and to work towards independence, generalization and transfer with the support of parents or significant others.

CO-OP requires that individuals have adequate attention and motivation to participate in the intervention process as well as the cognitive skills and language proficiency to understand and participate in the goal-plan-do-check process (Polatajko & Mandich, 2004). Consequently, it is uncertain if CO-OP could be applied to individuals with Down syndrome as a result of their difficulties in these areas, or if this client-centered intervention focused on guided discovery would help children with Down syndrome learn to ride a two-wheeled bicycle. Accordingly, a multiple baseline design single case study was undertaken to examine the possibility of using this approach to teach bike riding to children with intellectual disabilities. The findings of that study, reported in Chapter 2, were very positive; thus it was considered instructive to examine the specifics of the process in an in-depth case study—reported here.

Aims

- 1. To describe the learning trajectory and skill acquisition of a youth with Down syndrome learning to ride a two-wheeler
- 2. To carry out a detailed analysis of two key features of CO-OP (guided discovery and strategy use) to determine their utility for a person with Down Syndrome

Method

Study Design

As part of initial investigations of the applicability of CO-OP with individuals with intellectual disabilities, an in-depth case analysis was undertaken to determine the process and outcome of CO-OP. This participant was enrolled in a larger study looking at the effectiveness of CO-OP for teaching cycling to individuals with intellectual disabilities (Chapter 2). The current study involved secondary analysis of data collected in Chapter 2 including additional coding of videos and review of field notes, information from a prior learn to ride course in which the first author was involved, and parent and participant interviews. The University of Alberta Research Ethics Board approved all procedures.

Participant and Settings

Characteristics. Justin,¹ a 19 year old youth with a diagnosis of Down syndrome participated in the study. He was 152 cm tall, weighed 76.7 kg, wore glasses and used a hearing aid in one ear. Justin's father described him as an active but fairly cautious child with definite sensory preferences (i.e., did not like the feeling of grass or rain). Based on his performance on the Stanford Binet-5 (Roid, 2005), administered in September 2013, he had a moderate cognitive delay (IQ= 53). His visual spatial skills, non-verbal memory and problem solving skills were better developed than his ability to recall verbal instructions and reason or express himself with words.

Biking history. When Justin was younger, his parents tried "a few times" without success to teach him to ride a two-wheeler. For the past two years, he has used a bicycle adapted with large outriggers and his parents reported him riding it about 20 times/ year. They revisited

¹ A pseudonym, Justin, replaced the participant's name to protect his identity.

the possibility of a two-wheeler after an email from the local Down Syndrome Society introduced them to a free learn-to-ride course for children with disabilities, with which the first author was involved. Justin's family purchased him a new mountain bike prior to his participation in the course. At the outset of the learn-to-ride course, his father rated Justin learning to ride as important as indicated by a score of 8 on a 10-point scale. His father estimated Justin would give learning to ride a score of 2/10 for importance, and Justin confirmed he did not really want to learn. Cycling skills in the learn-to-ride course were presented in a structured and sequential manner with a strong focus on skill practice. All five² 50 minute sessions were conducted in a parking lot, on a shallow hill and on the trail system of a local park over a seven-week period. Justin learned to coast (pushing the bike with both feet on the ground and gliding) but he did not learn to pedal while steering in a straight line, despite being able to so on his adapted bicycle. He consistently required two volunteers to assist him with balance when his feet were on the pedals and he often froze on the bike (e.g., needed help to bring his feet from the pedals to the ground). As suggested by Ulrich et al. (2011), fear of falling was a major constraint in Justin's ability to learn to ride. Following the learn-to-ride course, Justin indicated that he was not good at riding and did not like to ride without training wheels. Parent ratings before and after the learn-to-ride course of Justin's cycling performance and their satisfaction with his skills are reported in Table 3.

Procedure

CO-OP intervention. Justin and his father consented to participate in a CO-OP research study following completion of the learn-to-ride course. He was one of seven participants in a single case multiple baseline design across participants (described in Chapter 2). His father was

² one session was cancelled due to rain

interviewed approximately one month following the last session of CO-OP. The first author, trained by one of the developers of CO-OP, administered the intervention which differed slightly from the typical CO-OP approach. First, Justin did not select cycling as a goal he wanted to work on. Rather, he agreed to participate in the study. Second, cycling was the only skill addressed in the 8 sessions, although 3 skills are typically targeted in a standard 10-week CO-OP intervention (Polatajko & Mandich, 2004). Third, generalization and transfer of CO-OP cognitive strategy use was not a focus, although generalization and transfer of bike riding was. If homework was discussed, it dealt with practicing the cycling skills learned within the sessions rather than generalizing the CO-OP strategies to other skills. Justin's father participated in all sessions and was aware of the cognitive strategies used.

The global strategy of goal-plan-do-check was taught in the first intervention session, reinforced with written text and verbally throughout the session, and used in each subsequent session. Justin was asked to set sub-goals (i.e., choose a cycling focus) each session which then became the focus of the intervention session. When the goal of the session was unclear (e.g., if Justin did not want to select one), the areas of performance breakdown, as determined by the performance analysis, were used to focus the session. Throughout the intervention, Justin was guided to discover domain specific strategies to meet the ultimate goal of learning to ride independently.

Data Collection and Analysis

Systematic behavioral observation of Justin's videotaped sessions was conducted in addition to the measures of distance, time and skills described in Chapter 2. Measures of performance and satisfaction were obtained from both Justin's prior learn-to-ride course (which

used a similar format of questions to that of the study) and the current study. Each session was transcribed and the entire video-recorded intervention used for coding.

Behavioral observations were used to capture three things. First, the main sub-goal identified by Justin and/or the skill worked on most frequently in the session was coded as the primary focus. Second, Justin's comments and responses as a result of specific questioning, coaching and/or demonstrations were examined. These were used to indicate the principles he learned through guided discovery. Third, domain specific strategies were identified based on descriptions adapted from Mandich et al. (2001; see Appendix B). With the exception of supplementing knowledge and task specification, domain specific strategies were only identified if they occurred during times Justin was in the "do" phase. Each strategy was considered to be mutually exclusive, as only one was coded at any one point in time (e.g., when a skill was coded under "task specification," other strategies used within this time period were not recorded).

Inter-rater agreement was determined for the skill checklist scores and the behavioral observations using weighted and unweighted kappa respectively (Advanced Analytics, 2013). Though inter-rater reliability had previously been established for the skill checklist (see Chapter 2), scores for the intervention sessions were determined differently as they also included skills observed during the sessions themselves (rather than just the probes). A second rater coded two randomly selected intervention sessions (28.6%) after first training on a third session to reach 80% or higher agreement. Fidelity in the implementation of the intervention was previously established (see Chapter 2).

Results

Learning Trajectory at Baseline

When Justin began the baseline phase of the larger study, he pedaled no more than 2 rotations if at all. He frequently overcorrected when steering, causing the bike to lurch and was only occasionally successful in riding in a straight line. A cycling harness was a necessity for safety and Justin opted to also wear kneepads and elbow pads. Maximal support was initially provided through both the bicycle seat and the harness as Justin refused the help of a second adult. At baseline, Justin had already mastered all of the stationary skills associated with cycling (e.g., moving kickstand, getting on and off the bike) as well as coasting and stopping.

Learning Trajectory During Intervention

Justin showed a gradual increase in his cycling skills as soon as intervention began (Figure 5). Inter-rater reliability for the cycling skills checklist was very good during this period (κ_w = 0.891 (p<0.001), 95% CI [0.828, 0.955]), with reliability values consistent with those found in baseline (as reported in Chapter 2). Initially, Justin's cycling time and distance were most strongly related to his cycling skills. Once Justin could consecutively pedal 5 or more rotations at a speed surpassing 2m/s (session 2), he experienced his first success riding independently (3 seconds). This is in line with the suggestion by Brown (2008) that the critical speed of being able to ride without wobbling is 2.2m/s. In session 3, Justin experienced a near fall and a decrease in the observed skills and cycling distance was noted (Figure 6). Over this session and session 4, he was guided to discover how his body position could contribute to a fall, as well as the fact that leaning (e.g., around a corner) did not always mean he would fall. In session 5, once he was consistently able to pedal 15 consecutive rotations, Justin became far more confident on his bike as evidenced his ability to ride in the parking lot for periods of 10 or more seconds

without support and his laughter and positive comments following his successes. At this time, an almost exponential increase in distance and time cycled was seen that continued into the followup phase (Figure 6). As he gained speed and confidence with riding, physical supports were faded.

Learning Trajectory at Follow-up

Improvement in distance, time and skills mastered was seen even after completion of the CO-OP intervention. Without having practiced after the final intervention session, Justin demonstrated mastery of 5 new skills on follow-up: riding on a narrow trail, managing different terrains, turning corners, ascending hills and managing obstacles.

Once Justin had mastered the basic skills of riding and launching his bicycle, the environment became the biggest influence on his cycling distance and time. Justin's cycling speed over the 5 follow-up sessions averaged 2.76 m/s and ranged from 2.4 m/s on an unfamiliar busy trail with hills, to 3 m/s in the flat neighborhood around his home. Yield signs encountered on the route limited the maximum distances Justin cycled in his neighborhood, and the absence of hills explained the apparent decrease in skills shown for the second and third follow-up sessions. On an unfamiliar trail system (follow-up 4), the distance he rode was limited by a large hill; Justin, who was following his dad on the trail, forgot to brake, lost control and fell into the grass as the path curved at the bottom. He was unhurt except for a few scrapes. Justin's final intervention session occurred on the familiar trails used in the baseline and intervention sessions. Here, Justin rode his farthest distance (665m) with an average speed of 2.9 m/s, and demonstrated independence in all cycling skills assessed (including descending a steep hill). For details on measurement of distance and time cycled as well as cycling skills mastered see Chapter 2.

Key Features of Learning with CO-OP

Global strategy use. Justin remembered two to three words of the goal-plan-do-check process (typically goal and plan). He required consistent prompting to implement this global strategy. Though the overarching goal was to ride a bicycle, Justin was asked to set sub-goals for every session. For sessions 1, 2 and 4, his identified goal was "balance". In session 4 he had figured out his most efficient launch strategy so for session 5, he also wanted "to coast a bit then bike like last week." When Justin did not want to set a goal, challenges identified in the performance analysis (e.g., turning) were targeted. Justin's plans in the initial sessions and when working on unfamiliar skills were prompted by the guided discovery techniques of "one thing at a time", "ask, don't tell" (e.g., "what are you going to do with your feet?") and "coach don't adjust" (e.g., "is there a different way we can start?"). Modeling by "making it obvious" was also used. Beginning in session 5, he demonstrated some independence and initiative in setting plans. These plans focused mostly on routes he wanted to navigate, and became increasingly complex as he experienced success. In session 7, one of Justin's plans included 9 steps and involved straight riding, corners, full turns, obstacles and stopping. The time spent in the "do" phase increased over the course of the intervention as Justin's confidence and skill increased and his rides became longer. In all of the sessions, the "check" phase was fairly passive and consistently required therapist prompting. On some occasions, Justin forgot his initial plan so was unable to effectively check if it was successful. This only happened when his plan was prompted; he was consistently aware of plans he had come up with independently. On other occasions, Justin was not aware of his behavior, and checking was only possible through review of the video. Despite these challenges, with scaffolding Justin was able to identify, adjust or

abandon his plans in order to achieve his goal. The focus of each intervention session and the targets of the guided discovery are outlined in Table 4.

Guided discovery and domain specific strategy use. The "ask, don't tell" and "make it obvious" techniques were most frequently used to help guide Justin's discovery of plans and domain specific strategies. The questions that were asked invited Justin to make an evaluation (e.g., is it easier to go in a straight line when you are going fast or slow?) or solicited facts (e.g., how do you stop yourself from going too fast down a hill?). His abilities were consistent with his cognitive level; he could not answer questions that required analysis, though was generally on topic with his responses (e.g., Q- "you don't want to go too slow- why?" A- "If I fall again, then I do a brakes on and step"). When Justin did not have sufficient task knowledge to perform a skill and it was not possible for him to discover the strategies on his own or with prompting, the solutions were made obvious through direct teaching or modeling. These solutions (e.g., how to launch the bike) were demonstrated wherever possible in a way that invited Justin to choose which method worked best for him. Typically 1 to 3 sub-tasks were targeted and practiced in each intervention session. Skills often had to be presented several times during a session or in several subsequent sessions for Justin to master them.

The domain specific strategies used during intervention are outlined in Table 5. All strategies were used, though "feeling the movement" was only used once. Discussion about or practice of any component of a skill (e.g., where foot needed to be positioned on the pedal so that it did not hit tire) was coded under task specification. This was the most commonly used strategy, and was always prompted. "Attention to doing" and "body position" were the next most common strategies and often had to do with calling Justin's attention to his feet (e.g., if he was pedaling or where his foot was on the pedal) or where he was looking. Reliability for the

strategies used was lower than training values and was moderate κ = 0.618 (p<0.001), 95% CI [0.391, 0.844], presumably because raters identified both the timing and the strategy. Both timing of the strategy (i.e., whether conversation could be coded as a domain specific strategy) and type of strategy coded were sources of disagreement, with task specification and supplementing knowledge being coded differently most frequently.

Once Justin had the idea of a skill, there was very little strategy-use associated with "do". This was particularly evident in session 5 where Justin spent the most time riding and used the fewest strategies. Though only verbalized behaviors could be coded, Justin may have been using one or more strategies silently as suggested by Justin's strategy of positioning his pedals parallel to the ground before launching his bike; this plan was never suggested nor did Justin ever verbalize what he was doing.

Individual interviews. Immediately following the final intervention session, Justin was asked for feedback on his experiences. He liked practicing on his own better than in the learn-to-ride class and he "liked the plans" because they "made it easy." Justin rated his ability to ride as both "I can almost ride by myself" and "I can do it!" and reported a score of 5/5 when asked how much he liked to ride. His father's answers mirrored these sentiments, reporting that working one on one "allowed for more trust building and relationship building" and that "giving (Justin) choice and empowering him with his choice" was particularly helpful. He rated Justin's performance in cycling as 7/10 (see Table 3) as he could not quite do everything needed to participate in bike rides with friends and family. He reported that Justin had practiced 3 to 4 times during the study.

After the follow-up sessions, Justin's parents returned the adapted bicycle he had been borrowing to the Down syndrome society, as he was now able to ride a two-wheeled bike. Since

the conclusion of the study, they reported Justin had gone on a few bike rides, mostly with his siblings, but that he was not overly enthusiastic about biking. Despite this, they were thrilled with his ability to ride and reported satisfaction with his performance as 10/10.

Discussion

This case study investigated the learning trajectory, strategy use and guided discovery of a youth with Down syndrome. First, the fact that Justin was not able to balance independently until he was able to pedal consistently at a speed of over 2 m/s suggests that certain skills may be intricately tied with success. It appears that once a critical threshold of skills is reached, progress comes quickly. Before participating in the CO-OP intervention, Justin's biking exposure consisted of no reported home practice, five approximately 50 minute learn-to-ride classes and five 5-10 minute sessions of baseline probes spread over 3-5 days. Following two sessions of CO-OP over a one-week period, Justin rode independently for 3 seconds. After 6 more CO-OP sessions over the span of five more weeks, Justin was completely independent riding a twowheeler; supervision was required for road safety. While this was not a comparative study, it should be noted that a similar amount of training was provided in the CO-OP sessions (400 minutes) as is offered in the 5-day, 75 minute iCan Bike camps, albeit over a much longer timeframe (2 months). The challenges experienced by many children with Down syndrome in iCan Bike of not being able to start or stop independently (MacDonald et al., 2012) were not observed in this case, although other challenges arose. Despite efforts to minimize risk, Justin experienced a few minor falls over the sessions. However, over the three incidents his reaction to these falls changed. In session 3, his first near fall, he avoided the activity he perceived to have contributed to the loss of balance- in this case pedaling. In contrast, in session 7 he asked to use safety equipment after his fall and in follow-up 4 he was able to verbalize how to avoid a

similar occurrence. As is mentioned in other studies (Little, 2006), the presence of risk (and learning from his mistakes) made a positive contribution to Justin's learning.

Additionally, several CO-OP strategies were used in the intervention. While the global framework provided structure and engagement, Justin was not able to recall or apply the steps of the goal-plan-do-check process independently, suggesting it may not be effective for individuals with cognitive impairments. Similar challenges were found when looking at the effectiveness of CO-OP for individuals with acquired brain injury (Missiuna et al., 2010). While Justin evidently enjoyed and was successful coming up with his own plan, it had less to do with *how* he was going to perform a skill, than *where* he was going to cycle.

Third, the guided discovery of CO-OP allowed for more therapist-child interaction and collaborative decision making than Justin experienced in the learn-to-ride course. It allowed for Justin to choose what he wanted to focus on and helped structure the sessions in a way that was meaningful to him, rather than pre-supposing the sequence in which cycling skills should be introduced. Allowing him choices in determining the course of events and making it fun in the sessions may have had a similar positive effect on engagement as self-selecting bike riding as a goal.

The types of domain specific strategies used by Justin were also similar to those seen in other studies, with "task specification" being used most consistently (Bernie & Rodger, 2004; Ward & Rodger, 2004). To help Justin discover how a skill could be performed, direct questioning (often with the support of gestural cues) and making it obvious through modeling was most successful. Because of their challenges with verbal short-term memory and expressive language skills, and strengths in non-verbal memory and visual-spatial processing (Daunhauer & Fidler, 2011), the use of visuals and modeling is a particularly useful addition to the verbally-

based cognitive strategies of CO-OP for individuals with Down syndrome. These challenges also explain the need for enabling principles that promote learning such as direct teaching, modeling and prompting.

The case study methodology has obvious limitations, such as the inability to generalize to other persons with Down syndrome. As per the tenets of CO-OP, Justin was guided to discover his own movement solutions. His physical characteristics, his cognitive and behavioral profile, his bicycle, the environment he was riding in and his previous experiences all influenced the solutions he generated just as his interest in and his family's commitment to cycling will influence his subsequent participation. Video recording was not continuous and did not capture all of the dialogue between Justin and the instructor (first author). Transitions between locations (e.g., parking lot to trail) were not usually recorded in order to conserve battery life. Wind and environmental noise (e.g., lawn mowers, buses), difficulty understanding Justin and the proximity of the videographer to the participant also limited what dialogue could be transcribed. It is therefore possible that some strategies are under-represented. Finally, participation in a learn-to-ride course prior to this study likely had an influence on his skills (e.g., he learned to coast in the program) as did his years of adapted bike use.

Conclusion

This study shows that mastery of a two-wheeled bicycle is within the realm of possibilities for individuals with Down syndrome, even as they enter young adulthood. Further research on the learning trajectory for other children with Down syndrome may focus recommendations to help more children with this diagnosis learn to ride. Based on the results of this case study, aspects of the CO-OP approach, specifically guided discovery and domain specific strategies show promise as an effective way to teach cycling to this population, given some modification to accommodate

for cognitive and language challenges. While the global strategy did not appear to be particularly useful, with scaffolding, Justin was able to identify various domain specific strategies that enabled success. In particular, "task specification", "attention to doing" and "body positioning" have potential for use with individuals with Down syndrome. Based on the preliminary positive findings, further investigation should examine the potential application of CO-OP with this population.

Table 3

Ratings of Performance and Satisfaction

	pre course	Post course/	post CO-OP	post CO-OP
		pre CO-OP	intervention	tonow-up
Performance*	1	3	7	8
Parent rating				
Satisfaction	2	5	9	10
Parent rating				

*ratings range from 1 to 10 with higher scores being more optimal

Table 4

Focus and Guided Discoveries in CO-OP Intervention

Intervention	Focus	Principles learned through guided discovery
session		
1	Balancing on bike -	a) if you stop pedaling, the bike will slow down
	straight riding	b) it is harder to balance when the bike is moving slowly
2	Balancing during launch	a) it is easier to start with gliding and then put feet on pedals
		b) when you position the pedals, you know where they are even without looking at them
3	Building confidence after a near fall ³	a) toes can hit wheel if foot is too far forward on the pedal
4	Balancing when	a) which way bike leans when going around a corner
	starting to pedal	b) how to position pedals; what pedal position makes the
		bike go further when you step on it
5	Balancing and riding	a) bike has to be moving before you can put both feet on
	(in parking lot)	pedals and stay balanced
6	Balancing and riding (on trail)	a) speed down a hill can be controlled by pumping brakesb) can go straighter when you pedal faster and when you keep your arms strong
7	Balancing (how to	a) when foot is in "tiptoe position", it doesn't hit the
	avoid a fall)	wheel when you turn
		b) if you push handlebars too far, you will fall
8	Turning	a) you need to look where you want to go when turning a
		corner
		b) it is hard to balance around a corner if you are going too slow

³ Focus provided by therapist as Justin did not want to choose a goal



Figure 5. Cycling Skill Progression.

* decrease in skills shown in session 8 may have been partially a result of a videography error; not all skills practiced were captured on film.



Figure 6. Distance and Time Cycled (Pre and Post Intervention)

* Only 7 intervention probes are shown as there was a filming error in session 8; straight riding was not captured on video.

Table 5

Domain Specific Strategies Used

	I1	I2	I3	I4	15	I6	I7	I8	Total
Body position	2	0	2	0	0	1	5	5	15
Attention to doing	1	0	2	2	1	5	4	1	16
Task specification	3	9	6	6	2	3	3	1	33
Supplementing knowledge	1	2	3	4	0	3	3	1	17
Feeling the movement	0	0	0	0	0	0	0	1	1
Verbal motor mnemonic	0	0	0	1	1	3	2	0	7
Verbal rote script	2	0	0	1	1	0	1	0	5
Total	9	11	13	14	5	15	18	9	94

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		inperono
Strategy	Strategy Description ⁴	Cycling examples
Body position ⁵	Relative to the task, verbalization of:	"where is your foot on the
	• attention to any body part	pedal?"
	• the shifting of the body	
	• a cue to shift the child's visual focus	"where are you looking?"
	in order to allow for safe completion	
	of the task	
Attention to	Verbalization to cue attending to:	"I see you stopped
doing ^o	• the doing of the task	pedaling"
	• a specific component of the plan that	
	is <u>not</u> being performed or that is	"does your bike balance
	causing skill breakdown	when you are not moving?"
Task	Any:	practicing putting feet on
specification/	• discussions regarding the specifics or	pedals or coasting
modification	modification of the task or subtasks	
	• modification to the task or subtasks	"follow the center line"
	• discussion about, focus on, or practice	tuisling which soon is most
	of specific components of a subtask	trialing which gear is most
	outside of the task (bike riding) as a	appropriate to ride in
<u> </u>	whole	1
Supplementing	Any:	demonstrating/ discussing
task	• successful instruction of task specific	now the position of the
knowledge	information or of how to get task	pedals impacts now far the
	specific information	blke travels on a single
	• modeling (by making it obvious) of	push
	<i>new concepts or strategies</i>	1
Feeling the	verbalization of attention to the feeling	now did you move your
movement	of a particular movement of reflecting on	100t on the pedal)? did
Varia al martan	Now it was completed	you fill it up of slide it?
Verbal motor	Any name given to the component of the	power pedal position to
mnemonic	task or body position that evokes a	snow where pedal is
	mental image of the required motor	positioned for launch
	performance.	"strong arms"
Verbal rota	A rote pattern of batwaan 2 and 5 words	Suong anns "alide alide alide pedel"
script	that are meaningful to the child to guide a	gnue, gnue, gnue, peudi
script	motor sequence or comments by the	"I'm looking up"
	individual on what they are doing	I III IOOKIIIg up
	inairiaaai on what they are abing	

Appendix B Domain Specific Strategy Descriptions

⁴ Strategy descriptions are modified from Mandich, Polatajko, Missiuna & Miller (2001). Italicized sections indicate how definition has been modified

 $^{^5}$ does not include any directive cues such as "feet down" or "head up"

⁶ does not include comments on what is being performed effectively

Chapter 4: On a roll: Discussion, Implications and Future Directions

A large majority of children with Down syndrome (64-91%) are unable to ride a twowheeled bicycle (Buckley et al., 2002; Ulrich et al., 2011), yet research on teaching cycling to this population is sparse and focuses only on what is offered by iCan Bike. This final chapter first explores the advantages and limitations of iCan Bike, and the theoretical and practical advantages of CO-OP. Next, the three goals of the study are revisited: a) to explore the effectiveness of CO-OP for teaching cycling with individuals with intellectual disabilities; b) to evaluate the use and utility of the components of CO-OP for this purpose; and c) to describe the learning trajectory and cycling skill acquisition of a youth with Down syndrome participating in a CO-OP intervention. Further support for the conclusions reached in Chapters 2 and 3 is provided by drawing from the experiences of study participants and by using the ICF framework to consider contextual factors; this information was not appropriate elsewhere. Finally, the limitations and strengths of the studies are discussed, a plan for knowledge translation is outlined, and suggestions for future research provided. In conclusion, a justification for why one method might be chosen over the other is provided.

I Can Bike: Advantages and Limitations

The advantage of iCan Bike's method of modifying the cycling task by adapting the bicycles is that balance can be challenged incrementally as the child encodes effective motor responses (Klein et al., 2005), which in turn can help decrease a child's anxiety about falling (Witter, 2013). That balance mastery can be achieved in as little as four sessions for individuals with Down syndrome is exciting (Burt, 2002), particularly in light of how frustrating mastery of this task can be for them and their families (Ulrich et al., 2011). However, there are several limitations to the program, the first being the cost. The adapted bicycles of iCan Bike are not

available for purchase or for rent and can only be accessed through one of their 5-day camps (J. Sullivan, personal communication, November 4, 2012). While the camps are mobile, the cost of bringing one to a community in Canada is \$14 000 (Falloon, 2014), significantly limiting the number of families that might be able to access the program. A second limitation is that compared to children with other disabilities, individuals with Down syndrome appear to have far less success learning to ride. MacDonald et al. (2012) reported that following 75 minutes of intervention for five days, 82.9% of children with autism and 63.3% of children with Down syndrome could ride for 30 meters and brake. When launch was added, 70.7% of children with autism were successful compared with only 16.7% of children with Down syndrome (MacDonald et al., 2012). The reason for this discrepancy has not been explored.

Even when cycling skills are learned in the iCan Bike program (or using their equipment), these skills are often not retained and do not transfer to the home environment. Burt (2002) investigated ten children between the age of 7 and 11 as they learned two-wheeled cycling using iCan Bike's adapted equipment. Sessions were held indoors and ran three days each week for 45 minutes. Within seven sessions, all ten children were able to independently ride a distance of 12 meters, although only one of the three participants with Down syndrome was able to maintain the skill, and none of the three was able to navigate obstacles. Burt also reported that one child refused to ride outside. Witter (2013) and MacDonald et al. (2012) found similar issues with transfer to the home environment following participation in a typical 5-day iCan Bike camp; a significantly larger proportion of children were considered riders in the course than continued riding in their communities. Klein et al. (2005) suggested that 1/3 of children that learned to ride were not able to generalize these skills to their communities, though the limited time the bikes are available may contribute to this. Though advantages of iCan Bike's method

are irrefutable, particularly for children experiencing high anxiety about falling, there is enough difficulty with generalizing the skill to their own bicycles and transferring the skill to the community to warrant the exploration of other possible methods of cycling instruction for individuals with intellectual disabilities.

CO-OP: a Feasible Alternative

Cognitive Orientation to daily Occupational Performance (CO-OP) has been used to teach several motor skills including cycling to individuals with various disabilities (Dawson et al., 2009; Mandich et al., 2003; McEwen et al., 2009; Phelan et al., 2009)and is a feasible alternative to iCan Bike for several reasons. First, adapted equipment is not necessary for the delivery of the intervention, though it can be used to facilitate learning (Polatajko & Mandich, 2004). As a result, the goal of cycling can be practiced entirely in a natural environment, which has the added advantage of providing contextual interference (e.g., variability of surface, environmental distractions) not seen in an indoor environment. Second, being a task-oriented approach in which clients set their own goals, CO-OP has the advantage of ensuring the prerequisite of motivation. With scaffolding, clients are guided to discover strategies that enable them to achieve these goals (Polatajko & Mandich, 2004). Third, in performing a dynamic performance analysis to identify areas of skill breakdown, therapists are able to consider the influence of the child, the task and the environment (Polatajko & Mandich, 2004).

Summary of Research Findings

Effectiveness of CO-OP. As there are no published reports of CO-OP with individuals with intellectual disabilities, the first research goal was to examine its applicability when teaching two-wheeled cycling to this population. A multiple baseline design was used to

determine if the introduction of the CO-OP intervention would cause changes in participants' ability to cycle (Kratochwill et al., 2010). The CO-OP intervention was modified in three ways: the goal of cycling was given to the child rather than being one of three goals chosen, parent involvement was encouraged but not required, and the intervention format consisted of eight rather than ten sessions. As each of these factors would have increased the likelihood of a successful result (i.e., the youth would be more motivated to cycle if it was their goal, the parents would be more likely to practice the skills, and two additional sessions would be offered), not having these components resulted in a bias against finding CO-OP to be effective. A successful outcome despite this bias would increase confidence in the effectiveness of the key features of CO-OP that were retained, but also in CO-OP in its pure form. Additional pre-post measures of performance and satisfaction examined the clinical validity of the intervention.

At the outset of the study, while none of the seven participants were able to ride a twowheeled bicycle farther than 1 meter or for longer than a couple of seconds, many were independent with between 20% and 45% of the basic cycling skills measured by the checklist. Most participants had either never been on a two-wheeled bicycle or had not attempted riding one in several years, having participated in cycling in a different form (e.g., adapted bicycle, bicycle with training wheels). Witter (2013) noted similar findings in an iCan Bike camp; while some participants had recent experience on other types of bikes, over half were not riding at all. Because of participants' lack of cycling experience in the current study, it was often necessary to demonstrate how the specific bike components worked (e.g., kickstand and brakes) or how certain skills were performed (e.g., coasting on the bicycle) before they could be assessed. As a result, some participants improved slightly in their cycling skills over the baseline period, but this was not significant, nor did it impact their ability to ride independently. For all but participant 4, stability at baseline was demonstrated, with none being able to ride a distance of over 2 meters by the end of the five probes. Participant 4, who had been practicing on an almost daily basis with his father, showed an upward trend in both his skills and his ability to ride; he was able to cycle a distance of 30 meters by the fifth baseline session.

Overall, six participants demonstrated a change in the trend and the level of cycling skills following implementation of the CO-OP intervention. With the exception of participant 4 whose progress may have been due to his regular practice with the help of his father, the replication across participants strengthens the evidence that the changes were due to the introduction of CO-OP (Kratochwill et al., 2010). At follow-up, these six participants were independent with between 55% and 100% of skills and were able to cycle independently at speeds of between 2.4 and 4.8 m/s over 31 to 1756 meters. As evidenced by these numbers and the graphs presented in Chapter 2, variability of distance and time cycled and skills demonstrated was high within and between participants, as well as within and between sessions. Burt (2002), noting increased variability prior to cycling skill mastery, explained this as "the participant (being) in the process of exploring increased degrees of freedom and demonstrating self-organization prior to acquiring and refining new skills" (p. 130) as per the dynamic systems theory. That 6 of the 7 participants demonstrated this same pattern and ultimately learned to ride without assistance strongly suggests that this is true, and corresponds with the large effect sizes found in this study as defined for single case multiple baseline research (Petersen-Brown et al., 2012).

Similarly, positive changes were seen in parent ratings of children's cycling performance using a modified version of the Canadian Occupational Performance Measure (Law et al., 1990). At baseline, COPM ratings of performance and satisfaction ranged from 1-3 out of 10 and 1-8 out of 10 respectively; five parents reported a score of 1/10 for both performance and

satisfaction. There were two exceptions. The father of participant 6 rated his child's cycling performance as 3/10 and his satisfaction with these skills as 5/10 as his son had completed You Can Ride 2's learn to ride class and had improved slightly in his pre-cycling skills. The father of participant 4 who had rated his child's cycling performance as 2/10 and his satisfaction as 8/10, reported that his sole goal of the summer was to teach his son to ride a bike, and that they had already started practicing. Following intervention, significant increases were noted in the COPM ratings of performance ($M_{increase} = 6$) and satisfaction ($M_{increase} = 7$) for participants 1 to 6, which suggests that the CO-OP intervention produces clinically significant changes. Taken together, the results are strongly supportive of CO-OP as an effective intervention for individuals with intellectual disabilities.

Utility of key features of CO-OP. The second research goal examined the use and the utility of the components of CO-OP as they apply to individuals with intellectual disabilities. These components include cognitive strategy use, guided discovery, domain specific strategies and enabling principles. Client centered goals, parent involvement and specific intervention format were modified from the original format of CO-OP in that cycling was presented rather than chosen, parent involvement was not required and the intervention consisted of 8 rather than 10 sessions. Yang et al. (1999) and Missiuna et al. (2010) concluded that individuals with intellectual disabilities are not consistently able to apply, retain or generalize a global strategy (e.g., goal-plan-do-check). These findings were mirrored in both the case study (Chapter 3), and within the context of the larger group. While the four steps were helpful to frame the sessions and cue discussions, the words "goal," "plan," "do," and "check" appeared to be too difficult to remember and too abstract for most of the participants to comprehend. Many participants remembered different though similar words (e.g., go or gold offered by participants 5 and 2) or

understood the words to mean something different (e.g., participant 6 defined a goal as "working hard"). "Plan" was the word most frequently remembered from session to session, and only one participant could recall the four steps at the conclusion of the study. If the words in the global strategy were not initially in participants' vocabulary, it is likely that learning and remembering them would have been challenging, particularly since deficits in verbal memory are common in individuals with Down syndrome (Næss et al., 2011). Perhaps for these children, specific training in problem solving as a first step in using the CO-OP approach would be warranted.

Despite the broad language challenges of the participants with Down syndrome, guided discovery was possible and proved an effective method of instruction as evidenced by the learning pattern of participant 6 in Chapter 3. It is likely that the ease of using gestures to demonstrate concepts such as steering and braking contributed to the learning. Wang et al. (2001) found that gestures helped students focus longer on tasks, and served as a model to scaffold performance. Another advantage of guided discovery was that there was no presupposed order of instruction, nor was it assumed that there was a right way to perform a movement. In the You Can Ride 2 class all children were shown how to start by placing one foot on a pedal placed in a mechanically advantageous position and the other foot on the ground and pushing simultaneously. Rather than teaching this method to the study participants, they were guided to set plans and figure out the method that was most effective for them. A particularly strong justification for the use of guided discovery came early in the course of the study as participant 1 was setting a plan for launching her bicycle. When asked where she was going to put her feet to start she stated "one foot on the pedal, one foot on the ground;" however, she executed something quite different. Her actual performance involved placing her pedals in the "start position" as outlined above, then coasting (i.e., pushing the bike forward with two feet)

before putting both of her feet up on the pedals and continuing riding. This was not a method that had been previously introduced (or even seen), it was the participant's own method, and the first time she experienced success starting. This method also turned out to be the most effective way to launch for 3 other riders who chose it over other methods modeled.

As was the case for participant 6, many of the participants adopted a variety of the CO-OP strategies as they learned to ride. In some cases, the strategies were only necessary until a skill was mastered. For example, participants 5 and 6 only required the verbal motor mnemonic of "strong arms" until they figured out how to steer without weaving. Despite the language challenges of the participants with Down syndrome, verbal rote script was particularly effective for many as they figured out how to launch the bike. Participant 5 used a verbal rote script to help with this process, counting as she pushed her bike forward five times as per the strategy discovered by participant 1. Participant 4 used a slightly different method, placing one foot on the pedal and pushing the bike forward with the other; his father cued each step of the process (i.e., left foot on, 4 pushes—1,2,3,4—pedal!). While participant 6 used all of the strategies, task specification was used most frequently, followed by supplementing knowledge, attention to doing and body position. Because task specification included the direct teaching and modeling of specific cycling components, it is likely to be the most frequently used strategy for all participants although this was not assessed. This was also seen in other studies (Bernie & Rodger, 2004; Ward & Rodger, 2004).

The language and cognitive delays of the participants made it necessary to use significantly more enabling principles to promote learning than typically reported in other CO-OP studies. These used included prompting, fading, direct teaching, modeling, chaining, and reinforcement; all participants appeared to benefit from this scaffolding. As conventional two-
wheeled bicycles were used, physical support through the seat and/or a wingman cycling harness was initially necessary to help with balance, but was faded as the youth participated more in the steering process. Direct teaching and modeling was used when participants could not be guided to discover movement solutions on their own, which happened frequently. Praise and specific feedback about performance (e.g., using a stopwatch to measure speed) was provided throughout the intervention; sharing knowledge of performance is thought to be particularly important for learning of continuous skills such as cycling (Newell, 1991). For more complex sequences such as starting, chaining was used. The one enabling principle that appeared to vary between participants was the use of reinforcements. For participants 3, 5 and 6, the intrinsic reinforcement they got from experiencing success appeared to motivate them to try harder. Comments from these participants included "No daddy, I do it!" and "I'm staying focused. I like to stay that always. My eyes staying wide." For participants 1 and 4, extrinsic reinforcement appeared to be more motivating. Physical tokens, such as happy faces to define the length of the session (i.e., the session concluded when 10 happy faces had been earned), or rewards (e.g., from the treat box or from parents) were necessary to maintain engagement which also translated to decreased time spent in intervention. In one case (participant 2), both intrinsic and extrinsic reinforcements appeared to be motivating. In the last case, participant 7 who did not learn to ride, it was difficult to determine if any reinforcement was successful.

Cycling learning trajectory. The final research goal was to describe the learning trajectory and cycling skill acquisition of a youth with Down syndrome participating in a CO-OP intervention. At baseline, neither participant 6 described in Chapter 3 nor the other participants were able to ride any functional distance. When asked to pedal as far as possible, participant 6 quickly removed his feet from the pedals and replaced them on the ground. Some of the others

would complete a half pedal revolution before putting their feet down. The independent skills of the participants varied at baseline and averaged 7 out of 20. Participant 6 had a specific strength in coasting, and struggled with pedaling more than a couple times in a row; participant 1 was easily able to steer in a straight line; participant 5 had no difficulty moving her kickstand out of the way, and participant 7 was consistent with pedaling. Anxiety about falling was a limiting factor for several participants, and at the extreme could be paralyzing. Participant 6 was particularly fearful about this possibility and would occasionally freeze on his bicycle, saying that he could not move to put his feet onto the ground. The use of safety equipment (e.g., knee pads, elbow pads and harness) as well as giving reassurance, building trust, and outlining behavioral expectations, eventually caused this to disappear.

Following implementation of the CO-OP intervention, participant 6 demonstrated gradual improvements in his cycling skills. Once he discovered the importance of pedaling consistently at a certain speed, he was quickly able to increase his cycling distance. This "critical speed" was variable, but for most participants was around the 2.2m/s suggested by Brown (2008); participant 7 never reached this speed. Some of the youth appeared to pull everything together seemingly overnight; participant 2 went from cycling for 2-5 seconds in the fourth and fifth intervention session to riding for almost 4 minutes the following session. For many of the participants with Down syndrome, this jump was noted on or just after the eighth day of CO-OP intervention. Contrary to what was expected, the speeds at which the youth started riding were not necessarily related to their size or the size of their bicycle. Participant 6, who rode the largest size of bicycle in the study (24-inch wheels), rode the slowest at a speed averaging 2.8 m/s. Conversely, participant 1 rode the smallest size of bicycle (16-inch wheel) and averaged 3.2 m/s, and participant 4 rode the fastest on a mid-size bicycle (20-inch wheels) with an average speed of 4.4

m/s. Burt (2002) noted that the riders in her study pedaled at an average speed of 3.15 m/s once they began riding a conventional bicycle. In the present study, the average speed of the six riders was 3.3 m/s.

An ICF Perspective

The framework of the ICF can be used to help consider the influence of environmental and personal factors on cycling acquisition and participation. As seen by Witter (2013), body functions and structures were not limiters to riding, despite the balance challenges of many of the participants. In some skills, such as getting onto or off of the bicycle, the body structures of many of the youth with Down syndrome were an advantage; the flexibility of their hamstrings made it particularly easy to lift their leg over the seat.

Personal factors. Attitudes, personalities, fears and experiences of the youth contributed to their willingness (or refusal) to participate in the cycling activities. Witter (2013) also noted that confidence was a factor that separated the riders from the non-riders. Motivation significantly impacted participation as well, though it was difficult to assess. While three participants (1, 4 and 6) reported they did not want to learn how to ride, participants 1 and 6 were both easily engaged in the activity albeit with different reinforcement. On the other hand, participants 4 and 7 were far more difficult to engage, also in different ways. Participant 4 often protested participating in the session and significant encouragement by the therapist and his father (or caregiver) was required. For the baseline sessions, a visual schedule was necessary to get through all of the skills; his scores of 24 and 25/ 100, the lowest of all participants, were due more to a lack of interest than a lack of skill. During the intervention phase, when asked what he wanted to do, his most frequent response was "home" and while he could usually be encouraged to participate, the use of extrinsic motivators and a shortened session were what

motivated him most. Even during the follow-up phase when he was independent with most skills, he protested as he was riding his bicycle. Participant 7 didn't so much resist participating as he delayed it; he took significantly longer than the other youth in most activities (e.g., walking up the hill, pedaling) and would occasionally avoid answering questions by turning his back to the investigator or hiding behind a tree. He also offered few opinions, often responding to questions or choices with "I don't know" or with the last of the options presented to him. As no cognitive assessment was available for him, it is also possible that his cognitive skills were lower than those of the other participants. He was the only participant who did not demonstrate independent cycling and also made limited progress in his cycling skills.

Environmental factors. While participant 4 did learn how to ride, several environmental factors may have contributed to his success including equipment and family support: a cycling handle was attached to his bicycle and his father practiced with him on a regular basis. Equipment played an impact in the success of the other participants as well. Safety equipment (knee pads, elbow pads and safety harness) was made available for all participants. All bicycles were tuned up by a mechanic and set up to allow for the most confidence and success; brakes were adjusted to make them easiest to engage and seat heights were adjusted so participants could put their feet flat on the ground when seated. Seat adjustments were possible for all except participant 3 whose body proportions made it difficult for her to both put her feet flat on the ground and be comfortable pedaling. Several modifications were attempted to try to optimize her position including installing crank shorteners and moving her seat back relative to the pedals; however, nothing was effective. Ultimately, it was decided that her comfort while pedaling was more important than her stability when standing and her seat was positioned so that only her toes touched the ground when she sat on the seat. As a result, though she had a much easier time pedaling, she was not very efficient at moving her bicycle forward when her feet were on the ground and was less stable in stationary positions than other riders. Other environmental factors such the weather and the bugs also influenced task performance of the participants, most were less interested in cycling when it was too hot, or it was raining; Witter (2013) observed a similar trend, noting the deterrence of weather related factors on cycling.

Family support also played a major influence on cycling mastery. Only the families of participant 1 and participant 7 reported that they did not practice with their children. While it may be coincidental, participant 7 did not learn to ride, and participant 1 though able to ride, obtained an adapted bicycle as her cycling skills did not match the environment she was going to cycle in (i.e., she could not ride on the sidewalk). Conversely, participant 4—whose father practiced with him daily—was the first to learn how to ride despite his lack of interest in the activity. Temple et al. (as cited in Witter, 2013), also found that continued family involvement facilitated cycling. Families for whom cycling was a large part of their lives were more likely to continue the skill, regardless of the level of mastery acquired by their children. Participant 5 who had acquired only an average of 12.4 of the 20 skills by follow-up, subsequently went on a family cycling trip to the mountains. They shared videos of their daughter having mastered several more skills.

Based on the results of these studies, CO-OP appears to be an effective way to teach cycling to individuals with intellectual disabilities. With modifications to accommodate for language and cognitive delays with this population, the guided discovery, domain specific strategies and enabling principles of CO-OP can enhance learning. In addition, there appear to be several patterns in the learning trajectory that would be worthwhile to explore.

Limitations and Strengths

A number of limitations should be noted. First, having all participants start at the same time and systematically introducing the intervention once the response is stable would have strengthened the multiple baseline design (Byiers, Reichle, & Symons, 2012). While this was the goal, difficulties with recruitment and summer scheduling made this impossible and as a result, participants spent varying amounts of time in each phase. In addition, differences in participant engagement resulted in some of the sessions being shorter than the scheduled hour, which may have introduced bias.

A second limitation to the study is the fact that the CO-OP method was modified in a fundamental way that has not previously been tested. In all cases, the parents rather than the participants chose the goal of two-wheeled cycling. While this is contrary to the first key feature of CO-OP of having a client-chosen goal, it allowed for a specific focus on cycling. With the exception of participants 4 and 7, parents attended at least 90% of sessions with their child (i.e., 17/18 visits) and were involved to varying degrees in the intervention sessions. All reported practicing once their children began to demonstrate some skill independence, but only a couple of parents provided information with the exact dates and times that this occurred. The father of participant 4 was the only one who reported practicing cycling on an almost daily basis. Because participant 4 began riding during baseline, even though it is possible certain skills may have been influenced by guided discovery of CO-OP (e.g., launch, braking), this cannot be determined. The external influence of equipment (e.g., harness and bike handle), behavior reinforcements (e.g., happy faces, rewards) and visual supports (e.g., pictures, reviewing videos of performance) also cannot be determined. While all equipment can be classified under CO-OP's enabling

principles, they are also standard in most other therapies and the relative importance of these factors as compared to the metacognitive piece of CO-OP is difficult to distinguish.

A third limitation of the study lies in the measurement by video analysis. Environmental noise and distance from the participant made it difficult to hear some conversations and since not all of the session was recorded (to conserve battery life), certain domain specific strategies may have been missed. In addition, while video analysis allowed for accurate measurement of time cycled, an exact distance measurement was not possible since following the exact trajectory of the rider with the roller tape measure would have been impossible.

While limitations exist, this study is the first to examine the use of CO-OP for youth with intellectual disabilities, and the first to look at cycling skill acquisition with this population using only a standard bicycle. Because the research was inspired by You Can Ride 2's challenges with teaching children with Down syndrome to ride, the lessons learned immediately influenced programming. In addition, the biggest strength lies in the results; 86% of children learned to ride a bicycle with 75% of children immediately generalizing the skill to their home environment.

Knowledge Translation

The plans to translate this knowledge to parents and other professionals are extensive. Oral presentations were given at the Women and Children's Health Research Institute research day (November 6, 2013) and the International Congress of the World Federation of Occupational Therapists (June 18, 2014), and a poster and rapidfire presentation was given at the Canadian Physiotherapy Congress (June 20, 2014). In the future, information will be shared on the You Can Ride 2 website (youcanridetwo.ca) and with the eleven communities across Alberta currently offering the curriculum, and the possibility of presenting at the Canadian Down Syndrome Conference will be explored.

Future Research

To build support for the use of CO-OP with individuals with intellectual disabilities, more complex study designs with a stricter adherence to the CO-OP protocol are necessary. In particular, a study with client-chosen goals, parent involvement and a more dedicated focus on teaching the goal-plan-do-check process should be designed. Given the cognitive delays of the participants, it may still be helpful to limit the number of goals rather than focusing on the typical three goals of CO-OP. While the multiple baseline design provides initial support for the use of key features of CO-OP to teach cycling to this population, future research should include larger numbers, a more diverse group of children (e.g., individuals with other diagnoses, youth from different ethnic backgrounds) and a control group. Ulrich et al. (2011) noted low levels of motivation in 8 and 9 year old children with Down syndrome, and that many of these children used leg fatigue as an excuse to get off of the bicycle. As the youngest participant with Down syndrome in this study was 12, it would be interesting to investigate if results are similar in younger children. In addition, several additional factors should be considered when assessing the cycling learning trajectory using a CO-OP method: the use of visual and behavioral supports, language usage of the investigator, current family cycling participation, previous cycling exposure and parent involvement in cycling practice. A longer follow-up time should look at if cycling skills are maintained over the winter months. As an extension to the current study, it would be possible to compare the results of participant 6 related to strategies with the results of the other participants to get a more accurate picture of guided discovery and strategy use in this population.

Future research into cycling skill acquisition with individuals with intellectual disabilities should consider a microgenetic approach which examines change as it occurs and attempts to

identify and explain its underlying mechanisms (Flynn, Pine, & Lewis, 2006). It involves taking multiple repeated measurements over times of transition and "provides an opportunity to identify different groups which may require different treatment or intervention styles" (Flynn et al., 2006). This method would be a particularly beneficial way to compare skill acquisition using the CO-OP method with that of other methods of cycling instruction, such as iCan Bike.

Recommendations

Several recommendations can be made to improve the You Can Ride 2 course based on the results of this study. These suggestions may also be helpful for other programs offering cycling instruction for individuals with intellectual disabilities. First, to improve cycling success rates for individuals with Down syndrome the time of intervention might be increased to a minimum of 9 weeks. Information about IQ and language abilities might be collected in order to better tailor the cycling instruction, and with this in mind, participants should be guided to discover their own movement solutions with the support of visuals or video feedback. When modeling is used, at least two solutions should be presented to encourage the possibility of other movement options. In light of the success of participant 4 despite his reluctance to participate in intervention session, and the lack of improvement of participant 7, it may be beneficial to prescreen participants for levels of engagement prior to accepting them into the program. This is consistent with the first question in the CO-OP dynamic performance analysis decision tree: does the client want to do the occupation (Polatajko & Mandich, 2004). For those who are reluctant to participate, equipment could be made available to parents that would allow them to incorporate regular practice into their routine and work towards achieving a minimum pedaling speed of 2 m/s; consultation could be provided to these families to help guide the progression until the children are ready to and/or interested in attending a class. For You Can Ride 2, this

would help make better use of the limited volunteer resources as participants with Down syndrome receive 2 on 1 support. Finally, contrary to MacDonald's (2012) assertion that 30 meters was a large enough distance to demonstrate mastery, it is suggested that a distance of 100 meters may be a more accurate estimation and only when the environment that one needs to cycle in is considered carefully. In addition, to be able to accurately compare participants or outcomes of different methods, it may also be necessary to consider a rating of cycling performance such the performance quality rating scale (PQRS) similar to that suggested by McEwen et al. (2009) or the skill check checklist as outlined in Chapter 2. More communication with parents about their cycling goals will help bridge the transition from cycling as an activity to cycling participation.

Conclusion

Despite language and cognitive delays, as well as the cycling goal not being self-selected, the youth with Down syndrome and other intellectual disabilities in this study could be guided to discover a variety of domain specific strategies that facilitated their learning of cycling skills. Success with the CO-OP intervention was higher when the participant was interested and willing to engage in the activity. Teaching strategies such as modeling, chaining and fading were used and appear to be more necessary with this population than has previously been mentioned in other CO-OP studies. While CO-OP shows promise as an effective method to teach the activity of cycling to individuals with intellectual disabilities, multiple contextual factors may contribute to successful cycling participation. Positive predictors to continuation of cycling in the community appear to be parental support, and matching the child's cycling skills to the riding environment.

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Appendices

ALBERTA

RESEARCH ETHICS OFFICE

308 Campus Tower Edmonton, AB, Canada TGG 1K8 Tel: 780.492.0459 Fax: 780.492.9429 www.reo.ualberta.ca

Notification of Approval

Date: April 8, 2013

Study ID: Pro00036251

Principal Investigator: Janine Halayko

Study Supervisor: Joyce Magill-Evans

Study Title: Teaching two-wheeled cycling to children with a mild cognitive disability

Approval April 7, 2014 Expiry Date:

Approved Approval Date Consent 4/8/2013 Form: 4/8/2013 4/8/2013 Approved Document Adult consent information Consent signature Pre-study consent

Thank you for submitting the above study to the Research Ethics Board 2. Your application and the associated documentation has been reviewed and approved on behalf of the committee.

A renewal report must be submitted next year prior to the expiry of this approval if your study still requires ethics approval. If you do not renew on or before the renewal expiry date, you will have to re-submit an ethics application.

Approval by the Research Ethics Board does not encompass authorization to access the staff, students, facilities or resources of local institutions for the purposes of the research.

Sincerely,

Stanley Varnhagen, PhD Chair, Research Ethics Board 2

Note: This correspondence includes an electronic signature (validation and approval via an online system).

Appendix D Pre-Study Parent Questionnaire

Identifier number 1. Which strategies have you already tried (e.g., running beside the bike, taking off pedals, other biking groups, gyrowheel)? 2. What do you feel are the biggest issues that are making bike riding difficult for your child (e.g., fear of falling, tires easily, distractibility, learning disability, language delay, etc.)? 3. How would you rate your child's CURRENT PERFORMANCE riding a two-wheeled bicycle (i.e., without training wheels)? 7 1 2 3 4 5 6 8 9 10 unable some skills proficient 3. How would you rate your SATISFACTION with your child's bike riding ability on 2 wheels? 4 1 2 3 5 6 7 8 9 10 unsatisfied moderately satisfied extremely satisfied 4. List your child's strengths in regards to physical activity in general and learning to ride: 5. How does your child best learn (reading, visual/ picture cues, verbal directions, other): 6. How long has your child been working on riding a two wheeler? 7. On a scale of 1-10, how important is it to you that they learn to ride a bike? 8. On a scale of 1-10, how important do you think biking is to your child? 9. Does your child have a medical diagnosis (e.g., asthma, Developmental Coordination Disorder, Cerebral Palsy, diabetes, ADD, anxiety etc.)? 10. Do you know of any reasons why your child should not undergo physical activity? Y / N If yes, please explain_____

Appendix E Pre-Study Youth Questionnaire

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Identifier number

SATISFACTION WITH PERFORMANCE

Do you like riding with training wheels?

Do you like riding without training wheels?

PERFORMANCE

Look at the pictures. These kids are riding without training wheels.

Circle the one that is most like **your** riding.

Learning	Almost	I can!	_
Riding without training wheels is really hard.	I can almost ride by myself.	I can do it!	

IMPORTANCE

1) Do you want to learn to ride without training wheels? If yes, how much?



3) Think about riding without training wheels. How important is it to you?

I want to learn a little bit. I want to learn.	I really want to learn!
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Appendix F Post-Intervention Parent Questionnaire

Identifier number

1. Which strategies did you feel worked best when teaching to ride?

2. Hov	v would	d you ra	te your	child's C	URREN	IT PERI	FORMA	NCE ric	ling a tw	o-wheeled bicycle
(i.e., w	vithout	training	wheels)?						
	1	2	3	4	5	6	7	8	9	10
	unable some skills								proficient	
3. Hov	v would	d you ra	te your	SATISF	ACTION	V with y	our child	l's bike	riding al	oility on 2 wheels?
	1	2	3	4	5	6	7	8	9	10
unsatisfied					mode	erately s	extremely satisfied			
4. How much did your child practice on two-wheeler during this process?										
5. Doe	es your	child in	itiate/ sı	ıggest bi	ke riding	g as an a	ctivity?			
If no,	why do	you thi	nk that i	s?						

What are the challenges (if any) for your family to go on bike rides? (e.g., parents do not have bike, gravel etc)

If yes (or if bike riding is done anyway):

- 2. How often does your child ride?
- 3. Who does your child typically ride with?
- 4. Is your child able to do everything they want to/ need to do in order to be able participate in bike rides with friends or family?

Is there anything else you'd like to share about your experience or cycling in general?
	Strongly		
	Strongly Disagree/ Dislike Agree/		
			Agree/
1 The bike riding study met my	ЕПЈОУ	1 2 3 4	L 5
expectations:			•••••
2. The researchers helped me to			
understand how to better help my child		13	l5
with bike riding:			
3. I used the information/homework	NO YES		
discussed at the end of each class	If NO aldia to # 4	if VEO, along a proving the follow	ilia ai
	If NO SKIP to # 4,	If YES , please answer the follow	ling
This information:	questions		
a. Had useful activity suggestions to	Does not apply	1 2 3 4	5
help work with my child	Does not apply	······	
b. Was easy to understand	Does not apply	14	5
C. Was useful in my child's learning	Does not apply	14	5
4. My child improved in his/ her			
ability to			
a. Balance on the bike	Does not apply	14	5
b. Steer the bike	Does not apply	14	5
c. Stop the bike	Does not apply	14	5
d. Be safe on the bike	Does not apply	14	5
5. The researchers were:			
a. Respectful in their approach	Does not apply	12	5
b. Open to answering questions	Does not apply	1	5
	Does not apply	14	l5
6. Your child enjoyed coming to the			
study:	Does not apply	1	45
7. Please comment on the following:			
a. Length of study			-
b. Frequency of sessions	Does not apply	14	5 F
c. Information provided	Does not apply	14	5 5
d. Biking adjustments	Does not apply	1 2 3 <i>A</i>	5 5
e. Sequence of skills	Does not apply	1 2 3 <i>A</i>	5 5
		4	
How would you rate your child's			
current PERFORMANCE when riding	1 2 3 4	5 6 7 8 9 10	
How would you rate your	unaute	some skins proncient	
SATISFACTION with your child's	1 2 3 4	5 6 7 8 9 10	. C 1
riding abilities	unsatisfied	moderately satisfied extremely sa	tisfied
nuing autilities			

Appendix G Post Follow-up Parent Questionnaire

Appendix H One Month Follow-up Parent Questionnaire

1. Has your child been out on her bike since I saw him/her last? If so, how many times?

2. If going for a ride is difficult, what are the barriers?

3. What do you consider the most functional form of cycling for your child and/or what do you anticipate him/her using most in the future?

a) two-wheeled cycling

b) tandem cycling

c) trike or adapted bike

d) recumbent bike

e) none of the above

4. Is there any additional information you would like on two wheeled cycling (or any other cycling options mentioned above)?

5. Have you or your child used the CO-OP strategies (e.g., Goal Plan Do Check) in any other situations?