CURRENT EVIDENCE AND FUTURE DIRECTIONS FOR PHYSIOTHERAPY INTERVENTION IN CEREBRAL PALSY: PHYSICAL ACTIVITY AND PHYSICAL FITNESS

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I, **JANE GIRLING-BUTLER**, hereby declare that the work contained within this thesis is my own and has not been submitted to any other university or institution as a part or a whole requirement for any higher degree.

In addition, ethical approval from the University of Sydney Human Ethics Committee was granted for the studies presented in this thesis. Participants were required to read a participant information document and informed consent was gained prior to data collection.

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ABSTRACT

The aim of this thesis was to examine the evidence for physiotherapy intervention for children with cerebral palsy. Cerebral palsy is the most common activity limiting disability in children and adolescents with the incidence being approximately 2-3 per 1,000 live births worldwide. The severity of cerebral palsy is highly variable, but inevitably results in some degree of activity limitation due to motor impairment. However, from the physiotherapist's point of view the presence of motor impairments – and the subsequent effect that these impairments have on a child's activity and participation – is the most relevant for intervention. Therefore, determining the most effective form of physiotherapy intervention to address motor impairment, to promote physical activity and enable community participation is an important issue.

Four studies were undertaken in this thesis. The first study was a summary of systematic reviews of physiotherapy intervention for children with cerebral palsy and was conducted in order to identify the evidence located on electronic databases. This study found evidence for six physiotherapy interventions – casting, physiotherapy intervention in conjunction with botulinum toxin type-A, electrical stimulation, constraint-induced movement therapy, strength training and cardiorespiratory training. However, the evidence for the effect of intervention was equivocal due to the small numbers of randomised controlled trials, poor reporting of data in the trials and inconclusive results. Since there were a number of trials examining cardiorespiratory training in children with cerebral palsy, which is also an important focus in other areas of physiotherapy intervention, it was decided to investigate this further.

In the second study, a systematic review was conducted to examine the evidence for improving cardiorespiratory fitness in children with cerebral palsy. This review showed that it was possible to increase cardiorespiratory fitness in these children. However, the improved fitness did not carry over into their everyday activity. This raised the question of whether there was a relationship between cardiorespiratory fitness and activity and led to the third study.

The third study was a feasibility study which examined whether cardiorespiratory fitness or walking capacity influenced the amount of physical activity undertaken by children with cerebral palsy who had mild impairment and were able to walk. This study found that children with cerebral palsy had low cardiorespiratory fitness levels compared to reported values in typically-developing children. However, as there was a moderate correlation between peak heart rate and oxygen uptake during the progressive exercise test, these children demonstrated that they had a normal cardiorespiratory response to exercise of increasing intensity. To control for environmental influences, the children with cerebral palsy were compared to a matched group of siblings to examine the amount of their physical activity. The results revealed that the children with cerebral palsy were performing as much daily physical activity as their age-matched siblings, but that this activity was less than recommended guidelines for both groups. Overall, no relation was found between the cardiorespiratory fitness or walking capacity of the children with cerebral palsy, and the amount of their physical activity.

Given that the amount of activity was not determined by the child's physical characteristics (cardiorespiratory fitness or walking capacity), the fourth study was undertaken in order to identify whether there were barriers which were preventing the child performing activities. Parents were asked to indicate what they saw as being the barriers which were either preventing their child from performing an activity, or which were influencing how frequently their child performed the activity. This study found that these children were regularly engaged in physical activities which are fairly representative of typically-developing children. However, parents perceived cost as being the largest barrier to their child's activities. Of concern, was that cost was preventing the child from being engaged in activities which could be performed frequently.

Generally, children with cerebral palsy just want to engage in activities alongside their typically-developing peers. For children who are ambulant and capable of physical activity, it is important that they are given the opportunity to engage in regular activity which is performed at a moderate to vigorous level. Furthermore, identifying the barriers which are preventing physical activity means that there is a greater chance that children with cerebral palsy will have a more active lifestyle.

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This is what I have been doing, and yes, I have finished.

PREFACE

The overall theme of the thesis is to present current evidence and propose future directions for physiotherapy intervention for children with cerebral palsy in relation to physical activity and physical fitness. This thesis is arranged into 8 chapters which comprise four studies, background information and conclusions.

Chapter 1 (Introduction) provides an overview of cerebral palsy in terms of definition, aetiology and classification of the condition. In addition, this chapter outlines the evolution of physiotherapy intervention over the last century. The rise of evidenced-based medicine has challenged physiotherapists to use scientific investigation and evidence to form the basis of their practice. This raised the question of what evidence currently existed for paediatric physiotherapy intervention. In particular, what evidence was there for intervention for children with cerebral palsy? In order to address this question, the first study was undertaken to identify and summarise the best evidence available.

Chapter 2 (Study 1) is a summary of systematic review which investigated the best-evidence available for physiotherapy intervention for children with cerebral palsy. The aim of this study was to examine and summarise the best evidence available from current systematic reviews by only examining the results from systematic reviews of randomised controlled trials. The review identified 8 systematic reviews examining the effects of 6 physiotherapy interventions for children with cerebral palsy. While the reviews were deemed to be of high quality, the low number of randomised controlled trials within the reviews means that these results have limited credibility for application to clinical practice. In addition, the evidence identified

from this review is not convincing as there are contradictory results at both the impairment and activity levels and no evidence at the level of participation. However, more than 80 randomised controlled trials were located on the Physiotherapy Evidence Database (PEDro) which examined physiotherapy intervention for children with cerebral palsy. In particular, it was noted that there was emerging evidence for cardiorespiratory fitness training for these children which seemed a relevant intervention to investigate further.

Chapter 3 forms a background chapter to the second study and explores what is known about cardiorespiratory fitness in typically-developing children and in particular to compare this to children with cerebral palsy. This information was considered necessary before investigating cardiorespiratory fitness further. It has been noted that children with cerebral palsy tend to have poor cardiorespiratory fitness, lower physical work capacity and higher oxygen cost during exercise than their typically-developing peers.

Chapter 4 (Study 2) is a systematic review which examined the evidence for cardiorespiratory fitness training in children with cerebral palsy. This review showed that it was possible to increase cardiorespiratory fitness in these children. However, the improved fitness did not carry over into their everyday activity. This raised the question of whether there was a relationship between cardiorespiratory fitness and activity in children with cerebral palsy.

Chapter 5 is presented in a similar format to the background chapter in Chapter 3 and explores physical activity in typically-developing children and how this activity

compares to children with cerebral palsy. There is increasing evidence that on-going physical inactivity in these children may be contributing to their lower levels of cardiorespiratory fitness compared to their typically-developing peers. However, what is still largely unknown is whether these decreased levels of cardiorespiratory fitness are related to the amount and intensity of free-living physical activity. Therefore, the third study presented in Chapter 6 raises the question of the relationship between cardiorespiratory fitness and the amount of physical activity in children with cerebral palsy who are independently walking.

Chapter 6 (Study 3) is a feasibility study which examined the feasibility of investigating whether cardiorespiratory fitness or walking capacity influenced the amount of physical activity undertaken by children with cerebral palsy who had mild impairment and were able to walk. This study found that children with cerebral palsy had low cardiorespiratory fitness levels compared to reported values in typically-developing children. However, as there was a moderate correlation between peak heart rate and oxygen uptake during the progressive exercise test, these children demonstrated that they had a normal cardiorespiratory response to exercise of increasing intensity. Given that the amount of activity was not determined by the child's physical characteristics (cardiorespiratory fitness or walking capacity), the fourth study identified whether there were barriers which were preventing the child performing activities.

Chapter 7 (Study 4) is a descriptive study which examined the preferred choice of physical activity in children with cerebral palsy. This study found that these children were regularly engaged in physical activities which are fairly representative of typically-developing children. However, parents perceived many barriers which

were either preventing their child from being engaged in an activity, or from performing the activity more frequently. The frequency and magnitude of the perceived barriers were not examined in-depth in this study. However, these are both issues which could be investigated further.

Chapter 8 forms the discussion for this thesis and updates the current evidence for physiotherapy intervention in children with cerebral palsy. Overall, this evidence is limited and there is a need for improved quality in research. In particular, this evidence is limited for children who are ambulant and capable of physical activity. Therefore, the focus of physiotherapy intervention for children with cerebral palsy should be on increasing their opportunity to engage in regular physical activity alongside their typically-developing peers.

CHAPTER 1

INTRODUCTION

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CHAPTER 1

INTRODUCTION

RATIONALE OF THE PROJECT

Cerebral palsy is the most common activity limiting disability in children and adolescents with approximately 2-3 per 1,000 live births worldwide (Blair & Stanley 2001; Winter et al 2002; Rosenbaum 2007). The incidence rates for cerebral palsy have remained the same since first being recorded in the 1960's despite advances in the obstetric care of mothers and infants and improved knowledge of some causative factors. The unchanging incidence rates are largely attributed to the increased survival rate of very pre-term and low birth weight infants through enhanced post-natal medical care (Blair & Watson 2006). That is, as a result of improved medical care, pre-term infants are more likely to survive than previously but are more likely to have associated co-morbidities such as cerebral palsy.

The severity of cerebral palsy is highly variable but inevitably results in some degree of activity limitation due to motor impairment. The motor impairments associated with cerebral palsy can be classified under the framework of the International Classification of Functioning, Disability and Health (ICF) (World Health Organisation 2001). This framework was developed as a means to describe and measure health and disability, taking into account the physical and social aspects of disability, at both the individual and population levels (World Health Organisation 2001). The framework is divided into domains of body (comprising body functions and structure), activities and participation (Figure 1.1).

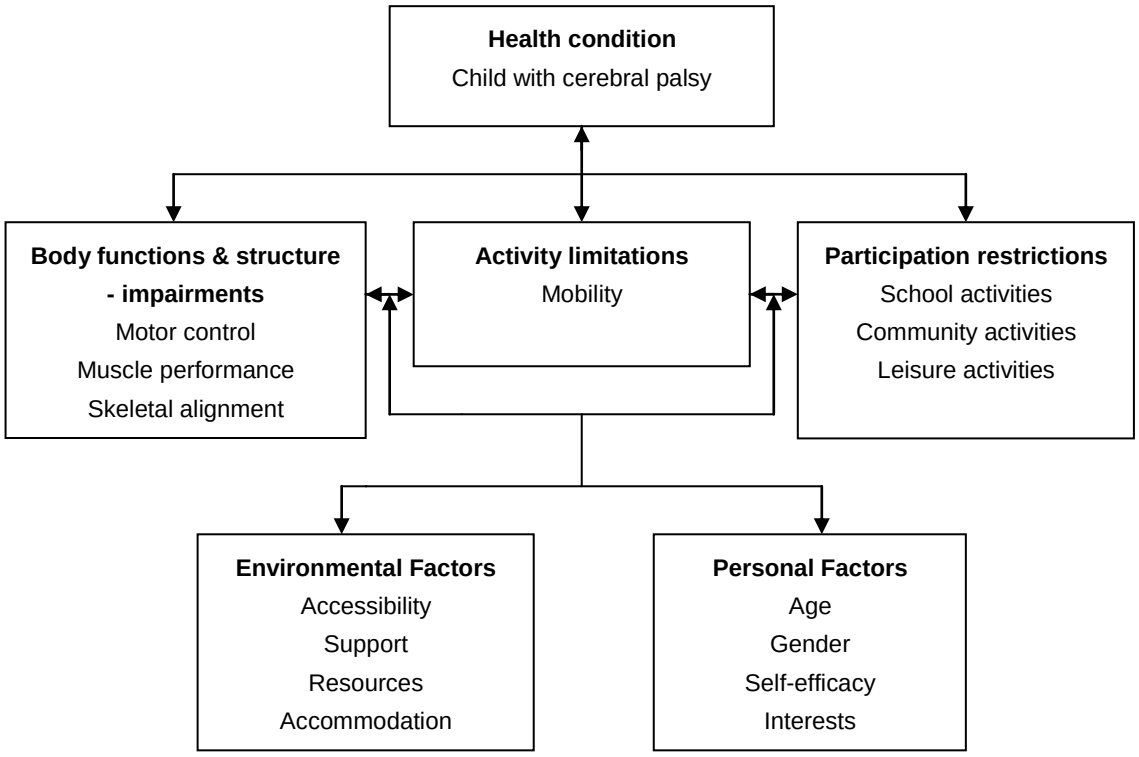

Figure 1.1: *The interaction of the domains of the International Classification of Functioning, Disability, and Health (ICF) (World Health Organization, 2001) in relation to children with cerebral palsy.*

This framework allows physiotherapists to consider the interaction between the child's impairments, their everyday activities and subsequent participation within their community. In this framework, impairments are classified under *body functions and structures* and describe problems such as impaired or decreased sensation, muscle weakness and contracture. The *activity limitations* domain describes a child's ability to move and change position in everyday circumstances such as reaching and manipulation, walking, and self-care. Within this domain, children with cerebral palsy may experience difficulties performing age-appropriate tasks or actions (Campbell et al 2006; 2012). For example, they may be unable to ride a bike or climb on outdoor playground equipment. The domain of *participation restrictions* identifies how an activity limitation would impact on the child's ability to participate in normally expected life activities (Campbell et al 2006; 2012). Such activities may

include being able to take public transport to go a movie or shopping centre with friends. The domain of *environmental and personal factors* consider whether such things as a child's age and gender, in addition to accessibility options, support and availability of resources, act as a facilitator or barrier to community participation.

Overall, cerebral palsy is a complex condition which spans physical, cognitive and behavioural domains. However, from the physiotherapist's point of view the presence of motor impairments, and the subsequent effect that these physical impairments have on a child's activity and participation, are the most relevant for intervention. The challenge for physiotherapists is to assist these children to develop effective motor performance in order that they can be independent in essential activities of everyday life (Campbell et al 2006; 2012). Children also present a unique challenge in terms of the need to consider the effects of growth and development. In particular, physiotherapists have long recognised the secondary physical impairments of cerebral palsy which become more obvious during periods of growth and the subsequent impact that these changes have on the child's everyday activities (Campbell 1991).

What then is the most effective form of physiotherapy intervention for children with cerebral palsy which will enhance their level of activity and their community participation? In physiotherapy, as in all areas of health care, there is an emphasis on providing services which are based on the best evidence available. Determining what evidence currently exists for physiotherapy intervention for children with cerebral palsy, how strong the evidence is and whether there are any gaps in the available evidence is the focus of this thesis.

CEREBRAL PALSY IN CHILDREN

Definition

Cerebral palsy can be broadly defined as a permanent, non-progressive neurological disorder caused by a lesion, or injury, to any part of the developing foetal or infant brain (Rosenbaum et al 2007). It is thought that most lesions occur in-utero, however some post-natal events may also be defined as cerebral palsy. For example, cerebral palsy is often suspected as a potential outcome where there are pre and peri natal complications of pregnancy. Likewise, where a baby is born extremely prematurely there is more likelihood that a lesion to the brain may have occurred. It is also known that similar brain lesions may produce different results when they have occurred at different chronological ages in the foetus (Morris 2007). For example, a brain lesion in a very preterm infant is more likely to lead to features associated with diplegic cerebral palsy, whereas the same lesion in an infant born at full term is more likely to lead to features associated with hemiplegia, quadriplegia or athetoid cerebral palsy. Cerebral palsy may also be the preliminary diagnosis when there is the presence of abnormal neurological findings in the neonatal period, such as inconsistent Apgar scores (Apgar 1953) or delayed developmental milestones.

The definition of cerebral palsy has varied over time, probably as a result of improved understanding of this condition. However, this variation has also led to a lack of consensus in reaching a common definition. As a result, an international workshop of recognised cerebral palsy specialists met in 2007 in an attempt to develop a more inclusive definition:

> Cerebral palsy describes a group of disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing foetal or infant brain. The motor disorders of CP are often accompanied by

> disturbances of sensation, cognition, communication, perception, and/or behaviour, and/or by a seizure disorder (Rosenbaum et al 2007, p.8).

This current definition highlights the importance of motor impairment in cerebral palsy while also recognising the influence of other associated impairments. Although cerebral palsy is acknowledged as being a non-progressive disorder, the impact specifically of musculoskeletal growth on primary physical impairments such as spasticity and weakness, is ongoing throughout childhood and into adulthood (Rosenbaum et al 2007). Often therefore the influence of secondary physical impairments such as muscle stiffness, shortening and contracture, impact the child's cognitive, behavioural and communication systems by preventing them from participating in life situations (Campbell et al 2006; 2012).

Aetiology

The cause(s) of cerebral palsy are not completely understood, however some pre-natal, peri-natal and post-natal factors have been identified (Torfs et al 1999). For example, birth asphyxia was commonly thought to be a peri-natal causative factor but is now considered to comprise only a small proportion of diagnosed cerebral palsy. Likewise, intra-cranial haemorrhage is a frequent concern in pre-term babies and, despite improved infant care, is known to be a causative factor that is more likely to lead to diplegic cerebral palsy. In addition, low to very low birth weight in infants is also known to increase the risk of cerebral palsy (Atkinson & Stanley 1983). More recently, brain imaging is being used to detect structural impairments of the developing brain, and may also give some indication of the timing and location of the lesion (Krageloh-Mann 2004). Imaging may also be useful in determining the type of motor impairment which may become apparent after the child is born. In

addition, there may be instances of cerebral palsy in which the cause is unknown (Blair & Watson 2006).

Classification

Cerebral palsy is commonly classified according to type, distribution and severity of motor impairment. The 'type' of cerebral palsy typically refers to the movement anomalies exhibited, eg, spasticity, rigidity, athetosis and chorea, while distribution refers to the observed 'paralysis', eg, hemiplegia, diplegia and quadriplegia (Mutch et al 1992). The degree of severity of cerebral palsy varies greatly and by convention has generally been described as being either, mild, moderate or severe. However these descriptions of severity are quite broad and subject to interpretation and therefore have often been used inconsistently.

Impairment

The primary motor impairments of cerebral palsy are the result of an upper motor neurone lesion leading to weakness, spasticity, contracture, incoordination, altered sensation and balance, and associated reactions (Rosenbaum et al 2007). Although cerebral palsy itself is non-progressive, progressive motor impairment is a feature which often becomes more noticeable during periods of growth and development (Graham & Selber 2003). As a child with cerebral palsy ages, they tend to develop increasingly abnormal movement along with accompanying increases in spasticity and contracture. These changes lead in turn to a general poverty of movement as a result of muscular atrophy, disuse and stretch weakness (Shepherd 1995). Long term musculoskeletal changes can be seen into adulthood with restricted muscle length causing bony abnormality and joint instability resulting in subluxation and

dislocation. In addition, cartilage damage, which occurs through altered compression and immobility, results in osteoporotic changes and painful arthritis (Campbell et al 2006; 2012). Therefore, the effect that these impairments have on developing skeletal muscles, and subsequently the motor performance of children with cerebral palsy, is of most concern to physiotherapists.

Activity limitation

The presence of impairments in children with cerebral palsy is likely to have a carryover effect into their everyday physical activities such as walking, climbing on play equipment and moving around at school and the community. Correspondingly, these children are more likely to engage in sedentary behaviours and therefore be less active than their typically developing peers (Capio et al 2010, Bjornson et al 2008, Murphy et al 2008, Maher et al 2007, Damanio 2006).

The Gross Motor Classification System (GMFCS) Palisano et al (1997) was developed to classify the amount of activity limitation seen in children with cerebral palsy and describes their activity in relation to walking (Figure 1.2). This system comprises 5 levels to best represent the child's current level of physical activity and mobility in their home, school and community settings (Campbell et al 2006; 2012). The GMFCS has been widely adopted for use in both research and clinical settings (Morris and Bartlett 2004).

Level	Abilities and limitations
I	Walks without limitations at home, school, outdoors, and in the community.
II	Walks without assistive devices but has limitations walking outdoors and in the community.
III	Walks using a hand-held mobility device (cane, crutches, walker). Limitations walking outdoors and in the community - child transported in a wheelchair or uses powered mobility.
IV	Child uses wheeled mobility in most settings; requires adaptive seating and physical assistance for transfers. Self-mobility can be achieved using powered mobility.
V	Self-mobility is severely limited even with the use of assistive technology. Child may achieve self-mobility using powered mobility with extensive adaptations for seating and control access.

Figure 1.2: *The Gross Motor Function Classification System (GMFCS) (Palisano et al 1997 and CanChild Centre for Childhood Disability Research http://www.canchild.ca/Portals/0/outcomes/pdf/GMFCS.pdf accessed, June 24, 2010).*

A similar system which complements the GMFCS, but has a focus on upper limb activities, was developed by Eliasson et al (2006). The Manual Ability Classification System (MACS) classifies the amount of upper limb activity in children with cerebral palsy (Eliasson et al 2006) (Figure 1.3). The five levels in the MACS are based on the child's self-initiated ability to manipulate objects and their need for assistance or adaptation to perform manual activities in everyday life.

Level	Abilities and limitations
I	Handles objects easily and successfully. At most, instead of handling objects with both hands, may have limitations in performing manual tasks requiring speed and accuracy. However, any limitations in manual abilities do not restrict independence in daily activities.
II	Handles most objects but with somewhat reduced quality and/or speed of achievement. Certain activities may be avoided or be achieved with some difficulty; alternative ways of performance might be used but manual abilities do not usually restrict independence in daily activities.
III	Handles objects with difficulty; needs help to prepare and/or modify activities. The performance is slow and achieved with limited success regarding quality and quantity. Activities are performed independently if they have been set up or adapted.
IV	Handles a limited selection of easily managed objects in adapted situations. Performs parts of activities with effort and with limited success. Requires continuous support and assistance and/or adapted equipment for even partial achievement of the activity.
V	Does not handle objects and has severely limited ability to perform even simple actions. Requires total assistance.

Figure 1.3 *The Manual Ability Classification System (MACS)*
Eliasson et al (2006).

Participation restriction

The World Health Organisation (WHO) (WHO, 2001) defines participation restriction as problems an individual may experience when attempting to be involved in a life situation. For children with cerebral palsy, this may mean difficulties when walking to a classroom, interacting with their peers during lunch and play breaks or participating fully in school physical education programs. Participation is very much an individual choice of activity and can be categorised by aspects of performance and capacity. For example, what a child can do (their performance), ie, such as walking measured using a 10-m Walk Test or a 6 Minute Walk Test, versus what they actually do (their capacity) ie the amount of physical activity carried out each day. The difference between performance and capacity is most likely to be dependent on

personal and environmental factors such as choice or availability of resources (WHO 2001). Subsequently, participation restriction is difficult to measure and is most commonly assessed by determining either the child's, or parents', perception of their quality of life. For a child, their perception of quality of life may be determined by their ability to interact and be involved in activities with their typically-developing peers. For parents of a child with cerebral palsy, the stress and everyday demands of caring for a child with a disability may determine their perceptions. It is recognised that there are many potential causes of poor quality of life factors which can impact on the child, their parents or caregivers, and subsequent family functioning. It is also likely that these factors are greater as the severity of cerebral palsy increases (Vargus-Adams 2005).

EVOLUTION OF PHYSIOTHERAPY INTERVENTION OVER THE LAST CENTURY

Orthopaedic approach

Some of the earliest documented evidence of physiotherapy intervention for children who have cerebral palsy comes through the work of Elizabeth Kenny (Kenny 1937, Kenny 1941). Initially her influence was with children who had been affected by polio whereby she applied heat packs, massage, splints and passive exercises to children's paralysed limbs. This intervention was considered radical at the time as most treatment for polio involved rest and the use of iron lungs for respiratory failure. With the discovery of the Salk vaccine in the mid 1950's, polio was virtually eradicated in developed countries and subsequently the focus of physiotherapy intervention changed. This focus became directed towards children who were

affected by other musculoskeletal anomalies as well as cardiorespiratory and neurological conditions. In the area of neurological conditions, such as cerebral palsy and spina bifida, Kenny and others largely applied some of the same orthopaedic principles which they had employed for children affected by polio. That is, stretching and splinting (via metal callipers and surgical boots) as well as positioning, passive movements and strengthening.

However, during this transition in physiotherapy intervention, it became apparent that intervention strategies of muscle strengthening and re-education, which were viewed as effective in treating disorders of the peripheral nervous system, were not able to address the problems seen in disorders primarily involving the central nervous system (CNS) (Gordon 1987).

Neurotherapeutic approach

The subsequent emergence of 'neurotherapeutic' approaches to address the problems seen in children with neurological conditions, such as cerebral palsy, saw a major shift in physiotherapy intervention (Gordon 1987). This shift probably came about as prominent physiotherapists of the time were reading more widely and in particular focussing on knowledge emerging from the behavioural and cognitive theory literature.

In the behavioural theory literature, the writings of authors such as Gesell (1928, 1934, 1940), Shirley (1931) and McGraw (1943) were based on their observations of child development and how the progression of motor development was thought to be dependent on the maturation of the child's central nervous system (CNS). As the

child matured, predetermined patterns of behaviour emerged and were not seen to be dependent on the surrounding environment of the child. In addition, more complicated patterns of movement became evident as higher levels of the child's CNS developed. Later authors termed these views 'neuromaturational' theories (Gordon et al 1987). Other specific motor behaviours, which only seemed to be apparent in newborn infants, were also observed and noted to be influenced by external stimuli such as positioning or touching. For example, putting pressure on an infant's palm caused flexion of the fingers, standing the baby and tipping them forward slightly led to spontaneous stepping movements. These behaviours were therefore thought to appear at different points during the first year of life and manifest in a stimulus response mode. Subsequently the terms *infantile or primitive reflexes* were coined (Gesell 1928, Gesell 1933, Gesell 1940, Gesell 1948, Shirley 1931, McGraw 1943). Furthermore, it was believed that these responses needed to 'disappear' before the child would be capable of volitional movement (Touwen 1984).

This combination of the appearance and disappearance of reflex activity and a progression of motor skills led to the term 'milestones' as a way of describing the point at which a child was likely to achieve a motor skill. That is, a predictable chronological age when a child's motor behaviours appeared and subsequently developed. It was also believed that these milestones occurred in a predictable order and that the appearance of one milestone was a prerequisite for the next (Gesell 1934, Shirley 1931, McGraw 1943). For example, a child needed to able to roll before they could sit and crawl before they would be able to walk independently. Subsequently, defining and outlining the emergence of motor development in terms

of ‘stages’ became common in developmental literature. Indeed, the writings of Gessell still continue to influence some developmental psychology textbooks.

During the 1970’s, behaviourists such as Watson (1971) and Connolly (1970; 1973; 1977) proposed a learning-based theory which described infants and children engaging in ‘trial and error’ motor behaviour as a way of learning solutions to motor problems and the subsequent development of their motor skills. Likewise, Skinner (1975) proposed that a child’s motor development was influenced by their interaction with their environment. That is, the environment motivated the child to move and subsequently shaped the type of motor behaviour seen. Furthermore, the child was seen to be interacting and continually adjusting to their environment and not just a passive recipient (Skinner 1975).

In the cognitive theory literature, authors such as Piaget (1952) and Erikson (1975) sought to explain the emergence and development of motor skills in relation to the child’s developing cognitive abilities. Piaget (1952) believed that a child needed to have a balance between their environmental experience and their existing cognitive structures in order to learn a new motor skill. That is, if they had not reached a certain level of maturity in their cognition they would not be able to learn or perfect a new skill no matter how much they might practice. This view was also used to explain why children with cognitive impairment had correspondingly delayed motor skills (Erikson 1975).

Despite numerous advances in knowledge relating to neural plasticity and biomechanics it would seem there is still a widespread belief in predictable

developmental ‘norms’. Universal descriptions of motor behaviour have become ingrained in child development textbooks and have subsequently formed the basis of many current developmental tests and physiotherapy intervention programs (Thelen 1995). Many of the approaches to physiotherapy intervention have been implemented worldwide and have been revised, modified or combined by physiotherapists over the last few decades. As a result, these modified approaches may have the same name as the original, but they may be quite different in practice. Commonly known approaches include neurodevelopmental therapy (NDT) (Bobath 1957, Bobath 1963, Bobath 1969); a neurophysiological approach (Rood 1956); proprioceptive neuromuscular facilitation (PNF) (Kabat and Knott 1954; Knott and Voss 1968); neuromuscular reflex therapy (Doman et al 1960); sensory integration (SI) (Ayres 1972); conductive education as developed by Andras Peto (Hari and Tillemans, 1984); and the Vojta method (Vojta 1984). A summary of these approaches is presented in Figure 1.4.

APPROACH	UNDERLYING ASSUMPTIONS
Bobath (1940's-1990's)	• Children with cerebral palsy have disordered postural and movement patterns ie *postural reflex activity*. • Theory of neurodevelopmental treatment (NDT) • When nervous system damaged, lower level reflexive behaviours released from inhibitory controlling system of the CNS leading to abnormal movement responses. • Intervention involves 'inhibiting' abnormal responses/tone/reflexes by positioning; facilitating normal postural responses and using key points of control; assumption that one task will generalise into another.
Rood (1950's-80's)	• Neurophysiological approach – facilitation of 'normal' movement patterns. • Assumption of carry over and/or generalisation to other motor skills.
Kabat, Knott and Voss (1960's–80's)	• Proprioceptive neuromuscular facilitation (PNF) • Focus on CNS dysfunction – emphasis on exercising muscles in specific diagonal and spiral patterns. • Stimulation of proprioception results in recruitment of additional motor units.
Doman-Delacato (1960's)	• Neuromuscular reflex therapy – psychomotor 'patterning' • Intervention for intellectual disability and motor disorders • Neurodevelopmental stages related to evolutionary amphibian/reptilian stages.
Ayres (1960's–80's)	• Sensory integration. • Assumption that child's ability to learn is dependent on utilising sensory information from the environment. • Learning dependent on intake and processing of sensory experiences from movement and the environment
Peto (1940's-1960's)	• Hungarian system - 'Conductive motor pedagogy' more commonly known as conductive education (CE). • Motor disorder a learning difficulty; assumption that brain capable of regenerating itself following injury. • 'Rhythmic intention' – inner and external speech to express intended motor action followed by movement performed rhythmically while counting (dynamic speech) to describe the action being performed. • Intervention comprises of series of tasks (task series) – children work predominantly in groups • Conductor responsible for teaching/guiding group, formulating program of tasks
Vojta (1950's)	• Reflex locomotion – motor responses occur as a result of specific stimulation • Intervention to facilitate the automatic regulation, or control, of the body's position; stimulate coordinated muscle activity.

Figure 1.4: *A summary of approaches to physiotherapy intervention for children with neurological disorders.*

Generally the aim of physiotherapists in using these approaches has been to improve the 'functional movement' of a child affected by cerebral palsy. In most cases it was thought that if impairments such as spasticity could be inhibited and contracture prevented, then the child's ability to perform everyday movements would be enhanced. The manner in which these motor impairments have been addressed by both physiotherapy and medical intervention has varied over time. For example, commonly, physiotherapists have addressed the issues of spasticity and muscle contracture by using 'reflex inhibiting positions' (Bobath 1963), passive stretches, splints and serial casting. Medical interventions for spasticity have comprised of orthopaedic surgery, nerve blocks, selective dorsal rhizotomy, and botulinum toxin type-A injections. Until recently, weakness has not been seen as an issue for priority for physiotherapy intervention. Indeed, physiotherapists have largely been influenced by persisting concerns that the increased effort associated with strengthening interventions would not only increase spasticity, but would also lead to an increase in muscle contractures (Bobath 1957, Bobath 1990).

Although improvement in activity has always been a desired outcome of physiotherapy intervention, it is probably fair to say that activity in itself was not necessarily the main goal of the intervention. Indeed, the emphasis has been more on reducing the activity limitation rather than promoting a specific activity. By addressing a specific impairment which was seen to be limiting a motor skill the assumption was that this skill would then generalise, or carry-over, into a more functional activity. For example by practising balancing on one leg the assumption might have been that improvement in this skill would lead to a child being able to stand on one leg and kick a ball as required for a game of soccer. As a result, the

majority of motor assessments have tended to have the measurement of impairment as their primary focus and it is only more recently that assessments which focus on activity performance have emerged (Russell 1989).

Likewise an assumption may be that improving a child's performance in activities would enable them to then participate in community and societal activities. However, again the issue of participation, and in particular participation restriction, has not been a focus of physiotherapy intervention or necessarily an issue which has been followed up in these children.

Movement science era

The era of 'movement science' emerged from the late 1970's into the 1980's with a renewed interest in the development and performance of motor behaviour. The previous emphasis of intervention for individual muscles and joints was being replaced by a focus on internal neuronal processes and how these processes influenced movement (Gordon 1987). In particular, there was a need to understand how the central nervous system coped with the demands of controlling movement in a task-oriented approach (Greene 1988). This task-oriented approach recognises that motor performance is continually being modified by both the demands of the task and the environmental conditions which will either enhance or detract from the performance (Kelso 1982).

In order to explain the emergence and development of motor skills, a dynamic systems theory based on the earlier work of Bernstein (1967), Kugler and Turvey (1987) and Kelso et al (1981, 1986), has been proposed by Thelen and colleagues

(Thelen & Ulrich 1991, Thelen 1995, Thelen & Spencer 1998, Clearfield, Feng, & Thelen 2007). This theory addresses the questions of what drives skill acquisition and how does the child move from one developmental stage to the next?

Developmental change is seen as a series of states of stability and instability in the child and is dependent on certain specific constraints. These constraints may be a combination of internal mechanisms such as neural maturation and cognition, and external mechanisms such as growth and biomechanical factors (Thelen 1995). Thelen (1995) proposes that for any new motor task there will be initial high variability in the performance of the task as the child explores the components of the task. This performance becomes less varied and progressively more stable as the child learns the requirements of the skill and repeatedly practices its execution (Thelen 1995). Furthermore, motor behaviour emerges as a result of the child's interaction with their environment with the subsequent behaviour being dependent on the task and the environmental demands. For example, the desire to look around while positioned in prone, influences an infant to lift their head off the surface. Likewise the presence of a toy within arm's reach will lead to rudimentary reaching attempts by the infant. Therefore, providing an environment which enables a child to perform self-initiated movements within naturally occurring restraints is more likely to encourage the development of motor behaviour (Shepherd 1995).

The effect of environmental factors in terms of promoting motor behaviour has been reported in cross cultural studies. These studies have challenged the idea of universal motor development by demonstrating the variability in the emergence of motor behaviour (Goldberg 1972, Solomons and Solomons 1975, Super 1976,

Konner 1977). For example, children in some African countries achieve independent walking sooner than Western-reared infants due to early practice of standing. In addition, these studies have further demonstrated that environmental influences on motor development may be determined by parental expectations and subsequent child rearing behaviours. Child rearing practices may be particularly crucial for influencing motor development during the first 18 months of life (Shepherd 1995). Likewise, environmental opportunities which allow repetition and variability of practice have been shown to influence the development of motor skills and also cognition and emotion (von Hofsten 1991).

Due to the differing rates of maturation in the body's systems, motor development has been seen to be non-linear and consists of spurts, plateaus and 'regressions'. These phases produce qualitative and quantitative changes in motor performance as a result of 'rate-limiting' or constraining factors (Thelen & Ulrich 1991, Thelen 1995). For example, a child's ability to walk independently may be affected more by their body morphology than their cognitive desire to be upright.

An increased understanding of the development of postural control and motor skills in children has been gained through the research of, among others, Shumway-Cook and Woollacott (1985; 2001; 2007); Hirschfeld and Forssberg (1991); and Forssberg (1985). These researchers have found that once a child is able to sit or stand independently they display similar postural responses to adults. In contrast, Nashner et al (1983) discovered that children with cerebral palsy tended to show postural responses in a reverse order to the norm. That is, when standing on a movable platform they activated proximal muscle groups (hip) prior to distal muscle groups

(ankle). However despite these differences the postural response in these children was still sufficient to prevent them from falling.

Over the years, physiotherapists have based their intervention on the collective experience of prior generations of colleagues who have passed their knowledge on to new practitioners (Campbell et al 2012). This knowledge has largely served physiotherapists well but new knowledge in the area of movement science, with a better understanding of the central nervous system and biomechanics, has challenged many of the assumptions and rationale for intervention. In addition, worldwide changes in health-care delivery are requiring physiotherapists to use scientific investigation and evidence to form the basis of their practice. Unfortunately many of the traditional interventions have little, or low, levels of evidence for effect.

Evidenced-based practice

So where are we now in terms of physiotherapy intervention for children with cerebral palsy? It would seem that there has been a major change in thinking from the "we can derive implications for practice from basic science" view that has dominated since the 50's, to a "we should practice interventions that have been shown to be effective" view. This latter view forms the basis of the current evidence-based practice era.

The concept of evidenced-based medicine can be traced back to ancient Chinese medicine and is attributed in part to the interpretation of Confucion texts (Sackett et al 2000). In more recent times, the practice of evidenced-based medicine means

integrating individual clinical expertise with a critical appraisal of the best evidence available from external clinical evidence which has been obtained from systematic research (Sackett et al 1996).

The catalyst for the current wave of evidenced-based practice stems from a rapid surge of medical literature during the 1980's and early 1990's. However, much of this literature was perceived to be of poor quality and in some cases the positive results from high quality research were not being implemented even when there was evidence for effect (Herbert et al 2005). Consequently, a group of researchers from McMaster University in Canada formed a group, the Evidenced-Based Medicine Working Group, in an effort to improve medical teaching (Evidenced-Based Medicine Working Group 1992). The aim of this group was to use high quality evidence to develop research guidelines to improve patient care. In conjunction with The Oxford Centre for Evidence-Based Medicine (OCEBM), the Evidenced-Based Medicine Working Group have elaborated the five-level system first proposed by Sackett (1986) (Figure 1.5). This system categorises the strength of the evidence from research trials by allocating the highest score, Level I, to systematic reviews of randomised controlled trials. The lowest score, Level V, is allocated to research which is primarily based on expert opinion without critical appraisal.

Level	Type of evidence
I	Evidence from at least one systematic review (with homogeneity) of multiple randomised controlled trials
II	Evidence from systematic review of cohort studies (including at least one randomised trial)
III	Evidence from systematic review (with homogeneity) of case-control studies or individual case-control studies
IV	Evidence from well-designed case series (and poor-quality and case-control studies)
V	Expert opinion without explicit critical appraisal or based on physiology, bench research or 'first principles'

Figure 1.5: *Levels of evidence for research trials (Sackett 1986).*

Recently, the OCEBM have reformulated the levels of evidence table as a series of searching "steps". These steps are intended to help researchers go through a process of questioning and identifying the best type of research design which will answer the research question posed (Figure 1.6).

Question	Step 1 (Level 1*)	Step 2 (Level 2*)	Step 3 (Level 3*)	Step 4 (Level 4*)	Step 5 (Level 5)
How common is it? (eg, Pre-test Probabilities)	Most relevant local and current random sample survey (or censuses)	Systematic review of current surveys	Systematic review of local non-random sample	Systematic review of case-series	Opinion without explicit critical appraisal, based on limited/undocumented experience, or based on mechanisms
Is this test accurate? (Diagnostic accuracy)	Systematic review of cross sectional studies	Systematic review of cross sectional studies with consistently applied reference standard and blinding	Systematic review of non-consecutive studies, or studies without consistently applied reference standards.	Systematic review of case control study, or cross-sectional study with on-independent reference standard	Opinion without explicit critical appraisal, based on limited/undocumented experience, or based on mechanisms
What will happen if we do nothing? (Prognosis)	Systematic review of inception cohort studies	Inception cohort studies	Cohort or control arm of randomized trial	Systematic review of case-series	Opinion without explicit critical appraisal, based on limited/undocumented experience, or based on mechanisms
Does this treatment help? (Treatment Benefits)	Systematic review of randomized trials or *n*-of-1 trial	Randomized trial or (exceptionally) observational studies with dramatic effect	Non-randomized controlled cohort/follow-up study	Systematic review of case control studies, historically controlled studies	Opinion without explicit critical appraisal, based on limited/undocumented experience, or based on mechanisms
What are the COMMON harms? (Treatment Harms)	Systematic review of randomized trials or *n*-of-1 trial	Systematic review of nested case-control or dramatic effect	Non-randomized controlled cohort/follow-up study	Case-control studies, historically controlled studies	Opinion without explicit critical appraisal, based on limited/undocumented experience, or based on mechanisms
What are the RARE harms? (Treatment Harms)	Systematic review of case-control studies, or studies revealing dramatic effects	Randomized trial or (exceptionally) observational study with dramatic effect	Non-randomized controlled cohort/follow-up study	Case-control studies, historically controlled studies	Opinion without explicit critical appraisal, based on limited/undocumented experience, or based on mechanisms
Is early detection worthwhile? (Screening)	Systematic review of randomized trials	Randomized trial	Non-randomized controlled cohort/follow-up study	Case-control studies, historically controlled studies	Opinion without explicit critical appraisal, based on limited/undocumented experience, or based on mechanisms

Figure 1.6: *The Oxford 2011 Table of Evidence (OCEBM 2011).*

****** Level may be graded down on the basis of study quality, imprecision, indirectness because of inconsistency between studies, or because the absolute effect size is very small, level may be graded up if there is a large or very large effect size.*

This system has been adopted by others in the health area for appraising research literature. Likewise physiotherapists are able to use this system to evaluate research relating to physiotherapy intervention and therefore to be better informed when making clinical decisions.

The wave of evidenced-based medicine has influenced many areas of health care. In physiotherapy, some of the early drivers of this movement have originated in the Netherlands and has led to high quality clinical research being conducted (Herbert et al 2005). In addition, texts have been published which help physiotherapists to understand what evidenced-based medicine is and how this knowledge could be relevant to their clinical practice (Bury & Mead 1998; Herbert et al 2005). Likewise free electronic databases such as The Cochrane Library (http://www.cochrane.org/cochrane-reviews) and the Physiotherapy Evidence Database (PEDro) (http://www.pedro.org.au) provide access for physiotherapists to randomised trials and reviews on the effects of intervention.

The need to based physiotherapy intervention on the highest levels of evidence available has been proposed by Herbert et al (2005) and also the need to identify what constitutes evidenced-based physiotherapy. "Evidenced-based physiotherapy is physiotherapy informed by relevant, high quality clinical research" (Herbert et al 2005 p.2). Therefore, 'evidence' is high quality clinical research (Herbert et al 2005). However, where high quality evidence is not available, then knowledge derived from other sources such as lower quality research, clinical expertise or patient preferences should guide the physiotherapist in their clinical decision making (Herbert et al 2005).

How can physiotherapists use the increasing amount of evidence to support the effectiveness of their clinical interventions? Physiotherapists are encouraged to use scientific evidence and reliable decision-making methods as the basis of their clinical practice (Palisano et al 2006). However, accessing evidence from research is not always an easy task as there are a vast number of medical journals and databases which have published clinical trials and systematic reviews, and many physiotherapists would have constraints to access this information during a normal working day (Moseley et al 2000). This problem may be overcome to some extent by accessing the health specific databases of The Cochrane Library and the Physiotherapy Evidence Database (PEDro) which are relevant to physiotherapy clinical practice.

Of particular interest to physiotherapists is whether a physiotherapy intervention has a beneficial, detrimental or null effect on the domains of impairment, activity and participation. Physiotherapists are most likely to find evidence of effect from randomised controlled trials and systematic reviews. Over the last decade there have been several reviews which have combined randomised controlled trials and reported on the effect of physiotherapy intervention in cerebral palsy. A review by Antila et al (2008) examined the effect of 'usual' physiotherapy intervention compared to Conductive Education in children with cerebral palsy. However, as most of the studies in these reviews were not randomised controlled trials, the authors concluded that the effects of intervention remained unclear or unsupported by data. Unfortunately, these findings are not uncommon and are largely due to the lack of randomised controlled trials in many reviews.

Moseley et al (2002) examined the randomised controlled trials and systematic reviews located on the Physiotherapy Evidence Database (PEDro) for the effect of physiotherapy intervention. Their study found that there had been an exponential increase in the number of trials and systematic reviews conducted in the last 8 years. Interestingly, Moseley et al (2002) also noted that paediatric physiotherapy represented the smallest number of trials and reviews compared to other areas of physiotherapy intervention. This raises the question of what evidence there is in those trials and reviews located on PEDro, for paediatric physiotherapy intervention. In particular, what evidence is there for intervention for children with cerebral palsy?

Evidence for physiotherapy intervention in cerebral palsy

In March 2006 a search of the Physiotherapy Evidence Database (PEDro), similar to the Moseley et al (2002) study, was conducted to identify the systematic reviews and randomised controlled trials located on this database which pertained to both cerebral palsy and other neurological conditions in children. An advanced search was conducted by using the 'neurodevelopmental therapy, neurofacilitation' terms in the therapy field and the 'paediatrics' and 'neurology' terms into the subdiscipline field. 'Clinical trials' and systematic reviews' were both included in the methods field. No time limit was placed on the search.

The search revealed a total of 102 records of which there were 83 randomised controlled trials and 19 systematic reviews pertaining to physiotherapy intervention in children with neurological disorders. A calculation of the cumulative totals of randomised trials from this search shows a twofold increase in the publication of trials within a 6 year period (from 28 publications during the period 1975 to 1999 to

55 publications from 2000 to 2006.) However, there was an even larger increase in the publication of systematic reviews with an increase from 2 to 17 during this same time period (Figure 1.7). This overall level of increase in publications is similar to the results found by Moseley et al (2002) for other areas of physiotherapy intervention.

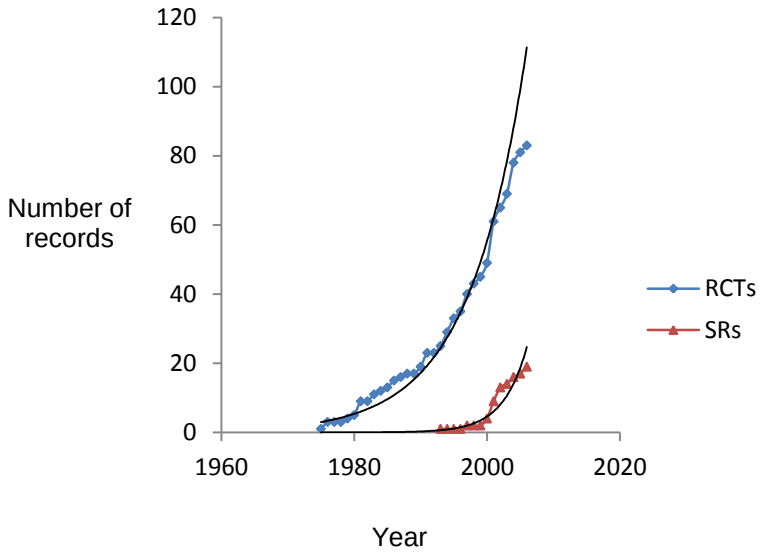

***Figure 1.7**: Calculation of cumulative totals of randomised controlled trials and systematic reviews from 1975 – 2006 showing an exponential trend.*

Of the 83 randomised controlled trials in paediatrics, just over half (n=46) related to cerebral palsy or a neurological condition. The remaining trials investigated conditions such as Duchenne Muscular Dystrophy, Down Syndrome, Developmental Coordination Disorder and cognitive deficits. Within the 46 trials 3 were not specifically a physiotherapy intervention and comprised occupational therapy, traditional Chinese medicine (acupuncture), and parental compliance to a home program. In terms of the scope of physiotherapy intervention, neurodevelopmental therapy comprised 1/3 of all randomised controlled trials. Other interventions included constraint induced movement therapy, efficacy of standing frames and

standing programs, casting, orthoses, botulinum toxin type-A in conjunction with a physiotherapy intervention, hippotherapy, hydrotherapy, biofeedback and electrical stimulation.

The methodological quality of the 43 trials ranged from 1 out of 10, to 7 out of 10 on the PEDro score. Overall, approximately 60% of the trials were of moderate to high quality, with a score of 5 or more (the mean quality score was 4.80). However, despite the increase in trials seen from 2000 to 2006, the quality of those trials did not increase and the mean for this time period stayed largely the same. Figure 1.8 shows the distribution of PEDro scores.

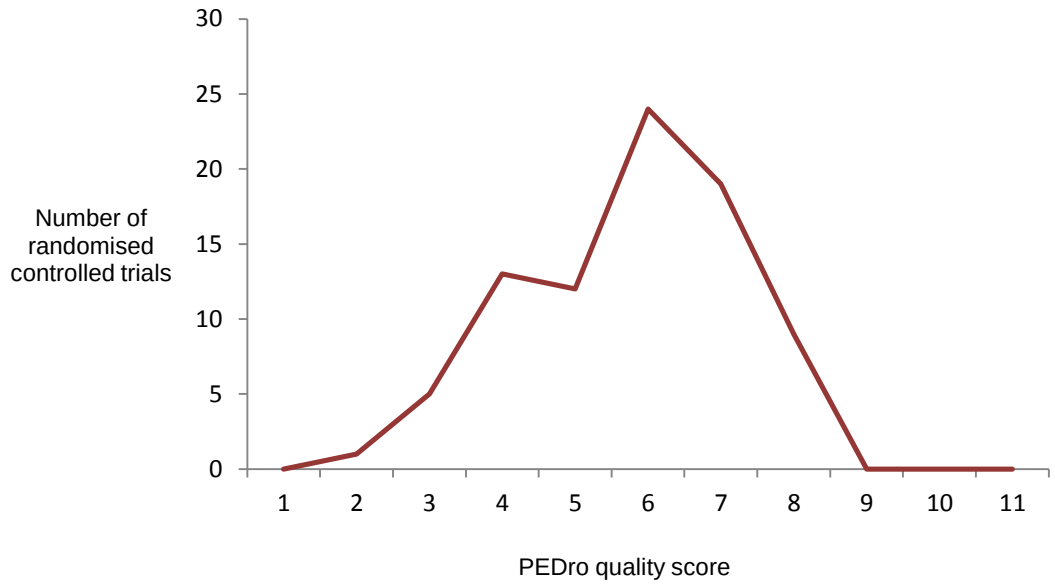

Figure 1.8: *Frequency distribution of the PEDro score for randomised controlled trials in paediatric physiotherapy.*

In order to see how the trials and systematic reviews may have changed over the 30 year time period both the first and last trial and systematic review were investigated.

The first trial relating to physiotherapy intervention in paediatrics was conducted in 1976 (Scherzer et al 1976). This trial investigated the effect of physiotherapy intervention over a 6 -12 month period on children under 18 months of age. Twenty four children were allocated into either an experimental or control group in a double-blind study. The experimental group received *'neurophysiologic physical therapy to stimulate motor milestones'* while the control group received *'traditional passive range of motion exercises'*. Each group was seen twice weekly for 6-12 months. The main outcome measure for both groups was a *'Motor Development Evaluation Form'* which was specifically constructed for the study. The authors reported 'definite changes' in the experimental group as a result of intervention however no significance was found between the two groups. This trial scored 4 on PEDro scale.

The most recent trial (Bar-Haim et al 2006) compared the efficacy of wearing an Adeli™ suit in children with cerebral palsy in comparison to neurodevelopmental treatment (NDT) over a one month period. This study also had 24 participants with half being randomly allocated into the experimental and control groups respectively. The experimental group wore the Adeli™ suit according to the original Russian protocol and Russian physical therapists, considered to be experts in this protocol, treated all children in the experimental group. The control group received an individual structured NDT program. At the conclusion of the study there were no significant differences either within or between the experimental and control groups. This trial scored 6 on the PEDro scale.

Of the 19 systematic reviews 14 related to cerebral palsy or a neurological condition. The remaining 5 reviews covered developmental coordination disorder, preterm

infants or issues dealing with unconscious children. The scope of intervention within the relevant reviews included casting, strength training, botulinum toxin type-A in conjunction with a physiotherapy intervention, neurodevelopmental therapy and electrical stimulation.

The first systematic review was published in 1993 (Turnbull 1993) and examined early intervention for children with, or at risk of, cerebral palsy. This review included 15 studies which were conducted from 1973 to 1991. The author concluded that the studies in the review failed to find convincing evidence for the efficacy of physiotherapy intervention largely due to inadequate methods of assessment. The latest review in 2006 (Autti-Ramo et al 2006) summarises systematic reviews on the effectiveness of upper and lower limb casting and orthoses in children with cerebral palsy. This review included 5 systematic reviews which contained 32 published studies. However of these 32 studies, only 5 were randomised controlled trials with the remainder described as observational studies. The methodological quality between the 5 studies varied considerably and the authors therefore concluded that evidence was only apparent for positive short-term effect of lower limb casting on passive range of movement.

The author's comments in many of the systematic reviews, that the effect of intervention was either inconclusive or that there was little evidence to suggest the efficacy of one intervention over another, appears to be a recurring theme. It would be interesting to know whether the inconclusive results could be attributed to factors relating to the specific intervention applied, issues relating to the study design or a combination of both. Many studies of therapy intervention often involve quasi-

experimental or non-experimental designs which may well have been used in the observational studies within these reviews. Therefore there may be limited credibility for application to clinical practice as these types of experimental design tend to provide lower levels of evidence (Campbell 2006).

A quick scan of other electronic databases, such as Medline, Cinahl and Ovid, was then undertaken to see whether similar topic areas to those displayed on PEDro were being reported. This was in fact the case in addition to more recent studies investigating the effect of treadmill training in young children (Schindl et al 2000, Ulrich et al 2001), physical fitness (van den Berg-Emons 1995; 1998), strengthening (Darrah et al 1997, Dodd et al 2002, Dodd et al 2003) and electrical stimulation (Kerr et al 2004).

There are a growing number of randomised controlled trials and systematic reviews emerging on the electronic databases relating to physiotherapy intervention for children with cerebral palsy. However, it is not clear which intervention is more likely to be effective. There is a need then, to examine the available evidence by combining some of the randomised controlled trials into systematic reviews in order to find the best evidence. Therefore, such a systematic review was undertaken (Study 1) and is presented in the next chapter.

CHAPTER 2

STUDY 1: SUMMARY OF BEST-EVIDENCE AVAILABLE FROM SYSTEMATIC REVIEWS OF PHYSIOTHERAPY INTERVENTION IN CHILDREN WITH CEREBRAL PALSY

INTRODUCTION

METHOD USED TO CONDUCT THE REVIEW

- **Identification and selection of reviews**
- **Assessment of characteristics of reviews**
- **Analysis of reviews**

RESULTS

- **Flow of reviews through the study**
- **Characteristics of included reviews**
- **Effect of physiotherapy intervention versus nothing/placebo**
- **Effect between physiotherapy interventions**
- **Effect of physiotherapy intervention versus non-physiotherapy intervention**

DISCUSSION

CONCLUSION

INTRODUCTION

There are many systematic reviews located on the electronic databases relating to physiotherapy intervention for children with cerebral palsy. However, the majority of reviews have found little evidence to support some of the more 'traditional' physiotherapy interventions (Turnbull 1993, Butler & Darrah 2001, Brown & Burns 2001). Traditional intervention for children with cerebral palsy has primarily involved 'neurodevelopmental therapy', commonly known as NDT, and evolved from the earlier work of Karel and Berta Bobath (1957, 1963, 1969). Furthermore, most of these reviews have included a mixture of case studies and randomised controlled trials and have drawn conclusions based on all of the studies included in the review. Therefore, conclusions for effect of intervention are being made on lower levels of evidence.

So are there interventions which **can** influence motor outcomes in children with cerebral palsy? If so, is the evidence for effect strong enough to make clinical recommendations? It is known that randomised controlled trials provide the best-evidence for the validity or strength of conclusions (Herbert et al 2005). Therefore, revisiting the question of the effect of intervention by summarising reviews using only the highest level of evidence would enable stronger conclusions to be made which would guide clinical practice. The aim of this study therefore was to examine and summarise the best-evidence available from current systematic reviews by only examining the results from randomised controlled trials.

The research questions for this study were:

In children with cerebral palsy, is physiotherapy intervention effective in:

1. reducing impairment? (weakness, spasticity, contracture, motor in-coordination)
2. improving activity? (gait, upper limb dysfunction)
3. promoting participation? (home, school or community activities, or quality of life)

METHODS USED TO CONDUCT THE REVIEW

Identification and selection of reviews

The following electronic databases were searched from March 2000 to December 2008 for systematic reviews that evaluated the evidence for physiotherapy intervention for children with cerebral palsy (CP): AMED, CINAHL, CDSR, ACP Journal Club, DARE, CCTR, CLCMR, CLHTA, CLEED, Ovid MEDLINE(R) Search Strategy, Web of Science, and PEDro. Inclusion criteria were determined a priori and to be included in this study, reviews could include any research design but had to also contain randomised controlled trials of physiotherapy intervention, be published in English as a full paper, and participants had to be children with a diagnosis of cerebral palsy (any classification). Keywords/search terms used – exploded terms of cerebral palsy child, adolescent, paediatrics or pediatrics, physiotherapy or physical therapy, physiotherapy modalities, neurological paediatric/pediatric physiotherapy (including related terms), systematic reviews. A manual search of the reference lists from the published papers was also performed in order to identify other potential papers which might not have appeared in the electronic database search. One reviewer applied the search strategy to each of the electronic databases and screened the papers for inclusion. The methods sections of

the retrieved papers were extracted and then reviewed by 2 independent reviewers using predetermined criteria (Figure 2.1).

Research design
- reviews of RCT. Where reviews also contain non-randomised trials (eg, pre- post- trials, case series, etc), results for RCT are reported separately
- published as a full paper, review published after 2000, English language only.

Participants
- children under the age of 18 years
- diagnosis of cerebral palsy (all classifications)

Intervention
- any physical therapy intervention

Outcomes
- impairment (weakness, spasticity, contracture, motor incoordination), activity limitation (gait, upper limb) or participation restriction (home, school, community activities, QOL)

Comparison
- physiotherapy vs placebo/nothing: PT vs placebo/nothing OR PT1 + PT2/other vs nothing/placebo + PT2/other
- physiotherapy vs physiotherapy interventions : PT1 vs PT2 OR PT1 + PT3/other vs PT2 + PT3/other
- physiotherapy vs non-physiotherapy interventions: PT vs other

RCT = randomised controlled trial; PT = physiotherapy (1, 2 and 3 refer to the other type of physiotherapy intervention)

Figure 2.1*: Inclusion criteria for summary of systematic reviews.*

Reviewers were therefore blinded to journal, author, title, results and place of publication. A third reviewer was available to resolve issues where reviewers had difficulty reaching a consensus for inclusion.

Assessment of characteristics of reviews

Participants, intervention, and outcome measures

Details on the participants, intervention and outcome measures were extracted in order to examine similarities between the reviews. Information obtained from the

reviews included the number and research design of the trials, number and age of the participants, classification and severity of cerebral palsy, description of the intervention, and identification of the control group.

Quality

The quality of the included reviews was assessed using a scale developed by Hoving et al (2001) (Appendix A). This scale was used as it has been developed specifically to assess the quality of systematic reviews in rehabilitation research. The scale is comprised of 9 criteria with 18 points being the maximum score. The scale assesses search methods (Criteria 1 and 2), selection methods (Criteria 3 and 4), validity (Criteria 5 and 6) and synthesis of findings (Criteria 7, 8, and 9). The reviews were rated as high quality (12-18 points), moderate quality (6-11 points) or low quality (0-5 points). The quality assessment was performed by 2 reviewers independently with any discrepancies resolved through discussion until consensus was reached. A third reviewer was available to adjudicate if a consensus could not be reached.

Analysis of reviews

Data extraction was completed by one reviewer and then independently checked by a second reviewer. This data was summarised under one of the following three comparisons: physiotherapy intervention versus placebo/nothing, physiotherapy intervention versus another physiotherapy intervention, and physiotherapy intervention versus non-physiotherapy intervention. In the first comparison, a physiotherapy intervention was either compared to a placebo or no intervention, or could also be combined with another physiotherapy intervention and then compared

to a placebo/ no intervention plus the other physiotherapy intervention. In the second comparison, a physiotherapy intervention could either be compared with another intervention alone, or in combination with a third intervention. In the third comparison, physiotherapy was just compared with a non-physiotherapy intervention. Where some reviews included uncontrolled clinical trials, results were extracted only from the randomised controlled trials. Results were reported on the number of trials from each review producing each outcome. The effects of the physiotherapy intervention are reported as either better, worse, or no different to the comparison at the level of impairment, activity and participation.

RESULTS

Flow of reviews through the study

The initial search yielded 1180 reviews. After initial screening, 38 potentially relevant reviews were retrieved for further analysis. Following the application of the inclusion criteria, 8 systematic reviews were finally included (Ade-Hall & Moore 2000, Blackmore et al 2007, Boyd & Hays 2001, Boyd et al 2001, Hoare et al 2007, Kerr et al 2004, Mockford & Caulton 2008, Rogers et al 2008) (Figure 2.2).

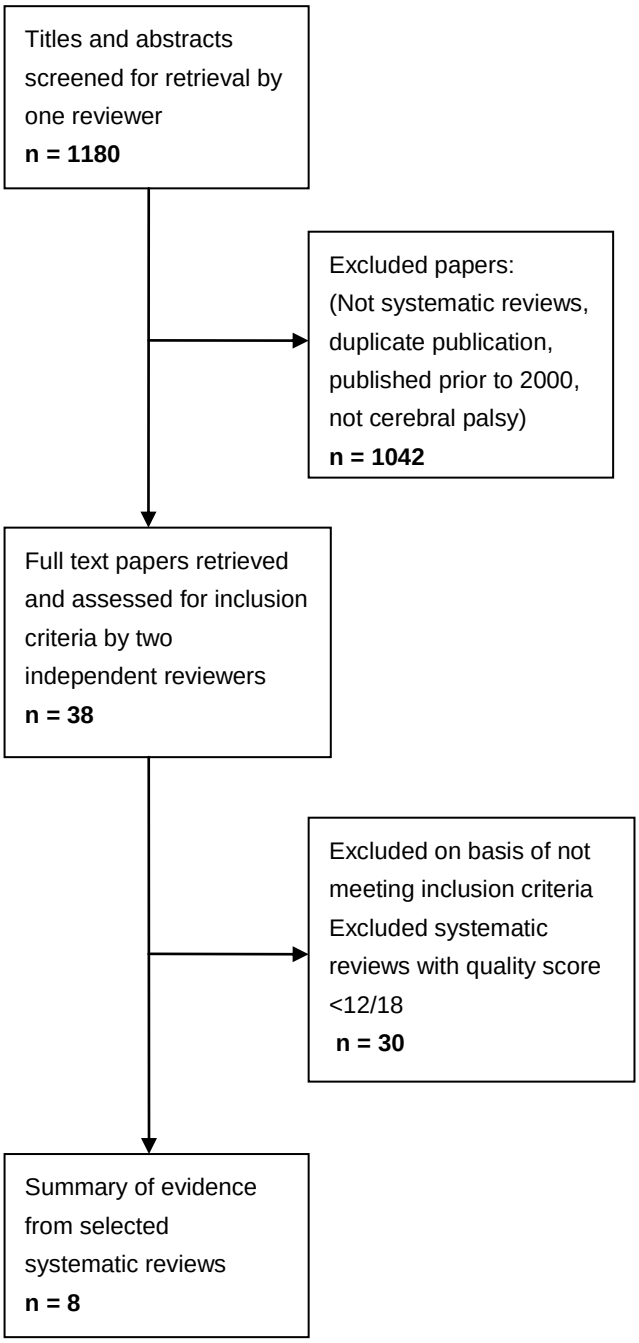

Figure 2.2: *Identification and selection of systematic reviews.*

Characteristics of included reviews

The characteristics of the reviews are summarised in Table 2.1. Six of the eight reviews included a mixture of randomised controlled and uncontrolled trials. The eight reviews comprised 24 randomised controlled trials.

Table 2.1: Summary of included systematic reviews of physiotherapy intervention (A) versus comparison (B).

Systematic review	Design*	Randomised controlled trials		
		Participants	**Intervention**	**Outcomes investigated**
Ade-Hall & Moore (2000)	RCT = 2 (Other = 1)	n = 40 Class = diplegia, hemiplegia, quadriplegia, triplegia Age = 2–11 yr	A = casting, B = BTX-A (n = 2)	Imp = spasticity, contracture Act = gait Part = quality of life, parental perception
Blackmore et al (2007)	RCT = 6 (Other = 13)	n = 142 Class = diplegia, hemiplegia, quadriplegia Age = 2–11 yr	A = casting, B = BTX-A (n = 3) AND A = casting + BTX-A, B = BTX-A (n = 2) AND A = casting, B = usual PT (n = 1)	Imp = weakness, spasticity, contracture Act = gait Part = none
Boyd & Hays (2001)	RCT = 3 (Other = 7)	n = 77 Class = diplegia, hemiplegia, quadriplegia Age = 1 yr 7 mth–9 yr	A = casting, B = BTX-A (n = 2) AND A = usual PT, B = BTX-A (n = 1)	Imp = weakness, spasticity, contracture Act = gait Part = quality of life
Boyd et al (2001)	RCT = 1 (Other = 4)	n = 29 Class = diplegia, hemiplegia, quadriplegia, triplegia Age = 2–10 yr	A = PT, B = BTX-A (n = 1)	Imp = weakness, spasticity, contracture Act = upper limb Part = quality of life
Hoare et al (2007)	RCT = 2	n = 49 Class = hemiplegia Age = 7 mth–8 yr	A = CIMT, B = PT (n = 2)	Imp = weakness, spasticity, contracture Act = upper limb Part = quality of life
Kerr et al (2004)	RCT = 6	n = 181 Class = diplegia, hemiplegia, quadriplegia Age = 8 mth – 18 yr	A = ES + PT, B = PT (n = 5) AND A = ES + PT, B = Placebo- ES + PT (n = 1)	Imp = weakness, spasticity, contracture Act = gait, sitting balance Part = none
Mockford & Caulton (2008)	RCT = 3 (Other = 10)	n = 61 Class = diplegia, hemiplegia, quadriplegia Age = 4–18 yr	A = strength training, B = PT (n = 3)	Imp = weakness, spasticity, contracture Act = gait, upper & lower limb Part = none
Rogers et al (2008)	RCT = 1 (Other = 2)	n = 20 Class = diplegia, tetraplegia, Age = 7–13 yr	A = aerobic ex. + PT, B = PT (n = 1)	Imp = weakness, spasticity, contracture Act = upper limb Part = quality of life

BTX-A = botulinum toxin type A; Imp = impairment, Act = activity, Part = participation; PT = usual physiotherapy intervention; ES = electrical stimulation; CIMT = constraint-induced movement therapy; RCT = randomised controlled trial.
*Type and number of studies - only the RCTs in the reviews which answered the questions for comparison were analysed

Participants

There were 599 children in the reviews who had cerebral palsy and ranged in age from 7 months to 18 years. The classifications of cerebral palsy included diplegia, hemiplegia, quadriplegia and tetraplegia. Only one review (Boyd et al 2001) reported the severity of cerebral palsy from one trial using the Gross Motor Function Classification Scale (GMFCS) (Palisano et al 1997).

Intervention

The physiotherapy interventions examined comprised electrical stimulation, casting, constraint-induced movement therapy, strength training, fitness training and usual physiotherapy intervention. The term 'usual physiotherapy' was used where the physiotherapy intervention was not stated or was reported as either 'usual neurodevelopmental therapy' or 'regular physiotherapy'. The comparisons were either, nothing/placebo, usual physiotherapy intervention or botulinum toxin type-A.

Outcome measures

Impairments investigated included spasticity (Ashworth Scale; Modified Ashworth Scale; Tardieu scale), contracture (goniometric range of motion; x-ray), and weakness (unilateral heel raises; manual muscle test; grip strength; hand-held dynamometer; cycle ergometry). Activities investigated included amount of physical activity (Timed stair test; Timed 10-m walk; total energy expenditure/resting metabolic rate; Physician's Rating Scale; 3D gait analysis, 2-D kinematics, Gross Motor Function Measure (Dimensions D & E), Pediatric Motor Activity Log, Paediatric Evaluation of Disability Inventory; Child Arm Use Test, Emerging Behaviours Scale, WeeFIM, Quality of Upper Extremity Skills Test; Box and Blocks

Test; Erhardt Developmental Prehension Assessment). Participation investigated included quality of life and parent perception (Global Scoring Parent Satisfaction; parent questionnaires; parent perception). All eight reviews reported impairment and activity outcomes whereas only two of those reviews reported participation outcomes.

Quality

All reviews were high quality with a mean score of 15 out of 18 (range 12-16) on the criteria developed by Hoving et al 2001 (Table 2.2).

Table 2.2: Quality of systematic reviews based on criteria developed by Hoving et al (2001).

Systematic review	Score									
	Search methods		Selection methods		Validity		Synthesis			Total
	Criteria 1 Stated	Criteria 2 Comprehensive	Criteria 3 Reported	Criteria 4 Unbiased	Criteria 5 Reported	Criteria 6 Appropriate	Criteria 7 Findings combined	Criteria 8 Combined approp.	Criteria 9 Conclusions approp.	(0-18)
Ade-Hall & Moore (2000)	2	2	2	1	1	2	2	2	2	16
Blackmore et al (2007)	2	2	2	1	2	2	1	1	2	15
Boyd & Hays (2001)	2	2	2	0	2	2	2	2	1	15
Boyd et al (2001)	2	2	2	0	2	2	2	2	1	15
Hoare et al (2007)	2	1	2	1	2	2	2	2	2	16
Kerr et al (2004)	2	2	1	1	2	2	0	0	2	12
Mockford & Caulton (2008)	2	2	2	1	2	2	1	1	2	15
Rogers et al (2008)	2	2	2	1	2	2	1	1	2	15

2 = Yes, 1 = partial or can't tell, 0 = No, 0-5 = poor quality, 6-11 = moderate quality, 12-18 = high quality

Effect of physiotherapy intervention versus placebo/nothing

Table 2.3 summarises the effects of casting and electrical stimulation on impairment, activity or participation from 9 randomised controlled trials in 2 reviews (Blackmore et al 2007, Kerr at al 2004). Both reviews included uncontrolled trials.

Casting

One review (Blackmore et al 2007) with 3 randomised controlled trials examined casting. Two trials (Ackman et al 2005, Bottos et al 2003) found casting and botulinum toxin type A to be no different than botulinum toxin type A alone in improving weakness, spasticity, contracture or activity. One trial (Bertoti 1986) found casting and usual therapy to be more effective than usual therapy alone in improving activity. There was no meta-analysis and the authors concluded that there was no clear evidence that casting is beneficial, either alone, or in combination with Botulinum Toxin A.

Electrical stimulation

One review (Kerr et al 2004) with 6 randomised controlled trials examined electrical stimulation. One trial (Steinbock et al 1997) found electrical stimulation and usual therapy to be more effective than usual therapy alone in improving weakness and contracture. One trial (Dali et al 2002) found electrical stimulation and usual therapy to be no different than placebo electrical stimulation and usual therapy in improving weakness, spasticity or contracture. Two trials (Steinbock et al 1997, Park et al 2001) found electrical stimulation and usual therapy to be more effective whereas one trial (Sommerfeldt et al 2001) found it to be no different than usual therapy alone in improving activity. One trial (van der Linden et al 2003) found electrical

stimulation and usual therapy to be no different than usual therapy alone in improving participation. There was no meta-analysis and the authors concluded that there was insufficient statistical power to conclude that electrical stimulation is beneficial, either alone, or in combination with usual therapy.

Table 2.3: Effect of physiotherapy (A) versus placebo/nothing (B) based on the randomised controlled trials only.

Review	Comparison	Results			Author's conclusions*
		Impairment	Activity	Participation	
Casting					
Blackmore et al (2007)	A = casting + PT B = PT (n = 1)	Weakness = no results * Spasticity = no results Contracture = no results	Gait = ↑ (n = 1)		No clear evidence that any of the three procedures (casting, Botulinum toxin type A, or a combination of the two) is superior to the others.
	A = casting + BTX-A B = BTX-A (n = 2)	Weakness = ↔ (n = 2) Spasticity = ↔ (n = 2) Contracture = ↔ (n = 2)	Gait = ↔ (n = 2)		
Electrical stimulation					
Kerr et al (2004)	A = ES + PT B = PT (n = 5)	Weakness = ↑ (n = 1) ↔ (n = 2) Spasticity = ↔ (n = 1) Contracture = ↑ (n = 1) ↔ (n = 1)	Gait = ↔ (n = 2) Sitting = ↑ (n = 2)	Parent perception = ↔ (n = 1)	Insufficient statistical power to provide conclusive evidence for or against electrical stimulation.
	A = ES + PT B = Plac- ES + PT (n = 1)	Weakness = ↔ (n = 1) Spasticity = ↔ (n = 1) Contracture = ↔ (n = 1)			

BTX-A = botulinum toxin type A; ES = electrical stimulation; n = number of studies; plac = placebo; PT = physiotherapy, ↑ = physiotherapy intervention is better than other: ↓ = physiotherapy is worse than other; ↔ = no difference between interventions; UL = upper limb; LL = lower limb, * = impairments, activities, participation aimed to be investigated by authors have no results reported in reviews

*Author's conclusions are based on all the studies presented in the review and not just the randomised controlled trials

Effect between physiotherapy interventions

Table 2.4 summarises the effects of constraint-induced movement therapy, strength training and cardiorespiratory fitness on impairment, activity or participation from 6 randomised trials in 3 reviews (Hoare et al 2007, Mockford & Caulton 2008, Rogers et al 2008). Two reviews (Mockford & Caulton 2008, Rogers et al 2008) included uncontrolled trials.

Constraint induced movement therapy

One review (Hoare et al 2007) with 2 randomised controlled trials (DeLuca 2002, Sung et al 2005) examined constraint induced movement therapy. The authors concluded that although there was a significant treatment effect for constraint induced movement therapy in a single trial, there was limited evidence for the use of this intervention.

Strength training

One review (Mockford & Caulton 2008) with 3 randomised controlled trials (Dodd et al 2003, Jiang et al 2006, Liao et al 2007) examined strength training. All three trials found strength training to be more effective than usual therapy in improving weakness and one trial (Jiang et al 2006) found it to be more effective at improving contracture. One trial (Jiang et al 2006) found strength training to be no different than usual therapy in improving spasticity. Two trials (Jiang et al 2006, Liao et al 2007) found strength training to be more effective than usual therapy in improving activity while one trial (Dodd et al 2003) found it to be no different. There was no meta-analysis and the authors concluded that there was no clear evidence that strength training is beneficial, either alone, or in combination with usual physiotherapy.

Cardiorespiratory fitness

One review (Rogers et al 2008) with one randomised controlled trial (van den Berg-Emons et al 1998) examined cardiorespiratory training. This trial found cardiorespiratory training to be more effective than usual therapy in improving aerobic power but no different in improving weakness or activity. There was no meta-analysis and the authors concluded that there was evidence that cardiorespiratory fitness training is beneficial.

Table 2.4: Effect between physiotherapy interventions (A) and (B) based on the randomised controlled trials only.

Review	Comparison	Results			Author's conclusions**
		Impairment	Activity	Participation	
Constraint Induced Movement Therapy					
Hoare et al (2007)	A = CIMT B = usual PT (n = 2)	Weakness = no results * Spasticity = no results Contracture = no results	Upper limb = ↑ (n = 1), ↔ (n=1)	QOL – no results*	Significant treatment effect using modified constraint-induced movement therapy in a single trial. Given the limited evidence the use of this therapy should be considered experimental in children with cerebral palsy
Strength training					
Mockford & Caulton (2008)	A = strength training B = usual PT (n = 3)	Weakness = ↑ (n = 3 Spasticity = ↔ (n = 1) Contracture = ↑ (n = 1)	Gait = ↑ (n = 2) ↔ (n = 1)		Isotonic strength training associated with moderate–to-large strength gains but inconclusive results regarding gait and function.
Cardiorespiratory fitness					
Rogers et al (2008)	A = aerobic training, B = usual PT (n = 1)	Aerobic power = ↑ (n = 1) Weakness = ↔ (n = 1)	Physical activity = ↔ (n = 1)	QOL – no results*	Evidence suggests that children with cerebral palsy who perform aerobic exercise training demonstrate improvements in physiological measures of aerobic fitness.

BTX-A = botulinum toxin type A; n = number of studies; PT = usual physiotherapy, QOL = quality of life; ↑ = physiotherapy intervention is better than other: ↓ = physiotherapy is worse than other; ↔ = no difference between interventions; * = impairments, activities, participation aimed to be investigated by authors have no results reported in reviews.

** Author's conclusions are based on all the studies presented in the review and not just the randomised controlled trials.

Effect of physiotherapy intervention versus non-physiotherapy intervention

Table 2.5 summarises the effects of casting and usual physiotherapy on impairment, activity or participation from 9 randomised controlled trials in 4 reviews (Ade-Hall & Moore 2000; Blackmore et al 2007; Boyd & Hays 2001; Boyd et al 2001). Three of these reviews included uncontrolled trials (Blackmore et al 2007; Boyd & Hays 2001; Boyd et al 2001).

Casting

Three reviews (Ade-Hall & Moore 2000; Blackmore et al 2007; Boyd & Hays 2001) reported on the same 2 randomised controlled trials (Corry et al 1998 & Flett et al 1999). One review (Blackmore et al 2007) also contained another trial (Ackman et al 2005). Two reviews (Ade-Hall & Moore 2000, Blackmore et al 2007) found casting to be no different than botulinum toxin type A in improving spasticity or contracture. Whereas one review (Boyd & Hays 2001) reported casting in the trial by Corry et al (1998) as being less effective than botulinum toxin type A. All reviews found casting to be no different than botulinum toxin type A in improving activity. One trial (Flett et al 1999) found casting to be less effective than botulinum toxin type A in improving participation. There was no meta-analysis in two reviews (Ade-Hall & Moore 2000, Blackmore et al 2007) and the authors concluded that there was no clear evidence that casting is beneficial, either alone, or in combination with botulinum toxin type A. Two reviews had a meta-analysis (Boyd & Hays 2001, Boyd et al 2001) and the authors concluded that there was high level evidence that botulinum toxin type A was beneficial. However, in these reviews examination of botulinum toxin type A, and not casting per se, was the stated aim of the study.

Usual physiotherapy

Two reviews with 2 randomised controlled trials (Boyd & Hays 2001, Boyd et al 2001) examined usual physiotherapy. One trial (Boyd et al 2001) found usual therapy to be less effective than botulinum toxin type A in improving weakness, spasticity, contracture and activity. One trial (Boyd & Hays 2001) found usual therapy to be no different than botulinum toxin type A in improving activity. Both reviews had a meta-analysis and the authors concluded that there was high level evidence that botulinum toxin type A was more beneficial than usual physiotherapy. However, in both reviews examination of botulinum toxin type A, and not physiotherapy per se, was the stated aim of the study.

Table 2.5: Effect of physiotherapy intervention (A) versus non-physiotherapy intervention (B) based on the randomised controlled trials only.

Review	Comparison	Results			Author's conclusions**
		Impairment	Activity	Participation	
Casting					
Ade-Hall & Moore (2000)	A = casting B = BTX-A (n = 2)	Spasticity = ↔ (n = 2) Contracture = ↔ (n = 2)	Gait = ↔ (n = 2)	Parental perception = ↓ (n = 1) Quality of life = no results*	No evidence to support or refute the use of Botulinum toxin type A in improving function.
Blackmore et al (2007)	A = casting B = BTX-A (n = 3)	Weakness = ↔ (n = 1) Spasticity = ↔ (n = 2) Contracture = ↔ (n = 3)	Gait = ↔ (n = 1)		No clear evidence that any of the three procedures (casting, Botulinum toxin type A, or a combination of the two) is superior to the others.
Boyd & Hays (2001)	A = casting B = BTX-A (n = 2)	Spasticity = ↔ (n = 1) Contracture = ↓ (n = 1)	Gait = ↔ (n = 1)	Quality of life = no results	High level and quality of evidence for Botulinum toxin type A in most aims of management; moderate size treatment effect for gait that is dose dependent.
Physiotherapy intervention					
Boyd & Hays (2001)	A = usual PT B = BTX-A (n = 1)	Weakness = no results* Spasticity = no results Contracture = no results	Gait = ↔ (n = 1)	Quality of life = no results	High level and quality of evidence for Botulinum toxin type A in most aims of management; moderate size treatment effect for gait that is dose dependent.
Boyd et al (2001)	A = usual PT (NDT) B = BTX-A (n = 1)	Weakness = ↓ (n = 1) Spasticity = ↓ (n = 1) Contracture = ↓ (n = 1)	Upper limb = ↓ (n = 1)	Quality of life = no results	Paucity of evidence for most treatment approaches for upper limb dysfunction in children with cerebral palsy

BTX-A = botulinum toxin type A; NDT = neurodevelopmental therapy; n = number of studies; PT = physiotherapy, ↑ = physiotherapy intervention is better than other: ↓ = physiotherapy is worse than other; ↔ = no difference between interventions; * = impairments, activities, participation aimed to be investigated by authors have no results reported in reviews.
**Author's conclusions are based on all the studies presented in the review and not just the randomised controlled trials.

DISCUSSION

This review identified 8 systematic reviews examining the effects of 6 physiotherapy interventions for children with cerebral palsy. While the reviews were deemed to be of high quality, the low number of randomised controlled trials within the reviews means that these results have limited credibility for application to clinical practice. In addition, the evidence identified from this review is not convincing as there are contradictory results at both the impairment and activity levels and no evidence at the level of participation. For example, the evidence in one review that electrical stimulation is beneficial as an adjunct therapy is contradicted by evidence in another review that it has no effect. The evidence for the interventions of casting and usual physiotherapy shows that they had no effect and in addition may be worse than botulinum toxin type A at the levels of impairment and activity. Likewise the evidence that constraint-induced movement therapy is beneficial at the level of activity is contradicted by other evidence that it has no effect. While there is evidence that strength training and cardiorespiratory fitness training are beneficial at the impairment level, this is less certain at the activity level. The strength of this evidence is further undermined by the fact that most of the reviews contained few controlled trials.

So what are the implications for further research? Several issues arise from this study which could be applied to future trials. For example, only conducting randomised controlled trials which investigate an intervention versus nothing or a placebo may provide more compelling evidence than uncontrolled trials at the level of impairment and activity. Likewise systematic reviews which only include randomised controlled trials would overall provide more robust evidence for

physiotherapy intervention. Many studies within the reviews included participants with a range of classifications of cerebral palsy. Only one trial in one review reported severity of disability as determined by the Gross Motor Function Classification Scale (GMFCS) (Palisano et al 1997) therefore the similarity of the participants between studies could not be assessed. Furthermore, no conclusions could be made regarding the effect of intervention on the level of severity of cerebral palsy and the lack of significant findings could be attributed to the diverse range of abilities within these participants. Including the most appropriate level for the intervention, ie, choosing Level I and/or II GMFCS for investigation of walking ability may be more appropriate. A further issue was identified with the choice of outcome measures used particularly at the activity level. For example the Gross Motor Function Measure (GMFM) (Russell et al 1989) was developed as a measure of motor performance but in many studies was frequently used as a measure of capacity. In addition, the failure of many studies to find any carryover to activity may be more reflective of the outcome measures used rather than the intervention.

Given that at this point there is no compelling evidence to guide clinical practice, how can these results be utilised in the meantime? For the interventions which showed some benefit at the impairment level but failed to carry over to activity, then examining the intensity and duration of the intervention may be useful. For the interventions which showed no difference, or were worse, than the comparison then discontinuing those interventions and examining other forms of intervention would be more beneficial.

CONCLUSION

There is limited and contradictory evidence about the effect of physiotherapy intervention in children with cerebral palsy. However, there are more than 80 randomised controlled trials located on PEDro which examine physiotherapy intervention for these children. Therefore, future research should be directed towards organising these trials, and those on other electronic databases, into systematic reviews, with meta-analyses, in order to provide specific evidence about the size of the effect of physiotherapy intervention in children with cerebral palsy. In particular, given that there is emerging evidence for cardiorespiratory fitness and strength training for children with cerebral palsy, these would seem to be relevant interventions to investigate. The following chapters outline preliminary investigations of cardiorespiratory fitness in cerebral palsy.

CHAPTER 3

CARDIORESPIRATORY FITNESS IN CHILDREN

DEFINITION

INCREASING CARDIORESPIRATORY FITNESS

TYPICALLY-DEVELOPING CHILDREN

Cardiorespiratory fitness values

Measurement of cardiorespiratory fitness

CHILDREN WITH CEREBRAL PALSY

DISCUSSION

DEFINITION

The promotion and maintenance of cardiorespiratory fitness in all ages is well recognised as being desirable for optimal health and for prevention of age-related disease (WHO 2001, Strong et al 2005). In typically developing children cardiorespiratory fitness is also necessary to ensure adequate levels of physical activity and subsequent development of motor skills (Stout 2012). However, children with cerebral palsy have been identified as having inadequate cardiorespiratory fitness which may be contributing to secondary physical impairments of pain, fatigue and osteoporosis (Fowler et al 2007). It is also recognised that these children have activity limitations compared to their typically-developing peers. In order to optimise cardiorespiratory fitness in children with cerebral palsy it is important for physiotherapists to have an understanding of what comprises 'normal' cardiorespiratory fitness in typically-developing children and how cardiorespiratory fitness is evaluated in children.

Cardiorespiratory fitness is an element of the commonly used term 'physical' fitness and is described as the body's response, or adaptation, to the demands and stress of physical effort (American College of Sports Medicine 2000). In particular, cardiorespiratory fitness relates to the ability of the circulatory and respiratory systems of the body to supply oxygen to skeletal muscles during sustained, moderate to high intensity level, physical activity (American College of Sports Medicine 2000). Aerobic capacity is the term used to describe the highest amount of oxygen consumed during maximal exercise in activities that use the large muscle groups in the legs or arms and legs combined (American College of Sports Medicine 2000). Aerobic capacity, aerobic power, functional capacity, functional aerobic capacity,

maximal functional capacity, cardiorespiratory endurance, cardiorespiratory fitness, cardiovascular fitness, maximal oxygen intake, and maximal oxygen uptake are all terms that are often used interchangeably. For the purpose of this thesis, the term cardiorespiratory fitness is used.

One of the main determinants of cardiorespiratory fitness is measured by maximum oxygen uptake (VO_{2max}). VO_{2max} is the highest amount of oxygen that an individual can consume while performing a dynamic exercise (Washington et al 2008). Adults generally reach a plateau at VO_{2max} with no further increases in rate of work. A VO_{2max} value of 42 ml/kg/min for males and 35 ml/kg/min for females is considered to be indicative of good cardiorespiratory health (American College of Sports Medicine 2000). However, studies show that VO_{2max} will be approximately 25% less than a young adult (Washington et al 2008). Table 3.1 shows VO_{2max} values for males and females aged 20 to 60 plus years.

Table 3.1: Maximal oxygen uptake ($VO_{2max)}$ mean (SD) values for adults measured in ml/kg/min*.

	Age(yr)				
Gender	**20-29**	**30-39**	**40-49**	**50-59**	**60+**
Males	42.8(5.4)	41.0 (5.6)	38.6 (5.5)	35.5 (5.4)	32.2 (5.9)
Females	35.6 (5.0)	33.6 (4.7)	31.3 (4.6)	27.9 (4.3)	26.7 (4.5)

*Modified data from American College of Sports Medicine (2000).

INCREASING CARDIORESPIRATORY FITNESS

The physiological elements required for increasing cardiorespiratory fitness in both adults and children are largely the same. In order to improve cardiorespiratory

fitness, it is generally recognised that an individual should perform a physical activity, or exercise, at an intensity level of between 40 to 85% of their aerobic capacity (American College of Sports Medicine 2000). In terms of physical activity, this range of aerobic capacity is referred to as moderate to intense (vigorous) and has been used in public health guidelines as a recommendation for daily physical activity (Department of Health and Ageing 2005). Moderate to intense exercise is usually between 40 to 60% of aerobic capacity and can be comfortably sustained up to 60 minutes if there is a gradual progression and the activity is generally non-competitive (American College of Sports Medicine 2000). Therefore, according to these guidelines, individuals should perform up to 60 minutes moderate to intense activity each day in order to maintain their cardiorespiratory fitness.

Subjectively, moderate to intense exercise should cause little or no discomfort, little increase in breathing, and should be well within a person's capability (American College of Sports Medicine 2000). For individuals who are less fit, or who have less capacity, cardiorespiratory fitness can still be achieved at intensity levels below 40% of aerobic capacity (American College of Sports Medicine 2000). The ability to achieve cardiorespiratory fitness at lower aerobic capacity levels may be particularly relevant for children with cerebral palsy.

TYPICALLY-DEVELOPING CHILDREN

In young children, it is hard to separate cardiorespiratory fitness over and above their natural physical activity levels (American Academy of Pediatrics 1994). In addition, it is also hard to measure cardiorespiratory fitness levels in children as many tests require voluntary participation to fatigue, a compliance issue which is unlikely to be

achieved particularly in very young children. Most estimates also take into account the growth and development changes which occur in childhood, particularly at periods of rapid growth such as adolescence (Pate et al 2006, Sharvtz and Reibold 1990, Rosenthal & Bush 2000). Unlike adults, the main changes in a child's cardiorespiratory response to exercise are seen as result of their age in relation to their cardiorespiratory system (Stout 2012). Children's responses are a direct result of gender, growth and maturation of their lung size and correspondingly, their respiratory capacity (Rosenthal et al 1993, Rosenthal & Bush 2000, Armstrong et al 1997) with approximately 80% of this change occurring between the ages of 6-12 years (Stout 2012).

Interpretation of a child's physiological responses to exercise is based on their age, height and weight in relation to their peak oxygen uptake (VO_{2peak}) (American College of Sports Medicine 2000). VO_{2peak} is expressed either as an absolute rate in litres of oxygen per minute (l/min) or as a relative rate in millilitres of oxygen per kilogram of bodyweight per minute (ml/kg/min).

The VO_{2max} plateau seen in adults is not usually found in children as it is hard to get them to work to exhaustion (Armstrong and Welsman 1994). Indeed, it has been reported that less than a third of children and adolescents achieve a VO_{2max} (Cooper 1995). Therefore, peak oxygen uptake (VO_{2peak}) rather than VO_{2max} is more commonly used when performing a maximal exercise test in children. Peak oxygen uptake (VO_{2peak}) is the maximal amount of oxygen observed during a specific exercise test and may or may not equal VO_{2max} (Washington et al 2008). VO_{2peak} can be measured directly during a specific exercise test by measuring the expired

ventilation and gas fractions using gas analysis equipment. Alternatively, VO_{2peak} can be estimated from an individual's performance during a sub-maximal test. This method involves calculating the VO_{2peak} using a regression equation of heart rate and oxygen uptake at submaximal workloads (Armstrong & Welsman 1994).

There is some debate in the literature, not only as to the relationship between heart rate and oxygen uptake when calculating VO_{2peak}, but also in the validity of using predictive equations for children which have been based on able-bodied adult populations (Shvartz and Reibold 1990, Armstrong and Welsman 1994, Braden and Carroll 1999). However, estimates taken from adult values have been suggested which take into account the developmental differences in children (Blair et al 1989). A minimum VO_{2peak} of 42 ml/kg/min is recommended for males aged 5 to 17 years, and 40 ml/kg/min for females aged 5 to 9 years. A decrease in VO_{2peak} of approximately one unit per year is suggested for females from 10-14 years as they go through puberty and a constant VO_{2peak} of 35 ml/kg/min suggested for 17 years onwards (Blair et al 1989).

Cardiorespiratory fitness values

A child's ability to increase their oxygen uptake during exercise increases with their growth and development. For example, peak oxygen uptake (VO_{2peak}) increases throughout childhood from approximately 0.5 l/min at 8 years to more than 2.5 l/min at adolescence (Shvartz and Reibold 1990, Armstrong and Welsman 1994, Braden and Carroll 1999, Freedson et al 1992, Rowland 1996). Males and females are likely to have similar VO_{2peak} levels until approximately 12 years of age (Shvartz and Reibold 1990, Krahenbuhl et al 1985). Typical VO_{2max} ,or $VO_{2peak,}$ values in healthy

children are 40-50ml/kg/min (Pryor & Prasad 2008) However, following puberty for both males and females, these levels change markedly. At 14 years of age males are likely to have 25% higher VO_{2peak} than females, which doubles to 50% higher at age 16 years (Armstrong and Welsman 1994). The increase in VO_{2peak} levels is closely aligned with skeletal maturity in males and females and also as a result of growth in VO_2 dependent organs such as the heart, lungs, blood volume and skeletal muscle (Braden & Carroll 1999). In addition, the higher increase in VO_{2peak} levels in males are attributed to corresponding increases in their muscular strength and size, but may also be related to higher levels of habitual physical activity than females (Armstrong et al 1990). Table 3.1 shows values for maximal oxygen uptake (VO_{2peak}) measured in l/min for males and females aged 8-14 years and Table 3.2 shows values for maximal oxygen uptake (VO_{2peak}) measured in ml/kg/min for males and females aged 8-18 years.

Table 3.2: Maximal oxygen uptake (VO_{2peak}) mean (SD) values measured in l/min for children aged 8-14 years.

Study	Age (yr)	Gender (M/F)	VO_{2peak} (l/min)
Shvartz & Reibold (1990)	8 – 14	M/F	1.5 (0.6)
Rosenthal & Bush (2000)	10 - 14	M	1.7 (0.2)
Krahenbuhl et al (1985)	8 - 14	M/F	1.8 (0.5)

VO_{2peak} = maximal oxygen uptake; l/min = absolute rate in litres of oxygen per minute; M = male; F = female; yr = year

Table 3.3: Maximal oxygen uptake (VO_{2peak}) mean (SD) values measured in ml/kg/min for children aged 8-18 years.

Study	Age (yr)	Gender (M/F)	VO_{2peak} (ml/kg/min)
Shvartz & Reibold (1990)	8 14	M/F	42.7(3.4)
Krahenbuhl et al (1985)	8-14	M	57(3.6)
Cooper et al (1984)	8-12	M/F	41(6.8)
Pate et al (2006)	12-14	M/F	42.4(4.1)
Ten Harkel et al (2010)	8-18	M F	47(7) 42(6)

VO_{2peak} = maximal oxygen uptake; ml/kg/min = millilitres of oxygen per kilogram of bodyweight per minute; M = male; F = female; yr = year

Measurement of cardiorespiratory fitness

Measurement of a child's response to exercise can either be determined indirectly via 'field tests' or directly using laboratory exercise testing. Field tests give an indirect measure of cardiorespiratory fitness via predictions of maximal oxygen uptake (VO_{2peak}). These tests are commonly standard school-type physical education exercises such as timed shuttle-run tests and timed distance runs/walk. They are generally sub-maximal tests, meaning the child does not perform to exhaustion, and outcome measures of cardiorespiratory fitness are based on the child's performance in the particular test. Measurement may also be supplemented with pre-test resting and post-test recovery heart rates (American College of Sports Medicine 2000). A decrease in resting heart rate is seen to be indicative of cardiorespiratory conditioning (Washington et al 1994).

The concern with sub-maximal tests is that incorrect assumptions for predicting VO_{2peak} may be made. The cardiopulmonary responses of children to submaximal

tests do not always parallel adults (on whom much of the normative data is based), and indeed children often have higher maximal heart rates and a greater variability in range of heart rate than adults (Armstrong & Welsman 1994). Furthermore, estimation of VO_{2peak} based on field tests has shown that in some children peak oxygen uptake may be underestimated (Artero et al 2011). At this point in time, the most reliable field test to predict VO_{2peak} is considered to be the 20-metre shuttle run test (Artero et al 2011, Leger et al 1982, Liu et al 1992, Ramsbottom et al 1988).

Direct measures of cardiorespiratory fitness on the other hand, via laboratory exercise testing, can evaluate a child's physiological responses to exercise using standardised exercise protocols (American College of Sports Medicine 2000). These protocols generally involve a progressive increase in rates of work without a resting period between changes of work increment rates (Washington et al 1994). Several of these protocols have been developed for children using either a cycle ergometer or treadmill eg the Balke Treadmill Protocol and the McMaster Cycle Test (Skinner 1993). Children perform a maximal exercise test until voluntary exhaustion, ie, when the child is unwilling or unable to continue the exercise despite strong verbal encouragement (Armstrong &Welsman 1994). Treadmills in particular, have an advantage over other methods of exercise testing in that they ensure consistent physical movement and can easily be adjusted to increase exercise workload.

In order to directly measure cardiorespiratory fitness, individual measures of heart rate, maximal oxygen uptake, anaerobic threshold, endurance time and total work may be made (Washington et al 1994). Heart rate response to exercise is often used as an indicator of cardiorespiratory fitness. Various formulas are used to estimate

individual maximum heart rates based on age. The most commonly used formula to calculate maximum heart rate (HR_{max}) is: HR_{max} = 220 – the individual's age.

However, maximum heart rates vary significantly between individuals and to date there are no reliable formulae which can accurately predict maximum heart rate (Washington et al 1994). Therefore, heart rate alone is not an adequate determinant of fitness and should be combined with one of the other measures (Washington et al 1994). Some examples of peak heart rates of children using either a treadmill or a cycle ergometer are presented in Table 3.3.

Table 3.4: Peak heart rates mean (SD) of children following an exercise test on a treadmill or cycle ergometer.

Study	Age (yr)	Gender	Peak heart rate (BPM)
Riopel et al (1979)	4-21	M/F	185 (5)
Wilmore et al (1982)	13-15	M	197 (5)
James et al(1980)	5-18	M/F	193 (6)
Alpert et al (1982)	6-15	M/F	190 (4)
Ten Harkel et al (2010)	8-18	M/F	184 (12)

bpm = beats per minute; yr = year; M = male; F = female

The most widely used, and probably most reliable, criterion measure for cardiorespiratory fitness in children is directly measured maximal oxygen uptake (VO_{2peak}) (Armstrong &Welsman 1994, Stout 2011). Direct measures of oxygen uptake are generally taken using a gas analysis system which may be portable or fixed, eg, Cosmed K4b2™, while a child performs a maximal exercise test on either a cycle ergometer or treadmill. Gas analysis systems measure the breath-by-breath gas exchange of oxygen and carbon dioxide. In order to ensure that valid and reliable measures of oxygen uptake are measured while on a treadmill certain

conditions should be achieved (Keefer et al 2004). The child must achieve a steady-state VO_{2peak} value, the amount of treadmill familiarisation should be standardised prior to commencing the test and the child should refrain from holding on to the treadmill while conducting the test (Keefer et al 2004).

Several studies have used a treadmill protocol to determine cardiorespiratory fitness in children with cerebral palsy (Hoofwijk et al 1995, Unnithan et al 1996, Unnithan et al 2003, Keefer et al 2004, Verschuren et al 2010, Maltais et al 2004, Potter & Unnithan, 2005). However, children with cerebral palsy included in these studies had to be ambulatory. The modified Balke has been used for children who are chronically ill and also those with physical disabilities who have varying physical fitness levels (Paridon et al 2006). This protocol involves either increasing the slope of the treadmill, in increasing increments while the speed remains constant, or by increasing the speed at pre-determined intervals. Using direct measures of cardiorespiratory fitness (and potentially in conjunction with field tests) for children with cerebral palsy means that comparison with typically–developing peers is more valid.

CHILDREN WITH CEREBRAL PALSY

It has been noted that overall, children with cerebral palsy do not tend to tolerate sustained periods of exercise and subsequently report early fatigue (Unnithan et al 2004). In addition, these children tend to have poor cardiorespiratory fitness, lower physical work capacity and higher oxygen cost during exercise than their typically-developing peers (van den Berg-Emons et al 1995, Fernhill and Unnithan 2002, Stout

2011). The higher energy cost in children with cerebral palsy subsequently results in increased energy expenditure required for any given exercise. Energy expenditure may be 2-3 times higher, for the same exercise, than seen in typically developing children (Unnithan et al 1999, Unnithan et al 2004, Maltais et al 2005, Piccinini et al 2007, van den Berg-Emons et al 1995, Rose et al 1989).

Several studies have shown that children with cerebral palsy have lower VO_{2peak} levels than their typically-developing peers (Hoofwijk et al 1995, van den Berg-Emons et al 1995, Unnithan et al 1996, Rose et al 1989). Specifically, these VO_{2peak} levels are not only lower but have been shown to be half the expected norm in children with cerebral palsy. Lower levels of VO_{2peak} may be attributed to lower efficiency of breathing as a result of higher than normal ventilator equivalent values for oxygen seen in children with cerebral palsy (Unnithan and Maltais 2004, Hoofwijk et al 1995). Likewise, the early fatigue to exercise seen in these children could be related to musculoskeletal impairment of lower limb weakness, spasticity and contracture. Table 3.4 shows values for peak oxygen uptake measured in L/min and ml/kg/min for males and females aged 10-16 years.

Table 3.5: Maximal oxygen uptake (VO_{2peak}) values measured in either l/min or ml/kg/min for children with cerebral palsy aged 10-16 years.

Study	Age (yr)	VO_{2peak}	
		l/min	ml/kg/min
Hoofwijk et al (1995)	10-16	1.58	32.7
Maltais et al (2005)	10-16	1.25	34.0
Verschuren & Takken (2010)	7-17		42.0

VO_{2peak} = maximal oxygen uptake; l/min = absolute rate in litres of oxygen per minute; ml/kg/min = relative rate in millilitres of oxygen per kilogram of bodyweight per minute; yr = year; M = male; F = female; yr = year

Although heart rate, combined with other measures, may be used to determine cardiorespiratory fitness (Washington et al 1994), in children with cerebral palsy there seems to be contradictory evidence when comparing peak heart rate (HR_{peak}) with oxygen consumption. One study found a linear relationship between heart rate and oxygen consumption when comparing walking speeds in children with cerebral palsy (Rose et al 1989). However in contrast, another study found that there was not a strong relationship between indirectly measured energy expenditure via heart rate and directly measured oxygen uptake (Keefer et al 2004). Regardless, using a heart rate monitor to determine a child's HR_{peak} during an exercise test is considered optimal (Verschuren et al 2011). Where heart rate monitoring is not available, or possible, for a child with cerebral palsy then using a mean of 194 beats per minute as an estimation of HR_{peak} is preferable to using a predictive formula (Verschuren et al 2011).

Both direct and indirect measures of cardiorespiratory fitness have previously been reported in studies on children with cerebral palsy (van den Berg-Emons et al 1995 Unnithan et al 1999, Unnithan et al 2007, Gorter et al 2009, Verschuren et al 2006, Verschuren et al 2009, Verschuren et al 2010, Keefer et al 2004, Keefer et al 2005). For children who are able to walk independently, a walking-based exercise test to directly measure cardiorespiratory fitness would be preferable (Verschuren et al 2006). Previous studies have shown that children with cerebral palsy are capable of performing a progressive exercise test on a treadmill (Hoofwijk et al 1995, Unnithan et al 1999, Keefer et al 2004, Keefer et al 2005, Maltais et al 2004). However, it would seem that indirect measures of cardiorespiratory fitness have generally been

favoured over direct measures due to the cost and availability of necessary measuring instruments and equipment (Keefer et al 2004).

DISCUSSION

It is known that cardiorespiratory fitness in children is important in terms of ensuring physical development and may prevent age-related disease associated with adulthood. Children improve their cardiorespiratory fitness as a result of the process of growth and maturation. However, for children with cerebral palsy, there is increasing evidence that they are more likely to have decreased cardiorespiratory fitness compared to their typically-developing peers. Furthermore, that this decreased cardiorespiratory fitness may be related to a corresponding decrease in physical activity levels (Bjornson et al 2006, Bjornson et al 2007, Rimmer 2001, Rimmer et al 2004, Pirpiris et al 2004, van den Berg-Emons et al 1995) and that for any given activity, the energy cost for children with cerebral palsy may be up to three times greater than for children of a similar age ((Unnithan et al 1999, Unnithan et al 2004, Maltais et al 2005, Piccinini et al 2007, van den Berg-Emons et al 1995, Rose et al 1989). In addition, children with cerebral palsy are more likely to fatigue at lower intensity levels of exercise than typically-developing children.

It is of concern that children with cerebral palsy have decreased cardiorespiratory fitness particularly in relation to lower levels of physical activity. Decreased cardiorespiratory fitness contributes to ongoing physical inactivity due to reduced stamina and aerobic capacity thereby discouraging, or limiting, participation in community activities. If these children are fatiguing earlier, and using twice as much

energy to perform the same activities as typically-developing children, they are less likely to want to be engaged in physical activities. Long-term behavioural patterns of inactivity may then be a consequence going into adulthood.

There are several issues which arise for physiotherapists in terms of improving cardiorespiratory fitness in children with cerebral palsy. Of particular interest is in determining whether physiotherapy intervention in the form of cardiorespiratory training can improve fitness in children with cerebral palsy. However, there is also a need to identify what evidence there is to support cardiorespiratory training as a physiotherapy intervention in these children. Furthermore, if cardiorespiratory fitness can be improved in children with cerebral palsy is there any evidence that there is carryover to physical activity in terms of increasing the amount and intensity of activity? These issues are investigated in the following chapter by a systematic review of the evidence for cardiorespiratory training in children with cerebral palsy.

CHAPTER 4

STUDY 2: EFFECT OF CARDIORESPIRATORY TRAINING ON AEROBIC FITNESS AND CARRYOVER TO ACTIVITY IN CHILDREN WITH CEREBRAL PALSY: A SYSTEMATIC REVIEW

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Assessment of characteristics of studies

Data analysis

RESULTS

Flow of studies through the review

Characteristics of included studies

Effect of cardiorespiratory training

DISCUSSION

CONCLUSION

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INTRODUCTION

The importance of cardiorespiratory fitness for all ages has been well documented. It is also well-known that children have quite different levels and patterns of physical fitness and activity to adults. Of increasing concern has been the realisation that children with cerebral palsy are more likely to have less cardiorespiratory fitness than typically developing children (van den Berg-Emons et al 1995, Fernhill and Unnithan 2002, Stout 2006). These lower levels of cardiorespiratory fitness may be related to a corresponding decrease in everyday physical activity as a result of musculoskeletal impairment. Also likely, is the presence of cardiorespriatory impairment in these children resulting in higher levels of energy expenditure than typically developing children for the same activities (Arvidsoon et al 2007, Dorminy et al 2008, Wickel et al 2007).

Physiotherapy intervention for children with cerebral palsy has rarely included training for cardiorespiratory fitness (Blanchard and Darrah 1999). However there have been two systematic reviews (Verschuren et al 2008, Rogers et al 2008) which have investigated the effect of cardiorespiratory training for children with cerebral palsy. One review included 5 studies (Verschuren et al 2008) and the other included 13 (Rogers et al 2008). Both reviews concluded that training may improve aerobic fitness but that there was no evidence for improvement in activity or participation. In the review with five studies on aerobic fitness (Verschuren et al 2008), one was a randomised trial (van den Berg-Emons 1998) while the other four were case series without a control group. In the review with 13 studies (Rogers et al 2008), the same randomised trial was included (van den Berg-Emons 1998) plus another trial (Dresen et al 1985), which was not randomised, and 11 studies were case series. As most of

the studies in these reviews were not randomised trials, the authors concluded that the level of evidence from which to make recommendations was low. Revisiting this question using the highest level of evidence would enable stronger conclusions to be made to guide clinical practice.

Therefore, we carried out a systematic review following the Cochrane Collaboration guidelines (Higgins & Green 2008) of including only randomised or quasi-randomised trials, where participants received cardiorespiratory training versus placebo or no intervention. Cardiorespiratory training was defined broadly as carrying out activities for an extended period which were progressed in terms of duration and/or intensity over time. Combination programs were only included if cardiorespiratory training comprised at least half of the program. Therefore the research questions for this systematic review were:

1. Does cardiorespiratory training improve aerobic fitness in children with cerebral palsy?
2. Is there any carryover to activity?

METHODS

Identification and selection of studies

Searches were conducted of AMED, CINAHL, CDSR, ACP Journal Club, DARE, CCTR, CLCMR, CLHTA, CLEED, EMBASE, Ovid MEDLINE(R) Search Strategy, Web of Science, and PEDro without language restrictions for studies published up to July 2008 (Appendix B). The search of the databases was optimised by using the terms recommended by the Cochrane Collaboration for the participants (cerebral

palsy), and using the terms used in previous Cochrane reviews for the intervention (children, adolescents, cardiorespiratory fitness, exercise [training], physical training, physical fitness, physical activity, aerobic training, exercise tolerance) and outcomes (fitness, activity). A manual search of the reference lists from the published papers which met the inclusion criteria of this review was also performed in order to identify other potential papers which might not have appeared in the electronic database search.

One reviewer applied the search strategy to each of the electronic databases and screened the papers for inclusion. The methods sections of the retrieved papers were extracted and reviewed by two independent reviewers using predetermined criteria (see Figure 4.1). Reviewers were therefore blinded to author, title, results and place of publication. A third reviewer was available to resolve issues where reviewers had difficulty reaching a consensus for inclusion.

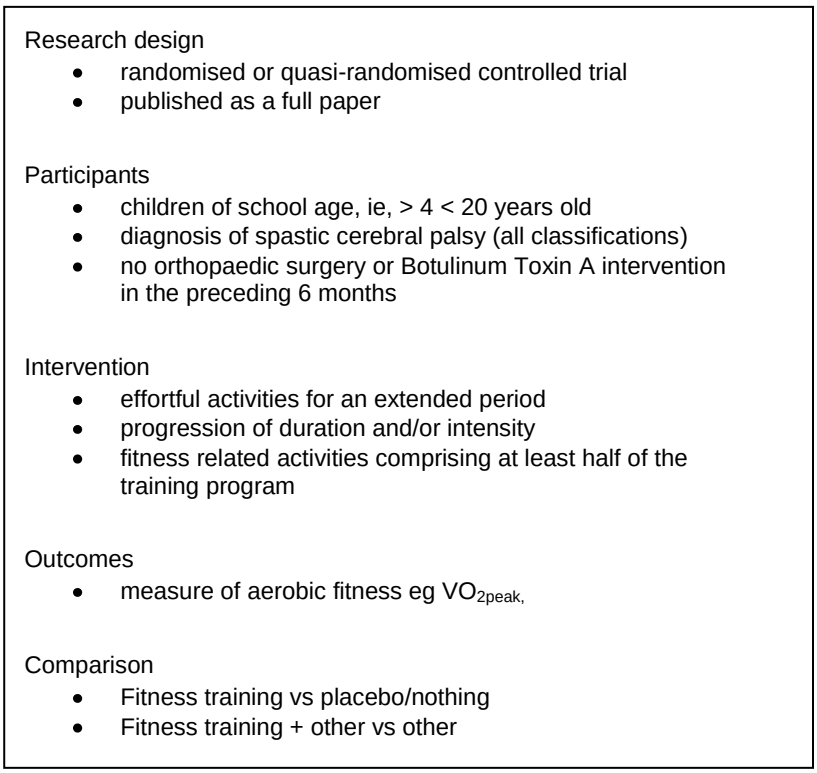

Figure 4.1: *Inclusion criteria for cardiorespiratory fitness.*

Assessment of characteristics of studies

Included studies had to be either randomised controlled trials or quasi-controlled trials. The quality of studies was assessed using the Physiotherapy Evidence Database (PEDro) scale (Maher et al 2003) by extracting the scores located on the PEDro website (http://www.pedro.fhs.usyd.edu.au). Where a study was not included on the database, it was assessed independently by two authors who had completed the PEDro Scale training tutorial located on the Physiotherapy Evidence Database. Participants had to be children of school age, with a diagnosis of spastic cerebral palsy which could be of any classification. Information about severity of disability (eg, using the Gross Motor Function Classification System, Palisano et al 1997) was extracted in order to assess the similarity of the participants between studies. To be

included, the intervention had to focus on cardiorespiratory endurance and outcomes had to include a measure of aerobic fitness, eg, peak oxygen uptake (VO_{2peak}). In addition, inclusions of measures of activity which reflect aerobic fitness, eg, 6 Minute Walk Test were also desirable. Information about frequency, duration, content and intensity of the training and the methods of outcome measures were extracted in order to assess the similarity of the intervention and measurement of outcomes between studies.

Data analysis

Information about the method (design, participants, intervention, outcome measures) and outcome data (number of participants and mean [SD] of aerobic fitness and activity) were extracted by one reviewer and checked by two other reviewers. Where necessary, authors were contacted to provide additional information. Where possible, data were pooled and the mean between-group differences (95% CI) were calculated. Where this was not possible, we calculated the mean difference (% difference) for each study and presented the between-group analysis reported by the authors.

RESULTS

Flow of studies through the review

The search strategy identified 342 studies. Following initial screening, 27 were retrieved for evaluation against the inclusion criteria. 24 studies were excluded at this stage leaving 3 randomised controlled trials for analysis (Unnithan et al 2007,

van den Berg-Emons et al 1998, Verschuren et al 2007). Figure 4.2 shows the process of study selection and exclusion.

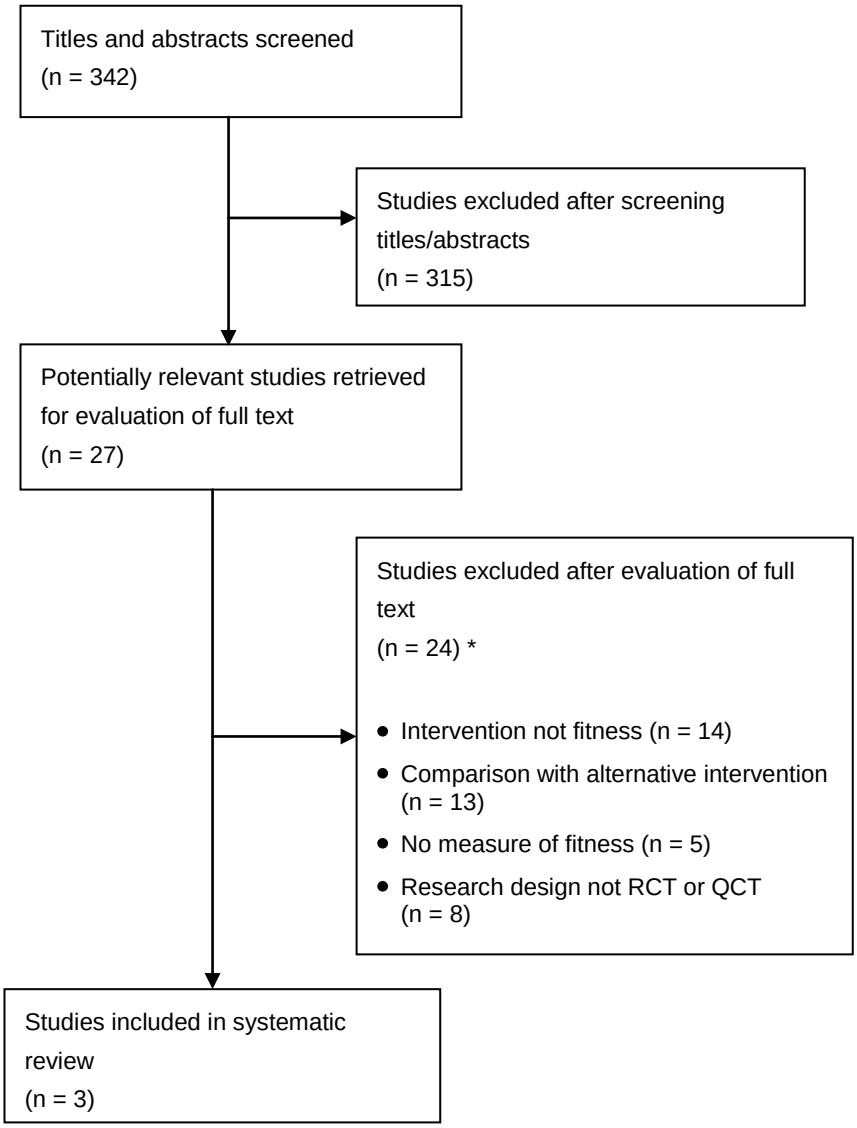

Figure 4.2: *Flow of studies through the review.*
** Studies may have been excluded for failing to meet more than one inclusion criteria.*

Characteristics of included studies

The studies (n = 3) included children with cerebral palsy (n = 101) who ranged in age from 7 to 20 years and comprised both male (n = 59) and female (n= 42) participants. The classification of cerebral palsy included diplegia (n = 52), hemiplegia (n = 45),

and tetraplegia (n = 4), with 2 papers reporting the Gross Motor Function Classification System scores of I – IV. The studies included one short-term intervention (3 months) and two long-term interventions (8 and 9 months). Both of the long-term intervention studies measured outcomes at 2-4 months as well as at 8-9 months after training was completed. The cardiorespiratory training included aerobic exercise such as walking, running, cycling, swimming or stair climbing, 2-4 times per week. One study investigated a combination program of cardiorespiratory and strength training (weights). Intensity of exercise was at a moderate level of 60-75% of maximum heart rate or 70% heart rate reserve. In all studies, the control groups had no additional intervention, but both groups maintained their usual therapy programs which had no aerobic fitness component. Table 4.1 shows the summary of included studies.

Table 4.1: Summary of included studies (n = 3).

Study	Design	Participants	Intervention	Outcome measures
Unnithan et al (2007)	RCT	n = 13 Age = 14-18 yrs Gender = 4 M, 9 F Class = diplegia GMFCS Level II/III	Both = usual PT (individualised NDT45 min, 2/wk) Exp = freq: 70 min 3/wk x 3 mth content: aerobic warm-up (walking), strength training (weights), drills, aerobic interval training intensity: 65 to 75% max HR Con = no additional intervention	Aerobic fitness = $VO_{2,peak}$ (mL kg^{-1} min^{-1}) Activity = GMFM Dimensions D (standing) & E (walking, running, jumping) (%) Follow-up = 0, 3 mth
Van den Berg-Emons et al (1998)	RCT	n = 20 Age = 7-13 yr Gender = 11 M, 9 F Class = diplegia (n = 16), tetraplegia (n = 4)	Both = usual PT (individualised); usual school (45 min 2/wk gymnastics) Exp = freq: 45 min 4/wk x 9 mth content: aerobic ex (cycling, wheelchair driving, running, swimming, mat) intensity: 70% HRR Con = no additional intervention	Aerobic fitness = peak aerobic power (watt/kg) Activity = energy expenditure during daily activity Follow up = 0, 2, 9 mth
Verschuren et al (2007)	RCT	n = 68 Age = 7-20 yr Gender = 44 M, 24 F Class = hemiplegia (n = 45), diplegia (n = 23). GMFCS Level I/II	Both = usual PT (ranged from no intervention to various therapeutic approaches) Exp = freq: 45 min 2/wk x 8 mth content: 8 aerobic & 8 anaerobic ex. (running, step-ups, stair climbing) intensity: 60 - 70% max HR Con = no additional intervention	Aerobic fitness = 10-m Shuttle Run Test (min) Activity = GMFM D & E Follow-up = 0, 4, 8 mth

Exp = experimental group, Con = control group, RCT = randomised controlled trial, class = classification, PE = physical education, PT = physiotherapy; Int = intervention, freq = frequency; HR = heart rate; HRR = heart rate reserve, ex. = exercise, max = maximum; VO_{2peak} = ,peak oxygen uptake, TEE = total energy expenditure, RER = respiratory exchange ratio, CP = cerebral palsy, GMFCS = Gross Motor Function Classification System, GMFM = Gross Motor Function Measure; M = male, F = female, m = metre, min = minute, mth = month; NDT = Neurodevelopmental Therapy.

All 3 studies were randomised trials which had a mean PEDro score of 6 (Table 4.2). Outcome measures of aerobic fitness were variable between studies and included peak VO_2, peak aerobic power and the 10-m Shuttle Run Test. Since the 10-m Shuttle Run Test has been shown to be correlated with aerobic capacity in children with cerebral palsy (Verschuren et al 2006), it was included in the analysis as a measure of aerobic fitness. Measures of activity included a ratio of energy expenditure during daily activity and the Gross Motor Function Measure (GMFM) Dimension D (standing) and E (walking, running, jumping). Although the GMFM does not reflect fitness as much as the 6-min Walk Test, since it was the only measure of activity in two of the studies, it was included in the analysis as a measure of activity.

Table 4.2: PEDro scores for included studies (n = 3).

Study	Random allocation	Concealed allocation	Groups similar at baseline	Participant blinding	Therapist blinding	Assessor blinding	< 15% dropouts	Intention-to-treat analysis	Between-group difference reported	Point estimate and variability reported	Total (0 to 10)
Unnithan et al (2007)	Y	N	Y	N	N	N	Y	N	Y	Y	5
Van den Berg-Emons et al (1998)	Y	N	Y	N	N	N	Y	N	Y	Y	5
Verschuren et al (2007)	Y	Y	Y	N	N	Y	Y	Y	Y	Y	8

PEDro scores from website http://www.pedro.fhs.usyd.edu.au

Effect of cardiorespiratory training

Outcomes were not pooled into a meta-analysis due to a lack of data. Visual examination showed a large variation in baseline scores between the groups and testing of statistical heterogeneity confirmed that this was most significant for aerobic fitness ($p = 0.09$). Heterogeneity of the baseline scores suggests that between-group differences in outcome could be misrepresented if the post-intervention scores were used in a meta-analysis. Although this problem could be overcome if the pre-post change scores were used, they were not available for two of the studies. Therefore, the results of the between-group analyses are reported.

The effect of cardiorespiratory training on aerobic fitness is presented in Table 4.3. After short-term intervention, all 3 studies reported an increase in mean aerobic fitness. The increase was around 20% (range 18-22%). However, not all were statistically significant, suggesting that wide confidence intervals reduced the precision of the estimate. After long-term intervention, both studies reported a statistically-significant increase in mean aerobic fitness of 26 and 41%.

Table 4.3: Effect of cardiorespiratory training on aerobic fitness.

Study	Duration of intervention	Outcome measure	Mean difference between groups Exp minus Con	% increase	Statistical significance of the difference between groups
Short intervention					
Unnithan et al (2007)	3 mth	$VO_{2peak,}$	3.2 mL/kg/min	18%	$p < 0.05$
Van den Berg-Emons et al (1998)	2 mth	Peak aerobic power	0.21 watt/kg (FFM)	21%	Not reported
Verschuren et al (2007)	4 mth	10-m Shuttle Run Test	1.5 min	22%	95% CI = -0.5 to 3.5
Long intervention					
Van den Berg-Emons et al (1998)	9 mth	Peak aerobic power	0.26 watt/kg (FFM)	26%	$p < 0.05$
Verschuren et al (2007)	8 mth	10-m Shuttle Run Test	2.8 min	41%	95% CI = 1.9 to 3.7, $p < 0.001$

Exp = experimental group, Con = control group, RCT = randomised controlled trial, class = classification, PE = physical education, PT = physiotherapy; Int = intervention, freq = frequency, CV = cardiorespiratory, HRR = heart rate reserve, ex. = exercise, VO_{2peak} = ,peak oxygen uptake, TEE = total energy expenditure, RER = respiratory exchange ratio, CP = cerebral palsy, GMFCS = Gross Motor Function Classification System, GMFM = Gross Motor Function Measure; M = male, F = female, m = metre, min = minute, mth = month, FFM = fat free mass

The effect of cardiorespiratory training on activity is presented in Table 4.4. After short-term intervention, improvement in mean activity ranged from 0 – 13%. However, only the improvement of 13% was statistically significant. After long-term intervention, both studies reported a statistically-non significant improvement in mean activity of 2 and 9%.

Table 4.4: Effect of cardiorespiratory training on activity

Study	Duration of intervention	Outcome measure	Mean difference between groups Exp minus Con	% change	Significance of the difference between groups
Short intervention					
Unnithan et al (2007)	3 mth	GMFM Dimensions D & E	4	13%	$p < 0.05$
Van den Berg-Emons et al (1998)	2 mth	Daily physical activity (ratio of daily energy expenditure over resting/sleeping energy expenditure in mJ/d)	-0.07	-6%	Not reported
Verschuren et al (2007)	4 mth	GMFM Dimension D	1	1%	95% CI = -3 to 5; $p > 0.05$
		GMFM Dimension E	0	0%	95% CI = -6 to 6; $p > 0.05$
Long intervention					
Van den Berg-Emons et al (1998)	9 mth	Daily physical activity (ratio of daily energy expenditure over resting/sleeping energy expenditure in mJ/d)	0.11	9%	$p > 0.05$
Verschuren et al (2007)	8 mth	GMFM Dimension D	3	3%	95% CI = 1 to 6; $p = 0.03$
		GMFM Dimension E	2	2%	95% CI = 0 to 5; $p = 0.27$

Exp = experimental group, Con = control group, RCT = randomised controlled trial, class = classification, PE = physical education, PT = physiotherapy; Int = intervention, freq = frequency, CV = cardiorespiratory, HRR = heart rate reserve, ex. = exercise, VO_{2peak} = ,peak oxygen uptake, TEE = total energy expenditure, RER = respiratory exchange ratio, RMR = resting metabolic rate; SMR = sleeping metabolic rate; CP = cerebral palsy, GMFCS = Gross Motor Function Classification System, GMFM = Gross Motor Function Measure; M = male, F = female, m = metre, min = minute, mth = month, mJ/d = mega joules per day.

DISCUSSION

This systematic review suggests that cardiorespiratory training may increase aerobic fitness in children with cerebral palsy. However, there is no evidence of carry over into activity. The strengths of this review are its comprehensive search strategy, lack of bias in selecting studies by blinding reviewers to journals, authors and outcomes, and the inclusion of only randomised (or quasi-randomised) trials (Autti-Ramo et al 2006, Hoving et al 2001, Oxman et al 1991). Although moderate to high quality studies were included in the review, the available evidence is not strong. This is not so much because of the small number of randomised trials of cardiorespiratory fitness in children with cerebral palsy, but because there was insufficient data reported in these trials to perform a meta-analysis. Our finding that cardiorespiratory training may increase aerobic fitness in children with cerebral palsy supports the conclusions of two previous reviews (Verschuren et al 2008, Rogers et al 2008). The present review strengthens these conclusions because the findings were based on randomised trials and also incorporated two recently published extra trials.

Clinical heterogeneity was noted across the studies in terms of participant characteristics. There was variability in the age range of participants between the studies and also in the level of disability reported. For example, there were only pre-adolescents in one study, adolescents in another and the full age range in the third. Likewise, there was variability in the level of disability with Levels I and II in one study, Levels II and III in another study and Levels I-IV in the third study. Participants' baseline fitness was approximately half their age expectation. While all participants showed some increase in their level of fitness, their percentage increase in fitness was less than would be expected for normal (Washington et al 1994).

These results are in line with Stout's (2006) observation, that children with cerebral palsy have a lower physical work capacity and higher oxygen cost associated with activity. The greatest increase in fitness was seen in those participants with Levels I and II. This is perhaps not surprising given that these participants were all ambulatory and have more potential to undertake training than participants with higher levels of disability.

Different measures of aerobic fitness were used across studies. However, all measures were either VO_{2peak} or peak aerobic power which have been shown to be valid measures of aerobic fitness for children (Armstrong 1994, Washington et al 1994). Therefore, given comparable measures of fitness albeit it with different units, a meta-analysis of the standardised mean difference between groups was planned. Unfortunately, insufficient data prevented this analysis from being performed.

In terms of intervention, the studies were clinically homogeneous. In all studies, the experimental intervention was cardiorespiratory training in addition to the normal physiotherapy regimen which did not include a fitness component. The intensity of the cardiorespiratory training of 60-75% maximum heart rate, or 70% heart rate reserve, for a duration of 12 or more weeks was adequate to produce a training effect (American College of Sports Medicine 2000). In addition, the recommendation for children of school age through to adolescence to undertake 20 to 30 minutes of moderate to vigorous activity 2 to 3 times a week (American College of Sports Medicine 2000) was achieved in all studies.

If it appears that fitness was improved, why then was there no carryover to activity?

This question can perhaps be answered by noting that in two studies (Unnithan et al 2007, Verschuren et al 2007), activity was measured using scales which reflect performance rather than fitness (eg, GMFM Dimensions D and E). However, where measures did reflect fitness (eg, energy expenditure during daily activity in Van den Berg-Emons et al 1998) there were only small increases of 6 to 9%, which were non-significant. The one study to show a significant increase in activity (13%) was the Unnithan et al (2007) study. However in this study, the children were extremely unfit prior to training (about half of normal) and therefore a 13% increase reflects little actual change. If assumptions are to be made regarding the carryover from fitness to activity, then measures of activity need to reflect fitness, eg, the 6-min Walk Test. The 6-min Walk Test is appropriate for children at Levels I and II (Maher et al 2008). However, a measure of activity that reflects aerobic fitness has yet to be identified for children with cerebral palsy at Levels III-V.

CONCLUSION

There is limited evidence about the effect of cardiorespiratory training in children with cerebral palsy. What evidence there is suggests that training may improve aerobic fitness, but that there is little carryover into activity. Despite some recent studies investigating physical activity in children with cerebral palsy, the patterns (types and overall amounts) of activity in these children are still largely unknown (Maher et al 2007).

CHAPTER 5

PHYSICAL ACTIVITY

DEFINITION

MEASUREMENT

TYPICALLY-DEVELOPING CHILDREN

CHILDREN WITH CEREBRAL PALSY

DISCUSSION

DEFINITION

Physical activity is traditionally defined as being any body movement produced by skeletal muscles which results in a substantial increase in energy expenditure (Caspersen et al 1985). Physical activity also encompasses any health-related behaviours or actions which enable an individual to carry out everyday actions and processes (Dollman et al 2009). Determining the components of physical activity generally involves identifying the duration, intensity and frequency of the specific activities (Dolman et al 2009).

Physical activity and physical fitness are often used interchangeably to describe body movements which result in increased energy expenditure. In addition, it is accepted that there is a close relation between these terms and that one may be dependent on the other in terms of the amount of energy expenditure which an individual is able to produce. However, the body movements produced with physical activity are generally considered to be more flexible, less structured and more unpredictable than physical fitness (Caspersen et al 1985). As such, the terms 'free-living' and 'habitual' are frequently used to describe an individual's physical activity in their everyday environment. Physical fitness refers to a set of attributes that people have, or achieve, that relates to the ability to perform physical activity (National Centre for Chronic Disease Prevention and Health Promotion 2000).

The World Health Organisation (WHO) classifies activity into domains using the International Classification of Functioning, Disability and Health (ICF) (2001) framework. These domains reflect both the performance and the capacity of the individual in relation to their habitual physical activity. Performance refers to an

individual being able to physically perform an activity if they choose to do so, but does not measure the activity in terms of duration or frequency. Capacity on the other hand has principles of frequency, duration, intensity and type of activity. Some authors would also suggest that adding 'purpose' to the measure of activity would be more meaningful, particularly in relation to physical activity seen in children (Dwyer et al 2009a; Dollman et al 2005, Kohl et al 1998).

It is well recognised that regular physical activity for adults and children is important for maintaining health and preventing disease and for providing benefits for self-esteem, social interaction and skill development (American College of Sports Medicine 2000). The promotion of regular physical activity and health-related cardiorespiratory fitness has also been associated with promoting longevity and reducing risks associated with adult diseases (Strong et al 2005). Furthermore, there is evidence that behavioural patterns of physical activity seen in childhood are likely to track into adulthood and therefore establishing regular physical activity habits in childhood will have long term benefits (Malina 2001).

Physical activity is also seen as a pre-requisite for optimal growth and development in children (Stout 2012). In particular, in children, that regular physical activity will lead to benefits in cognitive, cardiorespiratory and muscular development and the attainment of age appropriate motor skills.

MEASUREMENT

In order to measure physical activity it is necessary to quantify the parameters being measured. These parameters include duration, frequency, and intensity of the

activity (Dolman et al 2009). The duration of activity is measured in units of time and may also take into account periods of inactivity or sedentary behaviour if those periods comprise part of the original activity. Frequency is usually measured in 'bouts' of activity. These bouts of activity may include the number of sessions of the activity, or periods of time, such as days. The level of intensity of activity is commonly measured as energy expenditure and is expressed as metabolic equivalent multiples of resting metabolic rate or METS (Dolman et al 2009).

METs are defined as being the amount of energy an individual would expend during a period of rest. One MET is the amount of energy expended sitting quietly at rest and is adjusted to body weight, ie, 1 MET = 3.5 ml oxygen consumed per kilogram of body weight per minute (Dolman et al 2009). 1 MET is also described as being sedentary activity, with 1.5 and 3 METs being defined as light activity, ie, one and half, to three times, the energy expended compared to sitting quietly at rest (Dolman et al 2009). Light activity might equate with general physical movement within an individual's home or work environment, or walking at a slow pace. Moderate levels of activity are expressed as being between 3-6 METs, eg, walking at a 14 minute pace per kilometre would be expressed at an intensity of 6 METs, or 6 times the energy sitting quietly at rest (Dolman et al 2009). Vigorous level of activity are expressed as being >6 METs (Dolman et al 2009).

The duration, frequency, and intensity of physical activity may be determined by using criterion validity, objective and subjective measures (Sirard and Pate 2001, Trost 2007). Criterion validity measures are seen as the 'gold standard' measures as they give the most reliable information in regards to the relationship between activity

and energy expenditure (Sirard & Pate 2001). These measures commonly include direct observation and indirect calorimetry using the doubly labelled water method (Sirard & Pate 2001).

Direct observation of physical activity can provide relevant and comprehensive information regarding the duration and frequency of an individual's physical activity and also, to some extent, the intensity of the activity as judged subjectively by an individual's response to exertion. However, to give meaningful information, direct observation needs to be conducted over repeated time periods to ascertain an individual's usual physical activity patterns, and also to rule out potential bias of subject response to observation (Sirard & Pate 2001). A major drawback of direct observation is obviously the labour intensity and time required to conduct this method.

The doubly labelled water method originated from a study by Lifson et al (1949) and has been used extensively to measure energy expenditure in free-living humans (Schoeller 1999). Doubly labelled water, is water in which both the hydrogen and the oxygen has been partly, or completely, replaced in order for it to be traced, ie, labelled. This is done by administering a dose of hydrogen and oxygen isotopes, known as 'heavy water' and then measuring the elimination rates, in the subject, over time (Schoeller 1999). The method starts with a urine sample from subjects who are then given a single dose of 'heavy water'. Following the administration of the dose, urine is collected during the observation period, usually within the first 6 hours to determine total body water. The urine voided 24 hours later marks the beginning of the measured energy expenditure period (Schoeller 1999). Measurement of the

energy expenditure period ends after 7 to 21 days when a final urine sample is collected. Between the initial and final samples, the subject is free to engage in normal activities. A second dose of doubly labelled water is administered at the end of the study period and urine collected after 3 to 6 hours for a second determination of total body water. This second determination of total body water is used to measure any changes in the total body water pool during the observation period (Schoeller 1999). The doubly labelled water method has high validity as a measurement of energy expenditure. However, costs and availability of this method restrict it to primarily being used in laboratory settings.

Less rigorous, but still objective, measures of physical activity include heart rate monitors, pedometers and accelerometers. These devices have become popular in studies measuring free-living activities and can be used for both individuals and groups. Each of these devices is capable of reducing the subjectivity of measures such as surveys and self-reports, but may be unable to specifically estimate physical activity in an individual. For example, heart rate monitoring may be used to measure intensity of activity as there is a known association between heart rate and oxygen consumption, particularly when performing a more vigorous activity (Armstrong & Welsman 1994). However, heart rate is also known to be affected by other factors, such as stress, heat and caffeine. Therefore, heart rate monitoring may be valid to determine more vigorous physical activities where the heart rate is likely to be higher than140 beats per minute but may not be specific enough for heart rates lower than this (Armstrong & Welsman 1994).

Motion sensors, such as pedometers, provide information relating primarily to ambulation. 'Simple' pedometers estimate the distance covered when walking, or the number of steps taken over a period of time. Although this information may be useful in some circumstances, for example to determine if an individual is performing any upright activity, it is limited in that it does not detect differences in rate of walking, and thereby energy expenditure (Rowlands et al 1997). Specifically, energy expenditure would be underestimated when an individual is running, compared to walking, as fewer steps would be recorded for a set distance (Rowlands et al 1997).

More sophisticated pedometers have been developed in an endeavour to record both steps and energy expenditure during ambulation. The StepWatch Activity Monitor (SAM)® is a combined accelerometer and pedometer which is worn around the ankle (Coleman et al 1999). This monitor records both the frequency of steps taken as well as the number of steps at 1-minute intervals and can be worn for up to 4 weeks while continuously recording data. This monitor has been used in several studies of children with cerebral palsy to investigate physical activity patterns (Bjornson et al 2007, Stevens et al 2010, Van Wiley et al 2010). Findings from these studies look promising in terms of using the SAM as a measure of physical activity. However, discrepancies noted in other studies (Tudor-Locke et al 2004, Song et al 2006), in terms of recommended steps to reflect moderate to vigorous activity, indicate that further comparative studies are required.

Accelerometers have become widely used as a means of assessing the pattern, intensity, and total accumulation of activity (Clanchy et al 2011). Accelerometers

are motion sensors which measure physical activity and energy expenditure via accelerations of the body. Multiple planes of movement can be captured with more recent versions incorporating more planes, or axis, of movement. Accelerometers have evolved from being uni-axial sensors (measuring acceleration in a single, vertical, plane) to tri-axial sensors which measure acceleration in 3 planes (vertical, antero-postero, and medio-lateral) (de Vries et al 2009). In addition, many accelerometers have the capacity to assess physical activity and store data over a 4 week period (Sirard & Pate 2001).

Acceleration of the body is measured by sensors in the accelerometer which convert body movement into electrical signals, or counts (Sirard & Pate 2001). These counts are considered to be proportional to the muscular force producing the movement (Sirard & Pate 2001). The counts recorded by the accelerometer, are summed and stored over a specific period of time. This time period is referred to as an 'epoch' and may be adjusted depending on how much data is required to be collected (Sirard & Pate 2001).

In order to relate METs to activity counts, prediction equations have been developed for some accelerometers (Trost et al 2005, Freedson et al 1998; Puyau et al 2004; Evenson et al 2008). From these equations, 'cut-points' of activity counts have been proposed to relate to sedentary, light, moderate and vigorous levels of intensity of physical activity (Sirard & Pate 2001, Freedson et al 1998, Nichols et al 2000, Brage et al 2003, Hendelman et al 2000, Leeanders et al 2003). Table 5.1 shows activity counts for adults using the Actigraph® accelerometer.

Table 5.1: Activity counts for adults using the Actigraph® accelerometer.

Study	Age (yr)	Activity	Activity counts			
			Sedentary	Light	Moderate	Vigorous
Freedson et al (1998)	18-30	Walking/running	< 100	0-1951	1952	5725
Nichols et al(2000)	18-35	Walking/running	< 100	0-1576	3285	5677
Brage et al (2003)	23-30	Walking/running	< 100	0-1809	1810	5850
Hendelman et al (2000)	30-50	Walking/running	< 100	0-2190	2191	6893
Leeanders et al (2003)	18-30	Walking/running	< 100	0-1266	1267	6252

yr = year

Finally, subjective measures of physical activity vary considerably but may include interviews, self or proxy-report, questionnaires or activity logs and diaries (Trost 2007). Both objective and subjective measures tend to be the measurement of choice to determine an individual's free-living physical activity and are commonly known as 'field' tests'. Some measures of physical activity are quite difficult to achieve outside of laboratory conditions and therefore tend to be used more for research studies. Generally criterion validity measures fall into this category. Field studies on the other hand are more likely to include a combination of more portable objective and subjective measures.

TYPICALLY-DEVELOPING CHILDREN

Healthy young children differ to adults in that they tend to be relatively active without needing to engage in formal, or structured, physical activities. Active play tends to be the principal means of physical activity in young children being replaced with more formal exercise and recreational pursuits into adulthood (Dwyer et al

2009). Activity in children is generally conducted in short bursts rather than prolonged episodes (American College of Sports Medicine 2000). Bursts of activity tend to be intermittent and range from a few seconds to several minutes and may be interspersed with frequent rest periods. In addition, children are more likely to display frequent changes in activity choices, with play being the prime determinant of activity. In terms of promoting and maintaining physical health, for children aged 5-18 years, activity should ideally entail a cumulative amount of at least 60 minutes of moderate to vigorous intensity each day (Department of Health and Ageing 2005).

It is recognised that the parameters of duration, intensity and frequency of typical free-living activity in children is different from adults. As a result, there is a growing body of literature pertaining to measurement of physical activity in school-aged to adolescent children in an effort to identify their current levels and patterns of activity (Rowlands et al 1997, Coleman et al 1999, Sirard & Pate 2001, Trost et al 2005, Freedson et al 1998, Puyau et al 2004, Evenson et al 2008, Guinhouya et al 2009, Mattocks et al 2007, McClain et al 2008, Treuth et al 2004). From this literature, it is evident that the same principles for measurement of physical activity in adults are also applicable for children but with some modifications.

Measures of direct observation of physical activity are easily done with either individual or small groups of children. However, measures such as doubly-labelled water are less well tolerated by children. A combination of heart rate monitors, pedometers and accelerometers have been used in several studies and are seen to be valid measures of physical activity in children (Evenson et al 2008, Trost et al 2005, Mattocks et al 2007, McClain et al 2008, de Vries et al 2009). However, when using

heart rate as an indicator of physical activity in children, it is important to recognise that they are unlikely to achieve a heart rate of greater than the 140 bpm seen as necessary to indicate vigorous physical activity (Rowlands et al 1997).

Accelerometers are being used increasingly to measure physical activity in children using either structured or free living activities. The duration of investigated physical activity is quite variable and ranges from 15 minutes to 7 days. Shorter studies seem to investigate structured activities over a 15-30 minute period and often include a number of 5-7 minute activities within that time frame (Evenson et al 2008, Freedson et al 2005, Trost et al 2005, Mattocks et al 2007a, Puyau et al 2002, McClain et al 2008a). Whereas, longer studies have investigated the child's free living activities with a range of 1 hour to 7 days (Guinhouya et al 2009, Trost 2002, Treuth et al 2004). Table 5.2 shows activity counts for typically-developing children using the Actigraph® accelerometer.

The majority of the studies presented in Table 5.2 report similar cut-points for intensity of sedentary activity (≤ 100 counts) and light activity (>100 counts). However, one study (Puyau et al 2002) reports markedly higher cut-points for the same levels of intensity (≤ 800 counts for sedentary and 801-3200 counts for light activity). Interestingly, there are a variety of different cut-points across the studies reported for moderate and vigorous levels of intensity. These cut-points range from >2060 counts (McClain et al 2008) to 8200 (Puyau et al 2002) for moderate activity, and ≥ 4012 (Evenson et al 2008b) to >8200 (Puyau et al 2002) for vigorous activity. The study by Puyau et al (2002) again reports the higher cut-points for both of these

levels. Two studies (Guinhouya et al 2009, McClain et al 2008) combined moderate and vigorous activity into one category.

Table 5.2: Activity counts for typically-developing children using the Actigraph® accelerometer.

	Duration of Activity	Age (yr)	Epoch	Activity counts			
Free-living activities				**Sedentary**	**Light**	**Moderate**	**Vigorous**
Guinhouya et al (2009)	14hr x 3 days	8-11	1 min			≥ 3400	
Trost et al (2002)	12hr x 7 days	6-18	1 min	≤ 100	> 100	≥ 3000 - 5900	≥ 6000
Treuth et al (2004)	12hr 3 days	13–14	30 sec	≤ 100	> 100	≥ 3000	≥ 5200
Structured activities							
Evenson et al (2008)	63min (9 x 7min)	5-8	ROC	≤ 100	> 100	> 2296	≥ 4012
Freedson et al (2005)	Progressive treadmill exercise test to voluntary exhaustion	6-18	1 min	≤ 100	> 100	≥ 2220	≥ 4136
Trost et al (2005)	15min (3x5min)	10-18	1min	≤ 100	> 100	2945	4443
Mattocks et al (2007b)	30min (6 x 5min)	12	1 min	≤ 100	> 100	≥ 3581	≥ 6130
Puyau et al (2002)	6hr	6-16	1 min	0 - 800	801 - 3200	3201 - 8200	> 8200
McClain et al (2008)	30min	10	1 min	≤ 100	> 100	> 2060	

Min = minutes; hr = hour; yr = year; sec = seconds; ROC = receiver operating characteristic

CHILDREN WITH CEREBRAL PALSY

Children with cerebral palsy have been noted to be less active and more sedentary than their typically developing peers (Maher et al 2008). It is also noted that the activity limitations associated with cerebral palsy tend to result in a less than optimal self-reported quality of life (Bjornson et al 2008). The influence of musculoskeletal impairment particularly, is seen to be a significantly limiting factor to activity in children with cerebral palsy.

Therefore, children with cerebral palsy are less likely to meet the daily recommended guidelines for 60 minutes of moderate to vigorous intensity physical activity. However, determining the intensity, frequency and duration of physical activity required for improving the level of moderate to vigorous activity for children with cerebral palsy, remains largely unknown (Rimmer 2001). Furthermore, it is unknown whether children with cerebral palsy need to be engaged in the same levels of intensity and duration in physical activity as typically developing children in order to meet the expected daily levels.

Two studies have used accelerometers to investigate the level of physical activity of children with cerebral palsy (Capio et al 201,; Clanchy et al 2011). Both studies used the Actigraph® accelerometer to determine whether this accelerometer was valid to use in children with cerebral palsy. In addition, Clanchy et al (2011) investigated whether this accelerometer was able to differentiate between different intensities of activity and also whether the cut-points determined for typically developing children were valid for children with cerebral palsy.

The age of the children in both studies ranged from 6-16 years and measured timed walking trials and structured activities, as well as 'free play' activities. One study (Clanchy et al 2011) used 1 minute epochs to record the frequency of activity while the other study (Capio et al 2010) used 15 second epochs and converted these to 1 minute equivalents. The period of activity measured in both these studies was relatively short, ie, a total of 15 minutes in the Clanchy et al (2011) study, and 22 minutes total in the Capio et al (2010) study.

Both studies reported similar cut-points for moderate intensity levels of physical activity with Clanchy et al (2011) reporting ≥2400 counts and Capio et al (2010) reporting ≥2000 counts. Clanchy et al (2011) also reported on vigorous intensity levels of activity (≥4000 counts). These studies show similar results for moderate levels of intensity of activity when compared to typically-developing children. However overall, it is unknown what the level of intensity for vigorous physical activity would be in children with cerebral palsy.

Currently, there are few studies investigating the validity of accelerometry as a measure of physical activity in children with cerebral palsy. However, the preliminary studies using the Actigraph® accelerometer indicate that it may be a valid measure of physical activity in ambulatory children with cerebral palsy. In particular, that this accelerometer is likely to be appropriate for estimating time spent in moderate to vigorous physical activity (Clanchy et al 2011). Table 5.3 shows activity counts for children with cerebral palsy using the Actigraph® accelerometer.

Table 5.3: Activity counts for children with cerebral palsy using the Actigraph® accelerometer.

Study	Age	Epoch	Sed	Light	Mod	Vig	Activity measured
Clanchy et al (2011)	8-16yr	1min	≤100	>100	≥2400	≥4000	3 walking trials – 2x6min, 1x3min
Capio et al (2010)	6-14yr	15sec			≥2000	≥2000	6x 2 min structured activities and 1x 10min free play activities collected in school setting

Subjective measures of physical activity have been collected on children with cerebral palsy via self-report surveys. One study (Maher et al 2007), collected data on physical activity in adolescents and reported that physical activity was related to both age and level of ability. Overall, the adolescents in this study were less physically active over the observed week than their typically-developing peers. From this study, Maher et al (2007) also suggest that children with cerebral palsy have considerably different physical and sedentary behaviours than their typically developing peers. They are not only less active, but also tend to have higher periods of sedentary behaviour each day and participate in a smaller variety of physical activities. In addition, the choice of activity seems to be one which can be performed at a slower speed (Maher et al 2007).

DISCUSSION

There is growing recognition that children with cerebral palsy need to be similarly physically active as typically developing children (Bjornson et al 2008, Damiano 2006, Maher et al 2007, Murphy et al 2008). What is not known at this stage is, what the intensity, frequency and duration of physical activity needs to be in order to

improve physical activity in children with cerebral palsy. In particular, in order to gain health benefits from physical activity, what intensity of activity is required to specifically achieve levels of moderate to vigorous activity. Therefore, the challenge is to ensure that children with cerebral palsy are enabled to participate in physical activity which is not only enjoyable and achievable, but also of sufficient intensity to achieve physiological benefits (Maher et al 2007).

Overall, it would seem that further investigation of the intensity of physical activity in children with cerebral palsy is needed. This investigation is also needed, as there is increasing evidence that ongoing physical inactivity in these children may be contributing to their decreased levels of cardiorespiratory fitness compared to their typically developing peers. However, what is still largely unknown is whether these decreased levels of cardiorespiratory fitness are related to the amount and intensity of free-living physical activity. Therefore, the study in Chapter 6 investigates the relationship between cardiorespiratory fitness and the amount of physical activity in children with cerebral palsy who are independently walking.

CHAPTER 6

PHYSICAL ACTIVITY, CARDIORESPIRATORY FITNESS AND WALKING CAPACITY IN CHILDREN WITH CEREBRAL PALSY: A FEASIBILITY STUDY

INTRODUCTION

It is known that children with cerebral palsy have decreased levels of cardiorespiratory fitness compared to their typically-developing peers (van den Berg-Emons et al 1995, Fernhill and Unnithan 2002, Hoofwijk et al 1995, Unnithan et al 1996, Rose et al 1989). It is also known that decreased cardiorespiratory fitness in these children contributes to ongoing physical inactivity due to reduced stamina and aerobic capacity. Of concern, is the knowledge that these factors are likely to be discouraging, or limiting, participation in community activities (Bjornson et al 2006, 2007, Rimmer 2001, Rimmer et al 2004; Pirpiris et al 2004; van den Berg-Emons et al 1995).

Previous studies have shown that it is possible to increase cardiorespiratory fitness in children with CP (Unnithan et al 2007, van den Berg-Emons et al 1998, Verschuren et al 2007). Unfortunately, the intervention used in these studies did not lead to an improvement in physical activity. However in these studies, this result may be attributed to the outcome measures used to measure activity rather than a true reflection of the intervention.

One study investigated differences in cardiorespiratory endurance and walking capacity in children with cerebral palsy following a 9 week training program (Gorter et al 2009). In this study, a modified Bruce treadmill test was used to determine cardiorespiratory endurance with heart rate and time on the treadmill used as the outcome measures. However, maximal oxygen uptake was not measured as it was felt that an assumed relationship between heart rate and oxygen uptake could be taken as a proxy of the energy expenditure (Gorter et al 2009). Walking capacity

was determined using a 6-minute run test. As a result of this training program, it was reported that children had improved walking capacity in their home environment.

Another study investigated the relationship between the oxygen cost of walking and physical activity in children with cerebral palsy (Maltais et al 2005). Direct measures of VO_{2peak} were made using gas analysis during a progressive exercise test on a treadmill. Walking capacity was measured using the Gross Motor Function Measure (GMFM) – Domain E walking, running and jumping (Palisano et al 1997). The main findings were that there was no relationship between the oxygen cost of walking and physical activity. Although a direct measure of oxygen uptake was made in this study, walking capacity was not directly measured, ie, the GMFM reflects walking performance, along with other upright activities, but does not measure capacity directly.

Overall, despite physical training to improve cardiorespiratory fitness, children with cerebral palsy do not appear to use their physical reserve sufficiently to achieve optimal levels of daily physical activity (Gorter et al 2009). There are many children with cerebral palsy who are able to walk independently (Level I or II on the Gross Motor Function Classification Scale -GMFCS) and attend mainstream schools. Of interest would be to know whether these children are as physically active within their school and home environments as their typically-developing peers. In addition, if they are seen to be less active, could this be attributed to a decrease in their level of cardiorespiratory fitness?

One of the main drawbacks of many studies is that direct measures of both cardiorespiratory fitness and physical activity are not made in the same investigation. Often assumptions are made regarding a linear relationship between parameters, or indirect measures are made. In addition, to date there have been no studies which have investigated whether decreased levels of cardiorespiratory fitness in children with cerebral palsy are seen to affect the amount and intensity of moderate to vigorous physical activity.

The aim of this study was to examine the feasibility of investigating physical activity, cardiorespiratory fitness and walking capacity in independently walking children with cerebral palsy in order to answer the following research questions:

1. What is the amount and intensity (level) of their physical activity compared with their typically-developing siblings?
2. What is the relationship between cardiorespiratory fitness and amount of physical activity in these children? and
3. What is the relationship between their walking capacity and amount of their physical activity?

The hypothesis was that the procedures for measuring physical activity and cardiorespiratory fitness will be feasible in these children. Furthermore, the amount of physical activity undertaken by these children could be explained by their cardiorespiratory fitness and/or their walking capacity.

METHODS

Design

This was a descriptive study measuring physical activity, cardiorespiratory fitness and walking capacity in children with cerebral palsy. If participants had siblings who were aged between 8-12 years, they were also invited to participate in the study and have their physical activity measured.

Testing was conducted at the beginning and end of a 7 day period at either one of the NSW Spastic Centre/CP Alliance sites in the Sydney metropolitan area, or at the Lidcombe Campus of the University of Sydney. On the first day of testing, the participants with cerebral palsy had anthropometric measures (height and weight) and measures of impairment taken (range of joint motion, spasticity, strength and coordination). They then performed a cardiorespiratory fitness test of maximal oxygen consumption ($VO_{2peak)}$ while on a treadmill. Participants were then fitted with an accelerometer to wear for the following 7 days and given instructions for completing an activity record book for 4 of the 7 days.

On the second day of testing 7 days later (Day 8), participants performed a test of walking capacity. Participants and their caregivers were thanked for their involvement in the study and each participant was given a certificate of appreciation and a movie voucher at the conclusion of the testing.

Siblings had anthropometric measures (height and weight) taken on Day 1 and were fitted with an accelerometer to wear for the following 7 days. They were also given instructions for completing an activity record book for 4 of the 7 days.

Ethics approval was gained through The University of Sydney and The Spastic Centre of New South Wales (now known as the Cerebral Palsy Alliance) Ethics Committees to recruit participants for the study. Physiotherapists from The Spastic Centre of New South Wales invited children and their families registered with that organisation to participate in the study (Appendix C). A verbal and written explanation of the study was given to the parents of the participants (Appendix D). All data pertaining to the participants and peers was recorded on a Data Collection Sheet (Appendix E and F). The design and flow of participants through the study is presented in Figure 6.1.

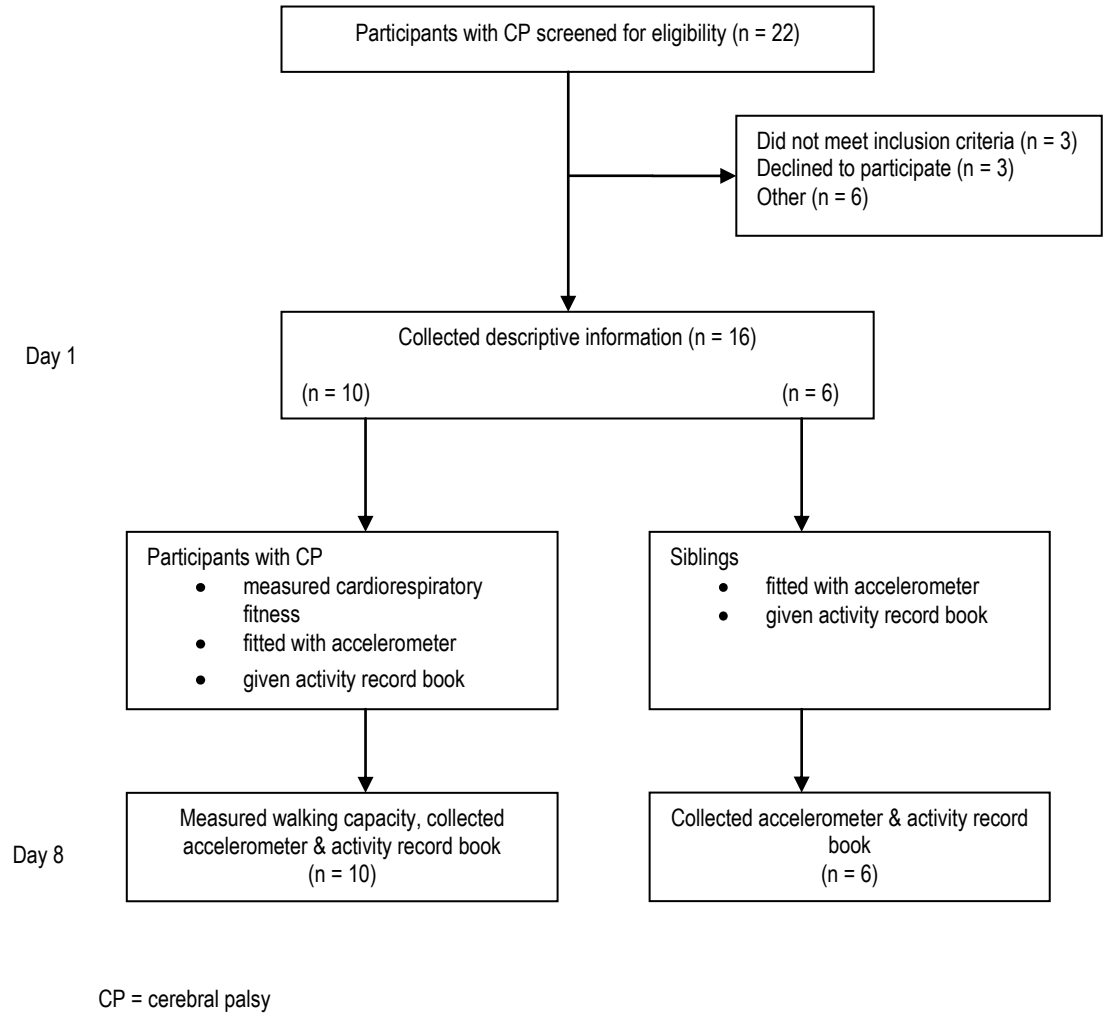

Figure 6.1: *Design and flow of participants through the study.*

Participants

Parents were invited to participate if their child had any classification of cerebral palsy, were aged between 8 and 12 years and had a Gross Motor Function Classification Score (GMFCS) of Level I or II, ie, they could walk independently. Potential participants were excluded if they had orthopaedic surgery or neurosurgery or Botulinum Toxin A intervention in the preceding 4 months, had cardiac or respiratory conditions which precluded them from exercising at high intensity or had severe cognitive and/or language deficits which precluded them from participation in

the exercise program. The latter exclusion category was made by clinical judgement. See Figure 6.2 for inclusion and exclusion criteria.

Inclusion criteria:

- Cerebral palsy Classification: hemiplegia, diplegia, other
- 8 - 12 years
- GMFCS Level I or II/can walk independently

Exclusion criteria:

- orthopaedic surgery or neurosurgery or Botulinum Toxin A intervention in the preceding 6 months
- cardiac or respiratory conditions which preclude from exercising at high intensity
- severe cognitive and/or language deficits which preclude participation in the exercise program (clinical judgement)

Figure 6.2: *Inclusion and exclusion criteria for the study.*

In addition, siblings within the 8-12 years age range were also invited to participate in the study to wear the accelerometer for 7 days and complete an activity record book.

Upon agreeing to participate, parents were given an Information Sheet (Appendix G) to keep and a Consent Form (Appendix H) to sign. In accordance with standard treadmill testing procedures, caregivers were instructed that the participant should not undertake any increased level of activity outside of their usual routine 24 hours before the day of the test. They should maintain their usual diet, but refrain from eating food 2-3 hours prior to testing. The participants were instructed to wear comfortable exercise type clothing and their usual walking or sports type shoes.

In order to describe the characteristics of the participants, anthropometric measures (height and weight), measures of impairment (contracture, spasticity, strength and coordination), classification of cerebral palsy, year and type of school attended were recorded. Only anthropometric measures of height and weight were recorded for peers.

Participants were weighed on standard scales (Seca, Hamburg Germany) and their weight recorded in kilograms. Height was recorded in metres with the participant standing against a wall, a spirit level ruler placed on top of their head and height measured with a standard tape measure (Stanley Powerlock®) to the nearest centimetre. Body mass index was calculated by dividing the participant's weight in kilograms (kg) by their height in metres squared (m^2) (National Centre for Chronic Disease Prevention and Health Promotion 2000).

Information was collected about impairment of the lower limbs that included contracture, spasticity, strength and coordination. Contracture was determined by measuring the passive joint range of the ankle with the participant positioned in supine with their knee extended. The participant was instructed to relax while the examiner pushed their ankle into dorsiflexion until resistance prevented any further movement. The observed angle was then recorded as either being <90°(score 1), =90° (score 2) or >90° (score 3) of ankle dorsiflexion for both left and right sides.

Because this study population may have had both spasticity and contracture of the ankle plantarflexors, the Tardieu Scale (Tardieu et al 1954; 1957) was used to determine spasticity. This scale was used as it is more likely to be able to

differentiate between spasticity and contracture (Patrick & Ada 2006). The CP participant was positioned in supine with their head in the midline and arms folded lightly across their chest. The participant was then instructed that they should relax while the examiner pushed their foot in dorsiflexion, first slowly (V1) and then quickly (V3). Both left and right ankles were tested and the quality of the muscle reaction to stretch was then graded on a scale from 0-4, with 4 representing normal. See Appendix I for the grading of the Tardieu Scale.

Strength of the ankle dorsiflexors was determined using a manual muscle test with the knee extended. The participant was positioned supine on a raised bed/plinth. They were then instructed to keep their knee straight and to pull their foot up towards their knee. This action was demonstrated first by the tester. The participant was instructed not to let the tester pull their foot back down out of this position. Both left and right ankles were tested and the strength of the dorsiflexors was then graded on a scale from 0-5, with 5 representing normal.

Coordination can be determined by evaluating an individual's performance during the execution of an accurate, fast and repeated movement (Desrosiers et al, 2005). Desrosiers et al (2005) have developed a lower-extremity motor coordination test, the Lower Extremity MOtor COordination Test (LEMOCOT) which has been found to be a valid and reliable outcome measure of motor coordination. Therefore, the LEMOCOT was deemed appropriate to be used to measure lower limb coordination in this study population. This test requires the participant to move the lower extremity as fast as possible from one target to another for 15 seconds. Targets are

6cm in diameter and positioned apart on either a thin piece of rigid foam, or non-slip matting, measuring 50×55cm. Figure 6.3 illustrates the LEMOCOT testing set-up.

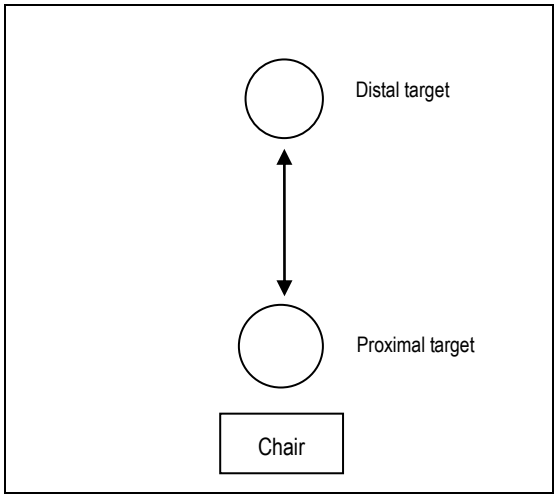

Figure 6.3: *LEMOCOT testing set-up (Desrosiers et al, 2005) (not drawn to scale).*

The matting was placed on the floor in front of the chair used for the test. Participants were seated on a regular chair which had back support and was appropriate for their height. Participants performed the test with bare feet. Before starting the test, participants were required to place one heel on the proximal target while maintaining the knee flexed at 90º. To start the test, the big toe was placed on the proximal target. At the examiner's signal, participants were required to alternatively touch the proximal and distal target with their big toe as fast, and as accurately, as possible in a 15-second period. The examiner counted the number of times the participant touched both targets with their big toe during the 15-second period. If a target was not touched by the participant's big toe, the target was not counted. Participants were told that the aim of the test was to touch each target accurately and not necessarily to go as fast as they could if it meant missing the target. Testing began with the unaffected lower extremity for five to ten seconds to

ensure the participant understood the procedure. The number of taps recorded over the 15-second period was then multiplied by 4 to get taps per minute for analysis.

Outcome measures

Feasibility

Feasibility of this study was measured as participant recruitment and retention, suitability of measurement procedures and participant compliance with the measurement procedures.

Physical Activity

Direct measures of physical activity were made using a uni-axial Actigraph® WAM 7164 accelerometer (Pensacola, FL). Accelerometers are motion sensors which measure physical activity and energy expenditure via accelerations of the body. The Actigraph® accelerometer was chosen for the study as previous studies (Capio et al 2010, Clanchy et al 2011) have found this accelerometer to be reliable and valid for measuring physical activity in children with cerebral palsy. In addition, this accelerometer has been deemed appropriate for estimating time spent in moderate to vigorous physical activity in these children (Clanchy et al 2011). One minute epochs were chosen in line with previous studies (Capio et al 2010, Clanchy et al 2011) to 'capture' the physical activity counts and also to enable comparison with data collected from typically-developing children (Freedson et al 2005, Mattocks et al 2007a, McClain et al 2008b, Treuth et al 2004, Trost et al 2002, Trost et al 2007, Evenson et al 2008, Guinhouya et al 2009, Puyau et al 2004).

Participants were fitted with a uni-axial Actigraph® WAM 7164 accelerometer (Pensacola, FL). Prior to fitting, these instruments had been initialised to collect data in 1 minute epochs. Each participant wore the accelerometer on a fitted elasticised belt which was worn around the waist and positioned over the right hip (Figure 6.4)

Figure 6.4: *Positioning of the Actigraph® accelerometer on participants.*

The participants were asked to put the accelerometer on each morning on waking and remove at the end of the day before going to bed. Participants were instructed not to get the accelerometer wet and to remove it for bathing, showering or swimming. Participants were also given an instruction sheet for wearing the accelerometer (Appendix J) and were asked to wear the accelerometer for 7 days.

In order to compare the data obtained from the accelerometer with the child's actual activity each participant and their sibling were asked to complete an Activity Record Book, which was devised for this study, for 4 days (Appendix K). The designated 4 days for recording activity in the book were standardised for each participant to Friday, Saturday, Sunday and Monday during the testing week. Siblings were also

fitted with their Actigraph® accelerometer and given the same instructions and Activity Record Book to complete.

Cardiorespiratory fitness

A direct measure of maximal oxygen uptake (VO_{2peak}) to reflect cardiorespiratory fitness was measured by a portable gas analysis system (Cosmed K4b2™) while CP participants performed a progressive exercise test on a treadmill. An exercise physiologist from The University of Sydney assisted with the progressive exercise test and performed the gas analysis. A progressive exercise test was performed on all participants using a modified treadmill protocol developed by Verschuren et al (2006) for children with cerebral palsy. Two protocols were used for this test: one protocol for participants who were classified at Level I on the GMFCS and one protocol for participants who were classified as Level II on the GMFCS. The starting speed for GMFCS Level I was 5 km/hr and the starting speed for GMFCS Level II was 2 km/hr. The speed was increased by 0.25km/hr for both groups every minute until the cessation of the test. In line with the protocol by Verschuren et al (2006), the incline of the treadmill was set at 2% to compensate for lack of air resistance. This incline is more likely to reflect results which would be obtained for outdoor running.

All participants were given a 3-minute practice session on the treadmill at a speed of 2 km/hr to familiarise themselves with the equipment. They were then given a 5-minute rest prior to starting the testing procedure. Following familiarisation with the treadmill, participants had their pre-test heart rate measured with a Smartsigns® MiniPulse (Huntleigh Healthcare) pulse oximeter and were fitted with a portable gas

analysis system (Cosmed K4b2™ device). This system consists of a facemask, a transmitting unit (containing different oxygen and carbon dioxide analysers), and a receiving unit. The transmitting unit with facemask and tubing (total weight = 0.57kg) was attached to participants via a head strap. The receiving unit was connected to a laptop computer located within 1.0m of the transmitting unit.

During the exercise test, two people stood alongside the treadmill on the same side as the emergency stop button. Participants were also instructed on the use of the emergency button prior to starting the test and were encouraged to use the button at any stage of the test if they felt concerned. Participants were given verbal encouragement throughout the test to walk for as long as they could (volitional fatigue.) The test was terminated when the participant indicated that they were either unable to continue (either verbally or through a pre-determined sign), refused to continue or exhibited signs of distress beyond what would be expected for this test.

The post-test heart rate was again measured using the pulse oximeter and the participant given a drink of water and instructed to sit and rest.

Walking capacity

In order to measure walking capacity, participants performed a 6-minute walk test (6-MWT) (American Thoracic Society 2002). The 6-MWT is a sub-maximal test of exercise capacity which was primarily developed for adults with cardiorespiratory disorders. This test measures the distance that a person can walk in 6 minutes, at a brisk pace, over a level surface. Use of the 6-MWT has been broadened to include other age populations, ie, children (Li et al 2005, Geiger et al 2007, Geiger et al

2011, Lammers et al 2008, Lammers et al 2011, Limsuwan et al 2010) and health disorders such as cerebral palsy (Maher et al 2008, Thompson et al 2008, Beard et al 2005, Mulligan et al 2004) (Table 6.1). The 6-MWT was chosen for this study as it was found to have high test-retest reliability in ambulant children with cerebral palsy (Maher et al 2008, Thompson et al 2008).

Table 6.1: The 6-Minute Walk Test in typically-developing children and children with cerebral palsy.

Study	Age (yr)	Distance (m)
Typically -developing		
Li et al (2005)	13-15	659 (58)
Geiger et al (2007)	3-18	621 (47)
Lammers et al (2008)	4-11	497 (145)
Cerebral palsy		
Maher et al (2008)	11-17	449 (97)
Thompson et al (2008)	4-18	334 (145)

Yr = year; m = metre

The 6-MWT was administered according to the American Thoracic Society (2002) guidelines. However, on some occasions the course length was adapted from the 30 metres recommended by the guidelines, to a 10 metre course. This change was necessary due to limited space in some test settings. It was not felt that the 6-MWT was being compromised as a result of this change as previous studies had also used variable distances and reported that reliability was not affected (Maher et al 2008, Thompson et al 2008).

For each test, the course was marked out using a metre wheel. Plastic cones were placed at each end of the course to indicate the turning point. The half-way point of the course was marked with red masking tape. A timer was set to count down for 6

minutes and each lap was recorded using a lap counter. Standardised instructions and encouragement were given in accordance with the test guidelines by the one assessor.

The participants wore the same Actigraph® accelerometer which they had been wearing for the previous 7 days while performing the test. Pre and post-test heart rate was recorded using a pulse oximeter. The accelerometer was removed after completion of the test and the participant instructed to sit and rest for 5 minutes. The accelerometer of the siblings was also removed at this time and both completed Activity Record Books were collected.

Data analysis

A convenience sample of 10 children with cerebral palsy who were able to walk independently, and their siblings, was collected in order to provide information to conduct a power analysis for a future study. Descriptive characteristics of the participants (anthropometric measures, measures of impairment, classification of cerebral palsy, and type of school attended) are expressed as mean (SD) unless otherwise indicated.

Analysis of data from the Actigraph® accelerometer was performed by using software provided by the Actigraph® company for the WAM 7164 accelerometer (Pensacola, FL). The cut-points set for physical activity were determined *a priori* and were considered as sedentary activity for ≤ 100 counts, light activity > 100 and < 2000 counts, combined moderate to vigorous activity ≥ 2000 counts. The counts for

moderate to vigorous physical activity are lower than for typically developing children (Trost et al 2005, Freedson et al 1998, Freedson et al 2005, Puyau2004, Evenson et al 2008) but were considered to be reasonable and in line with cut-points reported by Clanchy et al (2011) and Capio et al (2010) for children with cerebral palsy. Participants were asked to put the accelerometer on each morning on waking and remove at the end of the day before going to bed. However, the amount of 'wear time' varied between participants. On visual examination, a window of 12 hours between 8 am and 8 pm was available across participants. If data was not available for 80% (9.6 hr) of the 12 hour period, this was considered to be a non-wearing day and reported as ND (no data). This is in line with other studies (Guinhouya et al 2009; Trost et al 2002). Actigraph® data were collected over intervals of 1 minute epochs and the daily activity counts were recorded and downloaded when the device was returned after 7 days.

Analysis of cardiorespiratory data was performed by CosmedK4b^2 software to measure breath-by-breath ventilation, oxygen consumption (VO_2) and the respiratory exchange ratio ($RER = VCO_2/VO_2$). VO_{2peak} was reported as the mean VO_2 during the final 30 seconds of exercise. Maximal oxygen uptake (VO_{2peak}) is expressed as an absolute rate in litres of oxygen per minute (l/min) and as a relative rate in millilitres of oxygen per kilogram of bodyweight per minute (ml/kg/min).

The relation between variables was determined using standard Pearson correlations. A P value of $< .05$ was considered to be significant. Statistical analysis was performed using Statistica-10 which is a statistics and analytics software package developed by StatSoft Inc. (Tulsa, Oklahoma). The relations analysed were:

- cardiorespiratory fitness and amount of physical activity
- walking capacity and amount of physical activity

RESULTS

Flow of participants through the study

Ten children with cerebral palsy agreed to participate in this study and all participants completed the study. The mean age of participants ranged from 8 to 12 years and comprised 6 females and 4 males. All participants had a diagnosis of spastic cerebral palsy and were independent walkers either with or without aids. Eight participants were classified as Level I on the Gross Motor Function Classification Scale (GMFCS) (Palisano et al 1997) and two participants was classified as Level II. Eight participants had hemiplegic cerebral palsy and two had diplegic cerebral palsy. All participants attended a mainstream school in their residential area and were placed in classrooms with their typically-developing peers. Six siblings participated in the study, 3 males and 3 females, aged from 9 to 12 years. Table 6.2 shows the characteristics of participants and siblings and Table 6.3 shows the individual characteristics. Table 6.4 shows the individual characteristics of impairment for participants.

Table 6.2: Group characteristics of participants.

Characteristics	Children with cerebral palsy (n = 10)	Typically-developing siblings (n = 6)
Age *(yr)*, mean (SD)	9.8 (1.6)	10.0 (1.3)
Gender, number male *(%)*	4 (40)	3 (50)
Weight *(kg)*, mean (SD)	38.9 (12.6)	36.7 (8.8)
Height *(m),* mean (SD)	1.4 (0.13)	1.4 (0.10)
BMI *(kg/m^2)*	19.65 (4)	18 (2.3)
Distribution, n hemiplegic (%)	8(80)	n/a
GMFCS, n Level I (%)	8 (80)	n/a
Type (spastic), n (%)	10 (100)	n/a
School type, number mainstream (%)	10 (100)	n/a

BMI =body mass index; GMFCS = Gross Motor Function Classification Scale; yr = year, n = number

Table 6.3: Individual characteristics of all participants (n = 16).

	Age (yr)	Sex (M/F)	Height (m)	Weight (kg)	BMI	Distribution	GMFCS Level
CP participant (n = 10)							
1	8	F	1.3	24	14	Hemiplegic	I
2	12	M	1.6	54	21	Hemiplegic	I
3	11	F	1.5	55	24	Hemiplegic	I
4	9	F	1.4	40	20	Diplegic	II
5	11	F	1.5	40	18	Hemiplegic	I
6	10	M	1.4	52	27	Diplegic	I
7	9	M	1.2	23	16	Hemiplegic	I
8	8	F	1.3	33	20	Hemiplegic	I
9	8	F	1.3	29	17	Diplegic	II
10	12	M	1.6	55	21	Hemiplegic	I
Sibling (n = 6)							
1	9	F	1.3	28	15		
2	12	M	1.6	52	20		
3	9	F	1.4	32	16		
4	9	F	1.3	33	18		
5	10	M	1.3	33	18		
6	11	M	1.4	42	21		

yr = year, m = metre, kg = kilogram, BMI = body mass index (calculated by mass(kg)/height(m)^2), GMFCS = Gross Motor Function Classification Scale level: M = male; F = female

Table 6.4: Individual impairments of participants with cerebral palsy (n=10).

Participant	Strength (R)	Strength (L)	Dexterity (taps/min)(R)	Dexterity (taps/min)(L)	Spasticity V1 (R)	Spasticity V1 (L)	Spasticity V3 (R)	Spasticity V3 (L)	Contracture (R)	Contracture (L)
1	4	4	36	36	1	1	1	1	2	2
2	4	5	44	52	1	0	1	0	2	3
3	3	5	32	32	2	0	2	0	1	1
4	2	3	20	24	2	2	2	2	1	1
5	4	5	28	48	1	0	2	0	1	3
6	4	4	40	24	1	1	1	1	2	1
7	4	5	24	32	1	0	1	0	3	3
8	3	5	36	48	2	0	1	0	1	3
9	2	3	24	28	1	1	2	2	2	2
10	4	5	36	52	1	0	1	0	2	3
Mean (SD)/ Med (IQR)	3.4 (0.8)	4.4 (0.8)	32 (8)	38 (11)	1.3 (0.5)	0.5 (0.7)	1.5 (0.5)	0.7 (0.9)	1.7 (0.7)	2.2 (0.9)

yr = year; F = female; M = male; Hemi = hemiplegic; GMFCS = Gross Motor Classification Scale; Spas = spastic, (R) = right; (L) = left; min = minute; V1 = as slow as possible; V3 = as fast as possible; contracture scored 1 for < 90°, 2 for = 90°, 3 for > 90°.

Feasibility

Recruitment and retention of participants

Names and contact details of children with cerebral palsy aged between 8 - 12 years were provided by the Cerebral Palsy Alliance of NSW. Twenty-two families were contacted and 10 children were able to be recruited to the study. Of these 10 children, 6 siblings were also able to be recruited. All 16 participants completed the study.

Suitability of measurement procedures

All the participants with cerebral palsy were able to walk on the treadmill, walk independently for the 6-minute walk test and wear the Actigraph® accelerometer. All the siblings were able to wear the Actigraph® accelerometer.

Participant compliance with the measurement procedure

All the participants with cerebral palsy performed the progressive exercise test on the treadmill. Two of the participants did not wear the Cosmed K4b2™ mask for gas analysis while performing the test. All the participants with cerebral palsy completed the 6-minute walk test.

All participants reported wearing the Actigraph® accelerometer for the whole 7 days. However, when the data from the devices was downloaded, it was noted that for some accelerometers that no data had been recorded for both participants and siblings on some of the days. Five of the participants (50%) did not have complete data for 7 days. Three (30%) participants had missing data for 1 day and two (20%) participants had missing data for 2 days. Of the siblings, three (50%) did not have

complete data for the 7 days. Two (20%) siblings had missing data for 1 day and one (10%) had missing data for 2 days. On one of the days both the participant and their sibling did not wear the accelerometer as they were involved in an all-day swimming event. Therefore, 90% of the data for physical activity was available to be analysed.

Amount of physical activity

To allow for missing data, all data was averaged to take into account the number of days each participant had worn the accelerometer. Results for individual measurement of physical activity are presented as activity counts (in minutes) over 12 hours across the 7 day period (Table 6.5). The total number of minutes for each 12 hour day was 720 minutes across the 3 categories of physical activity – sedentary, light and moderate to vigorous. These counts have then been averaged across the 7 days as minutes, hours and proportion of the day spent in each physical activity category (Table 6.6).

The activity counts from the accelerometer were also compared with the participant's reported activities in their Activity Record Book over 4 days. Where a participant had recorded a high number of counts in the moderate to vigorous range, the book was checked to see if this matched with the type of activity the participant had been engaged in. Likewise, where there were periods of sedentary activity this was also checked to see whether it matched with the accelerometer counts. There were no obvious anomalies detected between the accelerometer counts and the reported activity.

Table 6.5: Individual activity counts for all participants in minutes over 12 hours across 7 days.

Participant	Day 1			Day 2			Day 3			Day 4			Day 5			Day 6			Day 7		
CP	**Sed**	**Light**	**Mod/vig**	**Sed**	**Light**	**Mod/vig**	**Sed**	**Light**	**Mod/vig**	**Sed**	**Light**	**Mod/vig**	**Sed**	**Light**	**Mod/vig**	**Sed**	**Light**	**Mod/vig**	**Sed**	**Light**	**Mod/vig**
1	431	256	33	321	374	25	345	326	49	344	312	64	296	379	45	332	341	47	335	347	38
2	437	252	31	415	277	28	ND	ND	ND	500	196	24	452	242	26	477	226	17	ND	ND	ND
3	246	434	40	435	278	7	359	349	12	ND	ND	ND	259	433	27	451	241	28	627	83	10
4	608	94	18	381	324	15	416	298	6	343	349	28	487	225	8	516	185	19	517	184	19
5	414	283	24	626	84	10	607	104	9	491	214	15	447	251	22	ND	ND	ND	596	114	10
6	157	375	188	290	363	67	530	157	33	225	427	68	191	431	98	564	147	9	62	553	105
7	388	260	72	369	316	35	320	357	43	350	288	82	351	344	25	381	280	59	603	110	7
8	290	363	67	376	303	41	266	416	38	210	447	63	ND	ND	ND	ND	ND	ND	322	353	45
9	474	240	6	424	281	15	388	314	18	654	64	2	395	321	4	ND	ND	ND	415	279	26
10	182	462	76	452	242	26	415	277	28	411	270	39	408	262	50	385	229	106	408	262	50
Sibling																					
1	342	261	117	560	127	33	524	173	23	408	262	50	433	225	62	436	246	38	337	320	63
2	344	340	36	417	296	7	611	107	2	414	269	37	397	297	26	ND	ND	ND	285	409	26
3	259	386	75	604	106	10	655	56	9	717	3	0	361	336	23	719	0	1	543	151	26
4	424	240	56	411	270	39	435	218	67	477	204	39	463	187	70	385	229	106	493	206	21
5	423	292	5	462	244	14	602	104	14	566	150	4	336	358	26	ND	ND	ND	317	363	40
6	ND	ND	ND	ND	ND	ND	282	373	65	198	458	64	280	404	36	347	340	33	432	234	54

CP = child with cerebral palsy Sed = sedentary; mod/vig = moderate to vigorous; ND = no data recorded

Table 6.6 Group activity counts over a 12 hour day averaged across 7 days

Participant	Sedentary			Light			Moderate to vigorous		
Cerebral palsy	**min/day**	**hr/day**	**Proportion of day**	**min/day**	**hr/day**	**Proportion of day**	**min/day**	**hr/day**	**Proportion of day**
1	343	5.7	0.47	334	5.6	0.45	43	0.7	0.05
2	456	7.6	0.63	238	4.0	0.32	25	0.4	0.03
3	396	6.6	0.55	303	5.0	0.42	21	0.4	0.02
4	467	7.8	0.65	237	4.0	0.32	16	0.2	0.01
5	530	8.8	0.73	175	3.0	0.24	15	0.2	0.01
6	288	4.8	0.40	350	5.8	0.48	81	1.4	0.10
7	395	6.6	0.54	279	4.7	0.38	46	0.7	0.05
8	293	5.0	0.40	376	6.2	0.50	51	0.8	0.06
9	458	7.6	0.63	250	4.2	0.35	12	0.2	0.01
10	380	6.3	0.52	286	4.8	0.39	53	0.9	0.07
Mean (SD)	401 (78)	6.7 (1.3)	0.55 (0.11)	283 (61)	4.7 (1.0)	0.40 (0.08)	36 (22)	0.6 (0.4)	0.04 (0.03)
Sibling									
1	412	6.8	0.57	286	4.7	0.39	23	0.5	0.04
2	434	7.2	0.60	231	3.8	0.30	56	1.0	0.08
3	551	9.2	0.76	148	2.4	0.20	21	0.4	0.03
4	441	7.4	0.60	222	3.7	0.30	57	0.9	0.07
5	451	7.5	0.72	252	4.2	0.42	17	0.3	0.02
6	308	5.2	0.42	362	6.0	0.50	50	0.8	0.06
Mean (SD)	433 (78)	7.2 (1.3)	0.60 (0.12)	250 (71)	4.1 (1.2)	0.35 (0.10)	37 (19)	0.7 (0.3)	0.05 (0.02)

Sedentary = <100 counts/minute; Light = >100 but <2000 counts/minute; moderate to vigorous = >2000 counts/minute; min = minute; hr = hour

On average, the participants spent over 6.5 hours each day in sedentary activity (range 5-9 hr), 5 hours in light activity (range 3-6 hr) and just over 30 minutes in moderate to vigorous activity (range 20 min-1.5 hr). In terms of proportion of the day, this shows that participants spent over half of each day in sedentary activity and approximately 4% of their day in moderate to vigorous physical activity.

On average, the results for siblings were similar with slightly more time spent in sedentary activity (7 hours, range 5-9 hours), less time in light activity (4 hours, range 2-6 hours) and the same amount of time in moderate to vigorous activity (range 30 minutes-1 hour). In terms of proportion of the day, this shows that siblings also spent over half of each day in sedentary activity and approximately 5% of their day in moderate to vigorous physical activity.

Therefore in comparison, the participants with cerebral palsy spent 5% of their day (95% CI -0.18 to 0.08) less in sedentary activity than their siblings, 5% (95% CI -0.05 to 0.15) more of their day in light physical activity, and 1% (95% CI -0.04 to 0.02) less of their day in moderate to vigorous physical activity (Table 6.7 and Figure 6.5).

Table 6.7: Mean (SD) values of proportion of day spent in physical activity of participants with cerebral palsy (n = 10) and siblings (n = 6) and mean (95% CI) difference between them over 7 days.

Physical activity	Groups		Difference between groups
	Participants with cerebral palsy (n = 10)	Siblings (n = 6)	Participants with CP minus siblings
Sedentary	0.55 (0.11)	0.60 (0.12)	-0.05 (-0.18 to 0.08)
Light	0.40 (0.08)	0.35 (0.10)	0.05 (-0.05 to 0.15)
Moderate/vigorous	0.04 (0.03)	0.05 (0.02)	-0.01 (-0.04 to 0.02)

CP = cerebral palsy; n = number

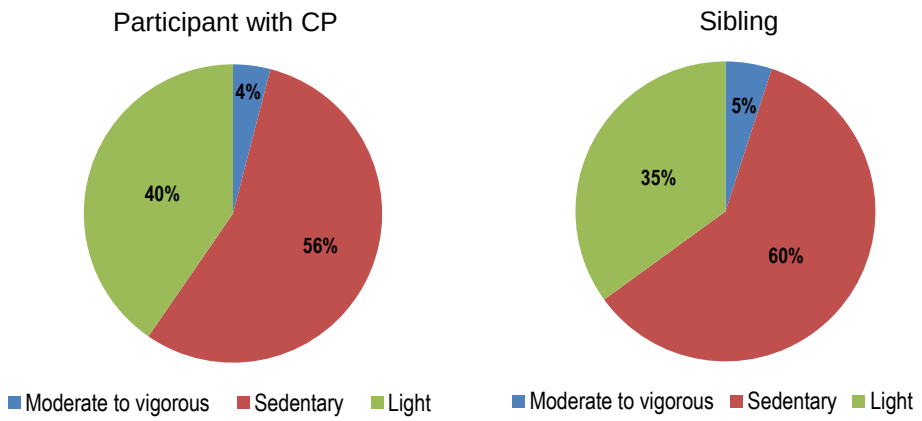

Figure 6.5: *Proportion of day in physical activity - participant with cerebral palsy (CP) and sibling.*

Level of cardiorespiratory fitness

All ten participants completed the progressive exercise test on the treadmill. However, two participants did not have respiratory gas analysis as they were unable to wear the Cosmed K4b2™ mask. Therefore, on the basis of results from other participants with similar exercise test times it was decided to use their results to equate to similar levels of VO_{2peak} for l/min and ml/kg/min respectively.

The duration of the progressive exercise test on the treadmill ranged from 6.5–23.0 minutes with the mean being 13.8 minutes. The VO_{2peak} in l/min ranged from 0.62–2.37 with a mean of 1.12 l/min. The VO_{2peak} in ml/kg/min ranged from 16–43 with a mean of 27 ml/kg/min. Results for VO_{2peak} in l/min and ml/kg/min are e reported in Table 6.8.

Table 6.8: Individual levels of cardiorespiratory fitness of participants with CP (n = 10).

Participant	Pre-test heart rate (bpm)	Peak heart rate (bpm)	Time on treadmill (min)	VO_{2peak} (l/min)	VO_{2peak} (ml/kg/min)
1	65	138	18.0	1.57*	29*
2	125	169	18.1	1.57	29
3	95	128	13.2	1.19	22
4	96	145	6.5	0.62	16
5	80	146	18.4	0.92	23
6	78	145	15.3	0.82	16
7	105	153	7.4	1.29	56
8	102	140	12.2	0.70	22
9	106	145	6.5	0.62*	16*
10	128	170	23.0	2.37	43
Mean (SD)	98 (20)	147 (6)	13.8 (5.7)	1.12 (0.56)	27 (14)

* substitution of results for missing data
BPM = beats per minute; VO_{2peak} = maximal oxygen uptake; l/min = absolute rate in litres of oxygen per minute; ml/kg/min = relative rate in millilitres of oxygen per kilogram of bodyweight per minute; min = minute

The peak heart rates for participants following the progressive exercise test ranged from 128–170 bpm with a mean of 147 bpm. There was a moderate correlation between peak heart rate and cardiorespiratory for both of the measures of VO_{2peak} in l/min or ml/kg/min (Table 6.9 and Figures 6.6 and 6.7).

Table 6.9: Correlation between peak heart rate and cardiorespiratory fitness in children with cerebral palsy presented as r (*p*-value).

	Cardiorespiratory fitness	
Variable	**VO_{2peak} (L/min)**	**VO_{2peak} (ml/kg/min)**
Heart rate	0.60 (p = 0.06)	0.50 (p = 0.14)

Heart rate = beats per minute; VO_{2peak} = maximal oxygen uptake; l/min = absolute rate in litres of oxygen per minute; ml/kg/min = relative rate in millilitres of oxygen per kilogram of bodyweight per minute

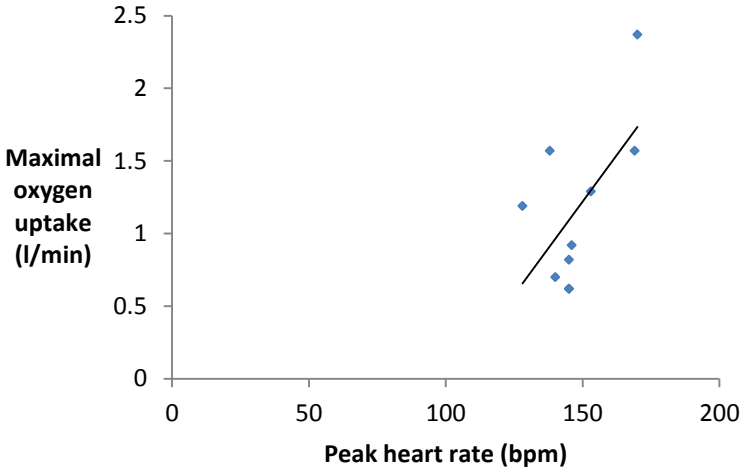

Figure 6.6: *The relation between peak heart rate (bpm) and cardiorespiratory fitness as measured by maximal oxygen uptake (VO_{2peak} l/min)*
(NB: Only 9 data points are visible as 2 results were the same for both heart rate and maximal oxygen uptake).

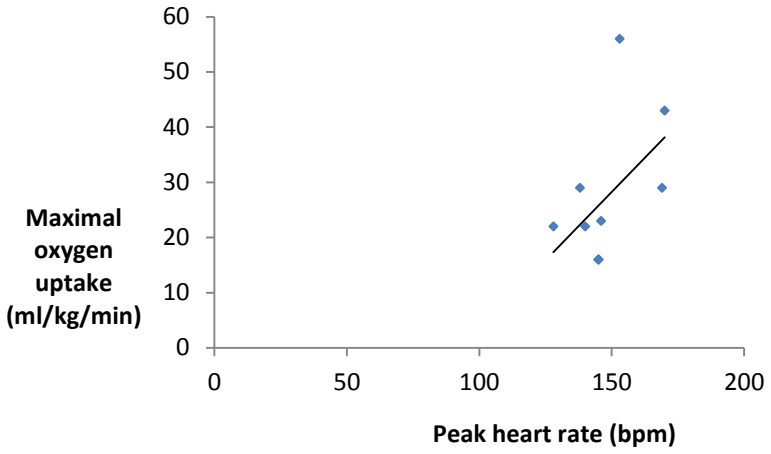

Figure 6.7: *The relation between peak heart rate and cardiorespiratory fitness as measured by maximal oxygen uptake (VO_{2peak} ml/kg/min)*
(NB: Only 8 data points are visible as 3 results were the same for both heart rate and maximal oxygen uptake).

Walking capacity

Nine (90%) of the ten participants completed the 6-Minute Walk test (6-MWT). One participant was unwell on the day of testing and did not want to attempt the test. Only one participant elected to have a break during the test and rested for 30 seconds before continuing with the test. The mean distance walked for the group was 382 (107) metres.

Participants wore the accelerometer during the 6-MWT with 8 of the 9 (88%) participants achieving moderate to vigorous levels, ie, >2000 counts, on the accelerometer, for each minute of the test. However, one participant only achieved 1 minute of moderate-to vigorous levels of physical activity throughout the entire test. Individual results for distance covered, pre-test heart rate, peak heart rate and activity counts are presented in Table 6.10.

Table 6.10: The 6-Minute Walk Test (n = 10).

Participant with CP	6 MWT Distance (m)	6MWT rests (#)	Pre-test heart rate (bpm)	Peak heart rate (bpm)	Activity counts during each minute						# of min >2000
					1 min	2min	3min	4 min	5 min	6 min	
1	263	0	65	96	4060	4792	4913	4622	4902	2878	6
2	DNA	DNA	DNA	DNA	DNA	DNA	DNA	DNA	DNA	DNA	DNA
3	315	0	95	106	1212	1506	1978	1663	1668	4203	1
4	203	1	96	105	2483	2602	2160	2340	2002	2312	6
5	495	0	80	127	4350	4468	4695	3931	4440	2892	6
6	428	0	78	104	2506	2610	3033	3002	3154	2439	6
7	477	0	105	124	4871	4914	4482	3620	4678	4779	6
8	490	0	102	140	4022	3965	3917	3687	4036	2348	6
9	330	0	106	150	3692	3952	3742	4594	4450	3234	6
10	440	0	96	128	2103	2425	2740	2966	2038	3016	6
Mean (SD)	382(107)		95(19)	125(25)	3255 (1219)	3470 (1212)	3518 (1091)	3381 (988)	3485 (1289)	3122 (849)	

CP = cerebral palsy; m = metres; # = number; 6-MWT = 6 Minute Walk Test; bpm = beats per minute; min = minute; DNA = did not attempt as unwell on the day of testing

Relation between cardiorespiratory fitness and amount of physical activity

There was little, or no, relationship between cardiorespiratory fitness and the amount of moderate to vigorous physical activity in children with cerebral palsy for either of the measures of VO_{2peak} in L/min or ml/kg/min (Table 6.11 and Figures 6.8 and 6.9).

Table 6.11: Correlation between cardiorespiratory fitness and amount of physical activity presented as r (*p*-value).

	Cardiorespiratory fitness	
Variable	**VO_{2peak} (l/min)**	**VO_{2peak} (ml/kg/min)**
Physical activity	0.25 (p = 0.48)	0.20 (p = 0.58)

Physical activity = moderate to vigorous as measured by the Actigraph® accelerometer; VO_{2peak} = maximal oxygen uptake; l/min = absolute rate in litres of oxygen per minute; ml/kg/min = relative rate in millilitres of oxygen per kilogram of bodyweight per minute

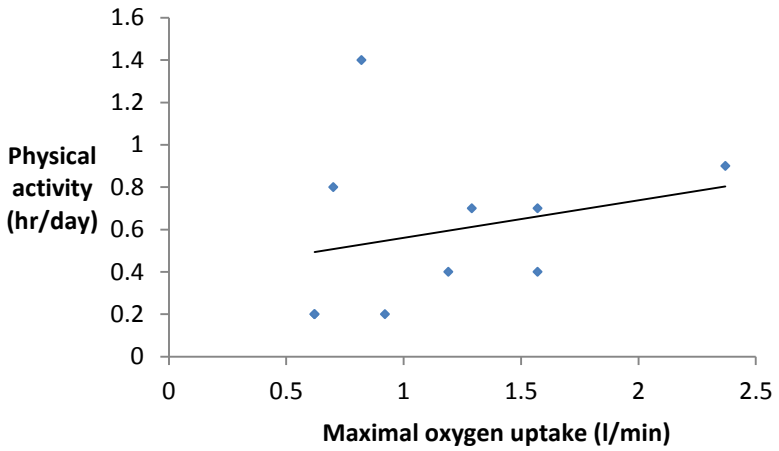

Figure 6.8 *The relation between cardiorespiratory fitness as measured by maximal oxygen uptake (VO_{2peak} l/min) and the amount of moderate to vigorous physical activity.*
(NB: Only 9 data points are visible as 2 results were the same for both physical activity and maximal oxygen uptake).

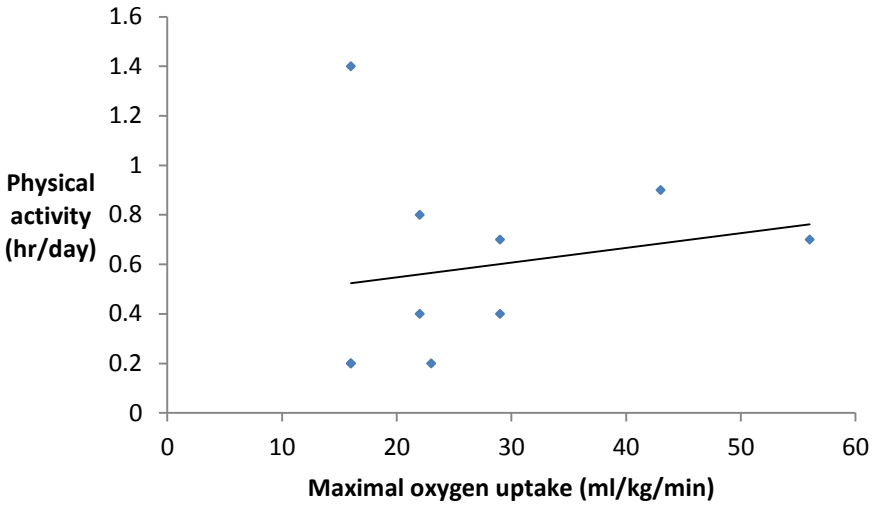

Figure 6.9: *The relation between cardiorespiratory fitness as measured by maximal oxygen uptake (VO_{2peak} ml/kg/min) and the amount of moderate to vigorous physical activity.*
(NB: Only 9 data points are visible as 2 results were the same for both physical activity and maximal oxygen uptake).

Relation between walking capacity and amount of physical activity

There was little, or no, relationship between walking capacity and the amount of moderate to vigorous physical activity in children with cerebral palsy (Table 6.12 and Figure 6.10).

Table 6.12: Correlation between walking capacity and amount of physical activity presented as r (*p*-value).

Variable	Walking capacity
Physical activity	0.07 (p = 0.84)

Walking capacity = measured by the 6- Minute Walk Test;
Physical activity = moderate to vigorous as measured by the Actigraph® accelerometer

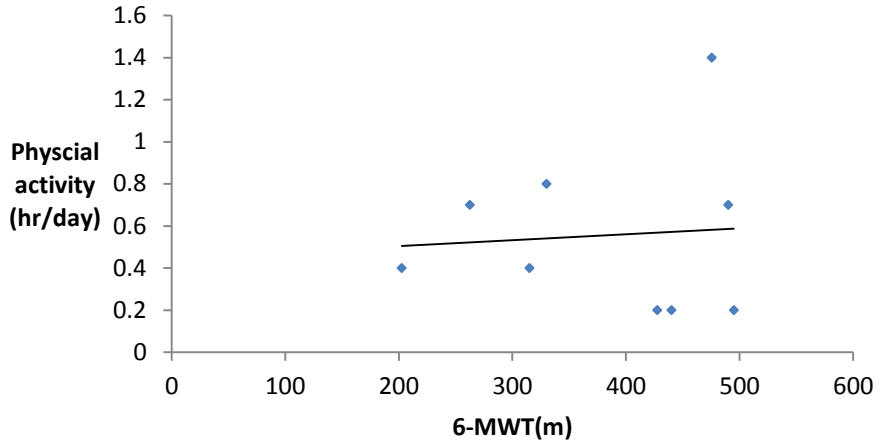

Figure 6.10: *The relation between walking capacity and amount of moderate to vigorous physical activity.*
(NB: Only 9 data points are visible as 2 results were the same for both physical activity and walking capacity).

DISCUSSION

The procedures used in this study are feasible to investigate physical activity, cardiorespiratory fitness and walking capacity in independently walking children with cerebral palsy. Participants were able to be recruited through the major cerebral palsy service provider in Sydney, the Cerebral Palsy Alliance. Staff from the Cerebral Palsy Alliance identified potential participants who were likely to be interested in participating in the study. In addition, as the testing for the study was conducted close to participant's homes, or in their usual physiotherapy intervention environment, it was considered less likely that there would be issues with retention. However, despite access to potential participants through the Cerebral Palsy Alliance, it took 18 months to recruit the 10 children with cerebral palsy and their siblings. Most of the recruitment was conducted by telephone invitation. Notably, 4 of the participants were recruited at one exercise class run by the Cerebral Palsy

Alliance. Therefore, in a future study it may be better to attend such classes in order to meet potential participants and their families face-to-face. Recruitment could also be extended to include other agencies which provide services for children with cerebral palsy eg the children's hospitals.

The measurement procedures used to measure physical activity, cardiorespiratory fitness and walking capacity have all been used in other studies examining physical activity in children with cerebral palsy (Capio et al 2010, Clanchy et al 2011; Verschuren et al 2006; Maher et al 2008) and were therefore considered to be feasible for this study. Overall, the participants were compliant with the measurement procedures. However, two participants were fearful of wearing the CosmedK4b^2 face mask during the treadmill test. In a future study, this could be overcome by having a familiarisation period where participants could observe a video of the testing procedure and also handle the face mask. In addition, pairing participants or conducting testing in small groups where participants could observe each other performing the test could allay fears.

When data was retrieved from the Actigraph® accelerometer, it was apparent that some participants had not worn the accelerometer for the whole 7 days. Participants reported spending the day at the beach or swimming in a pool and as the model of accelerometer used was not water resistant, it had to be removed for water sports. In a future study, the more recent water resistant model of the Actigraph® accelerometer, ie, GT3X+ could be used.

A fully powered study would require 20 participants which would provide 80% power to detect a 10% difference in sedentary or light activity between children with cerebral palsy and their typically-developing siblings at the p=0.05 level. However at this stage, it is worthwhile examining the findings so far.

In this study, the participants with cerebral palsy, were classified as either Level I or II on the GMFCS and were therefore independently walking in their community and school environment. Overall, there was only a small amount of difference in the amount of physical activity between the participants with cerebral palsy and their siblings. Both groups spent 95% of their day in either sedentary or light physical activity and neither group met the recommended guidelines of 60 minutes daily moderate to vigorous physical activity. Furthermore, the participants with cerebral palsy had decreased levels of cardiorespiratory fitness and walking ability compared to published literature of typically-developing children (Shvartz and Reibold 1990, Krahenbuhl et al 1985, Cooper et al 1984, Pate et al 2006, Ten Harkel et al 2010). However, these levels are similar to reported levels for children with cerebral palsy (Hoofwijk et al 1995, Maltais et al 2005, Verschuren & Takken 2010). In this study, neither cardiorespiratory fitness, nor walking capacity, seemed to explain the amount of physical activity in the participants with cerebral palsy.

Investigating a child's physical activity within their free-living environment is important to determine what they do on a day-to-day basis. Studies have examined physical activity in children with cerebral palsy within a structured environment (Capio et al 2010, Clanchy et al 2011), but less is known about their free-living activities. In particular, in terms of 'incidental' free-living physical activity, it may

also be relevant to know the child's method of transport to and from school, along with their activities before and after school.

Direct measures of the amount of free-living physical activity were made using an Actigraph® accelerometer which all participants wore for 7 days. Despite some participants not wearing the accelerometer for the whole 7 days, there was still 90% of the data available to be analysed. Over a 12 hour day, there was very little difference in free-living physical activity between the participants with cerebral palsy and their peers. In terms of proportion of the day, participants spent over half of each day in sedentary activity and only approximately 5% of their day in moderate to vigorous physical activity. From these results it is apparent that participants are engaged in less than the recommended guidelines of 1 hour each day of moderate to vigorous activity for typically-developing children (Department of Health and Ageing 2005).

There may be several reasons for participants not meeting recommendations for daily moderate to vigorous physical activity. It is commonly recognised that worldwide, typically-developing young children and adolescents are not meeting these guidelines and indeed, reduced physical activity is one of the main factors attributed to rising levels of obesity. Therefore, the amount of physical activity seen in this group may not be dissimilar to the general population and in particular, the child with cerebral palsy could be fitting in with their family's typical activity behaviours.

In contrast, the physical impairments of the child with cerebral palsy could be impacting the amount of physical activity which the typically-developing sibling

could be engaged in. Interestingly, as reported in the activity record book, while some participants with cerebral palsy were engaged in structured physical activities, such as horse-riding or swimming, the entry in the sibling's record book showed that they had been engaged in sedentary activities such as computer time or television viewing. Therefore, time may be spent ensuring that the child with cerebral palsy is engaged in physical activity however this could be limiting the amount of physical activity of the sibling.

Accessibility of free-living activities outside the family home may be another factor for reduced activity of the participant with cerebral palsy. While parents of children with cerebral palsy usually want them to be engaged in physical activity, it is not always easy to establish activities outside the home environment. Accessibility, time commitments and the cost of many community activities, may place limits on a family's capability to engage in habitual physical activity.

All participants with cerebral palsy were found to have low cardiorespiratory fitness levels, as measured directly by maximal oxygen uptake, for both of the measures of VO_{2peak} in l/min and ml/kg/min compared to expected values. The levels achieved by the participants, were approximately 30% less than might be expected for typically-developing children (Shvartz and Reibold 1990, Rosenthal & Bush 2000, Krahenbuhl et al 1985, Cooper et al 1984, Pate et al 2006, Ten Harkel et al 2010). In comparison to other children with cerebral palsy, there is little published data about VO_{2peak} during a maximal progressive exercise test. However, from the data available, the results achieved by the participants in this study are similar to reported

results from other studies which investigated maximal exercise testing in children with cerebral palsy (Hoofwijk et al 1995, Maltais et al 2004, Maltais et al 2005).

Children are known to increase their oxygen consumption up to 10-fold during intense exercise (Washington et al 1994). However, for children with cerebral palsy, this increase is likely to be higher, for the same exercise, than seen in typically developing children and may be related to lower breathing efficiency due to possible chest wall distortion and/or respiratory muscle spasticity (Unnithan et al 1999, Unnithan et al 2004, Maltais et al 2005, Piccinini et al 2007, van den Berg-Emons et al 1995, Rose et al 1989).

Children with cerebral palsy do not seem to tolerate sustained periods of exercise and are more likely to fatigue at low exercise intensities than typically-developing peers (Unnithan et al 2004). The results in the present study are consistent with this observation and the average time that participants spent on the treadmill was less than could be expected for typically-developing children. The early fatigue may also be related to musculoskeletal impairment commonly seen in children with cerebral palsy, with lower limb weakness, spasticity and contracture generally a feature in these children (Campbell et al 2011, Unnithan & Maltais 2004, Bar-Or 1996). Despite the participants in the study being independent walkers, they all had some level of weakness, spasticity or contracture in their lower limbs. The musculoskeletal impairment, of predominantly weakness, seen in children with cerebral palsy may mean that they are reaching their anaerobic threshold sooner than typically-developing children, and are therefore not as efficient. None of the participants had contractures in their lower limbs, but did have some spasticity. It

has been suggested that the presence of spasticity in the lower limbs of children with cerebral palsy, may cause a local obstruction of venous return thereby increasing muscular fatigue (Hoofwijk et al 1995). Muscle fatigue is particularly an issue during periods of intense exercise as it becomes necessary to increase the peripheral oxygen extraction from skeletal muscles (Washington et al 2008, American College of Sports Medicine 2000, Braden & Carroll 1999).

The presence of musculoskeletal impairment may also mean that some participants may not have had time to adjust to the speed increments on the treadmill. The speed of the treadmill was increased in 1 minute increments in line with previous treadmill protocols for a progressive exercise test (Verschuren et al 2006). However, some studies suggest that it may take up to 45-60 seconds for children with cerebral palsy to accommodate to an increase in treadmill speed (Hoofwijk et al 1995, Unnithan and Maltais 2004). Therefore, the participants may have still been adjusting to a change in speed when the next increment was implemented.

Another indicator of cardiorespiratory fitness is the heart rate response to exercise. The peak heart rate achieved by the participants on the progressive exercise test was on average 25% less than for typically developing children (Riopel et al 1979, Wilmore et al 1982, James et al 1980, Alpert et al 1982, Ten Harkel et al 2010). However, this result is similar to other studies comparing peak heart rate in children with cerebral palsy (Maltais et al 2005, Piccinini et al 2007, Keefer et al 2004). In addition, in the present study, there was a moderate correlation between peak heart rate and oxygen uptake during the progressive exercise test suggesting that these

participants had a normal cardiorespiratory response to exercise of increasing intensity (Washington et al 2008, Armstrong et al 1997).

The walking capacity of participant's was measured using the 6-Minute Walk Test. The distance walked by the participants was less than that found in one study of children with cerebral palsy (Maher et al 2008), but was within the range of another study (Thompson et al 2008). On average, the distance achieved by participants is also less compared to distances achieved by typically-developing children (Lammers et al 2008, Geiger et al 2007). Interestingly, the activity counts from the accelerometer showed that the participants in the present study were capable of achieving moderate to vigorous levels of physical activity during each minute of the test. However, there was little, or no, relationship between walking capacity and cardiorespiratory fitness. This result is in contrast to another study which reported improvement in cardiorespiratory fitness and walking distance (Gorter et al 2009). However, this study did not directly measure maximal oxygen uptake and instead assumed a linear relation between peak heart rate and oxygen uptake.

Limitations of study

Maximal exercise testing is the preferred method for determining the performance-limiting factors to exercise (Verschuren et al 2011). However, the difficulty of encouraging children to perform a maximal exercise test to exhaustion is well recognised. Indeed, in this study some participants did not seem exhausted when they stopped the test. Therefore, the overall results may not be a true reflection of maximal oxygen uptake for this group.

Previous studies have identified the importance of habituation to walking on a treadmill (Hoofwijk et al 1995, Unnithan et al 1996, Unnithan et al 2003, Keefer et al 2004, Verschuren et al 2010, Maltais et al 2004, Potter & Unnithan, 2005). It is recommended that participants have a period of familiarisation on the treadmill of approximately 5 minutes. It has also been suggested that more than 5 minutes familiarisation may be necessary for some children with cerebral palsy (Potter & Unnithan 2005). In addition, during both familiarisation and testing, participants should be instructed not to hold on to the handle bars of the treadmill. Participants in this study were given a 5 minute period of familiarisation at the lower treadmill speed and there were no observed difficulties in performance by participants once testing had commenced. However, 2 participants at Level II on the GMFCS, needed to have touch contact with the handle bars at various stages of the test.

CONCLUSION

In this study, the participants with cerebral palsy had similar amounts of moderate to vigorous physical activity as their typically-developing siblings. However, this finding is not consistent with other studies (Maher et al 2007, Law et al 2006, King et al 2003) and is also less than the recommended guidelines for daily physical activity. As there was no relation between cardiorespiratory fitness and walking capacity, with the amount of physical activity, it is worth examining why these children with cerebral palsy are not being physically active. In particular, there may be identifiable barriers which are preventing these children from participating in habitual free-living physical activity in their community environment.

CHAPTER 7

IDENTIFICATION OF BARRIERS TO PHYSICAL ACTIVITY AND PARTICIPATION FOR CHILDREN WITH CEREBRAL PALSY: A FEASIBILITY STUDY

INTRODUCTION

METHODS

Design

Participants

Outcome measures

Data analysis

RESULTS

Flow of participants through the study

Feasibility

Current physical activity

Preferred physical activity

Parent's perception of barriers

DISCUSSION

How can barriers be overcome to increase daily activity?

INTRODUCTION

Physical activity involves body movement produced by skeletal muscles which results in an increase in energy expenditure and enables an individual to carry out everyday actions and processes (Caspersen et al 1985, Dollman et al 2009). The benefits of regular physical activity are well known, and for children particularly, the habits they form in childhood are likely to influence their physical activity behaviours into adulthood (Malina 2001). Therefore, promoting lifetime physical activity is critical for reducing the risks associated with adult diseases (Strong et al 2005).

Anecdotally, it would seem that children with cerebral palsy want to engage in activities alongside their typically-developing peers. In addition, it has been reported that for children with cerebral palsy, the ability to participate in community events is closely related to their perceived quality of life and psychosocial function (Bjornson et al 2008). Children with cerebral palsy have been noted to engage in activities which are conducted at a relatively low intensity but have a high perception of enjoyment (Imms et al 2008). These types of activities are generally 'small screen' activities as they are based around television, computer or gaming consoles (Trost et al 2002). This choice of activity is commonly socially acceptable for their age, and also may be easier to perform if the child has a motor impairment which is preventing them from engaging in more physical activity. However, while the motor impairment in children with cerebral palsy is generally thought to be the main determinant for reduced physical activity, there may be other factors which are limiting their participation in common age-related activities.

From the study presented in Chapter 6, it was evident that the children with cerebral palsy had similar levels of moderate to vigorous activity as their typically-developing siblings but that neither group were meeting the recommended guidelines (Department of Health and Ageing 2005). For the children with cerebral palsy, this is concerning particularly as they are ambulant children who are not potentially using their physical capability. It is known that the motor impairments associated with cerebral palsy may constrain a child's physical activity. In particular, reduced cardiorespiratory fitness and/or walking capacity may limit the amount of physical activity possible. However, as no relation was found between the cardiorespiratory fitness and the walking capacity of the participants compared to the amount of their physical activity, the question arose as to whether there were other potential barriers which were preventing them from participating in activities.

One study has examined the potential barriers preventing children and/or adolescents with physical disabilities from engaging in their preferred choice of activity (Buffart et al 2009). However, the participants were young adults whose level of impairment ranged from mild to severe and therefore they did not necessarily have the potential to participate in many physical activities. The identified barriers were grouped into personal barriers and environmental barriers. Personal barriers included physical issues and psychological issues, and environmental barriers included lack of professional support, inclement weather, limited facilities, cost, and transport.

At the time of planning this study, there were no studies which examined barriers to participation in activity from the parents' perspective for children with cerebral palsy who had mild impairments which would theoretically enable them to engage in a

normal amount of physical activity. Therefore, the aim of this study was to examine the feasibility of investigating the choice of physical activity for children with cerebral palsy in order to answer the following research questions:

1. What are the physical activities that the children are currently engaged in?
2. Are there activities they would prefer to do if they had the choice?
3. What do parents see as the barriers preventing their child from being engaged in their preferred activities?

The hypothesis was that the procedures for measuring preferred choice of physical activity will be feasible in these children. Furthermore, the choice of preferred physical activity undertaken by these children could be explained by the potential barriers to activity as perceived by their parents.

METHODS

Design

This study was a descriptive study to investigate why children with cerebral palsy who were ambulatory were not participating in their choice of activity. The children and their parents attended either one of the NSW Spastic Centre/Cerebral Palsy Alliance sites in the Sydney metropolitan area, or the Lidcombe Campus of the University of Sydney. The children and their parents were asked about the child's current physical activity and what they saw as the barriers preventing them from engaging in some activities. In order to minimise responses being influenced by each other, the children and parents were located in separate parts of the same room while the questions were asked.

Ethics approval was gained through The University of Sydney and The Spastic Centre of New South Wales (now known as the Cerebral Palsy Alliance) Ethics Committees to recruit participants for the study. Physiotherapists from The Spastic Centre of New South Wales invited children and their families registered with that organisation to participate in the study (Appendix C). A verbal and written explanation of the study was given to the parents of the participants (Appendix D).

Participants

Children with cerebral palsy and their parents were invited to participate in the study. The child needed to have a classification of cerebral palsy, be aged between 8 and 12 years and have a Gross Motor Function Classification Score (GMFCS) of Level I or II, ie, they could walk independently. Upon agreeing to participate, parents were given an Information Sheet (Appendix G) to keep and a Consent Form (Appendix H) to sign.

In order to describe the characteristics of the children, anthropometric measures (height and weight), measures of impairment (contracture, spasticity, strength and coordination), classification of cerebral palsy, year and type of school attended were recorded.

Outcome measures

Feasibility

Feasibility of this study was measured as participant recruitment and retention, and suitability of measurement procedures.

Barriers to physical activity

In order to identify the potential barriers to physical activity, children and parents were asked questions relating to a list of 20 physical activities. The activities chosen were intended to be reflective of a mix of physical activities which the child might be engaged in either alone, or with other people, and in addition, be performing these activities either within their home or community. These activities were – martial arts, swimming, gymnastics, horse riding, athletics, team sports, dancing, walking, bike riding and skating, water sports, snow sports, swings and slides at the park, handball (and similar games), gardening, fishing, 'gym' activities (either at a commercial gym or at home), tennis or badminton, going to the movies, going to a live concert or other event and going on a general outing/excursion or picnic.

For each activity children were asked three questions:

1. Are you doing this activity at the moment? (yes/no)
2. If no, would you like to do this activity? (yes/no)
3. How much would you like to do this activity? (a little/a lot)

For each activity the parents were also asked three questions:

1. Is your child doing this activity at the moment? (yes/no)
2. If no, do you think they would like to? (yes/no)

3. How much do you think your child would like to do this activity? (a little/a lot)

Parents were asked to indicate what they saw as being the barriers which were either preventing their child from performing an activity or which were influencing how frequently their child performed the activity. Parents were given examples of potential barriers such as cost, location and availability of activities and time constraints, and could also indicate other barriers which they had identified. They could indicate as many barriers for each activity as were relevant.

Data analysis

A convenience sample of 10 children with cerebral palsy, who were able to walk independently, was collected in order to provide information to conduct a power analysis for a future study.

Descriptive characteristics of the children (anthropometric measures, measures of impairment, classification of cerebral palsy, and type of school attended) are expressed as mean (SD) unless otherwise indicated.

The analysis of the current physical activity of the children is presented as a simple tally of the number of activities engaged in.

The analysis of preferred activity and level of interest in engaging in the activity are presented as the number of responses of parents and children and the percentage agreement between them for each activity.

The perception of barriers to physical activity is presented as the number and percentage of parent responses for each activity and identified barrier.

RESULTS

Flow of participants through the study

The ten children with cerebral palsy, who had participated in the previous study presented in Chapter 6, agreed to participate in this study and all responded to the activity questions. The mean age of the children ranged from 8 to 12 years and comprised 6 females and 4 males. All children had a diagnosis of spastic cerebral palsy and were independent walkers either with or without aids. Eight children were classified as Level I on the Gross Motor Function Classification Scale (GMFCS) (Palisano et al 1997) and two children were classified as Level II. Eight children had hemiplegic, and two, had diplegic cerebral palsy. All children attended a mainstream school in their residential area and were placed in classrooms with their typically developing peers. Group characteristics of children participating in the study are presented in Table 7.1.

Table 7.1 Group characteristics of children

Characteristics	Children with cerebral palsy (n = 10)
Age *(yr)*, mean (SD)	9.8 (1.6)
Gender, number male *(%)*	4 (40)
Weight *(kg)*, mean (SD)	38.9 (12.6)
Height *(m),* mean (SD)	1.4 (0.13)
BMI *(kg/m^2)*	19.65 (4)
Distribution, n hemiplegic (%)	8(80)
GMFCS, n Level I (%)	8 (80)
Type (spastic), n (%)	10 (100)
School type, number mainstream (%)	10 (100)

BMI =body mass index; GMFCS = Gross Motor Function Classification Scale; yr = year, n = number

Ten parents (9 female, 1 male), participated in the study and all responded to the activity questions.

Feasibility

Recruitment and retention of participants

The 10 children with cerebral palsy, who had participated in the study reported in Chapter 6 and their parents, were able to be recruited to this study and all completed the study.

Suitability of measurement procedures

The physical activities which were chosen encompassed the range of activities children participated in. There were no additional physical activities which were identified by the children or their parents.

Current physical activity

All but one (martial arts) of the 20 activities was reported as being currently performed, or had been performed by at least one child in the preceding 12 months (Table 7.2).

Table 7.2: Number (%) of children participating in each activity (n =10).

Activity	Children participating
1.Martial arts	0 (0)
2.Swimming	9 (90)
3.Gymnastics	2 (20)
4.Horse-riding	2 (20)
5.Athletics	7 (70)
6.Team sports	6 (60)
7.Dancing	4 (40)
8.Walking	5 (50)
9.Bike/skating	5 (50)
10.Water sports	1 (10)
11.Snow sports	1 (10)
12.Swings/slides at park	8 (80)
13.Handball	9 (90)
14.Gardening	4 (40)
15.Fishing	3 (30)
16.Gym activities	8 (80)
17.Tennis/badminton/squash	5 (50)
18.Movies	8 (80)
19.Live event/concert	4 (40)
20.General outing/excursion	6 (60)
Mean (SD)	5 (50)

Eleven of the 20 activities were currently being performed by over half (50%) of the children. Swimming and unstructured games, such as handball, were being performed most frequently (90%). Performing activities on swings, slides and other equipment at a park, gym type activities, and going to the movies were the next

highest group of activities (80%). Figure 7.1 shows the percentage of children participating in the activities.

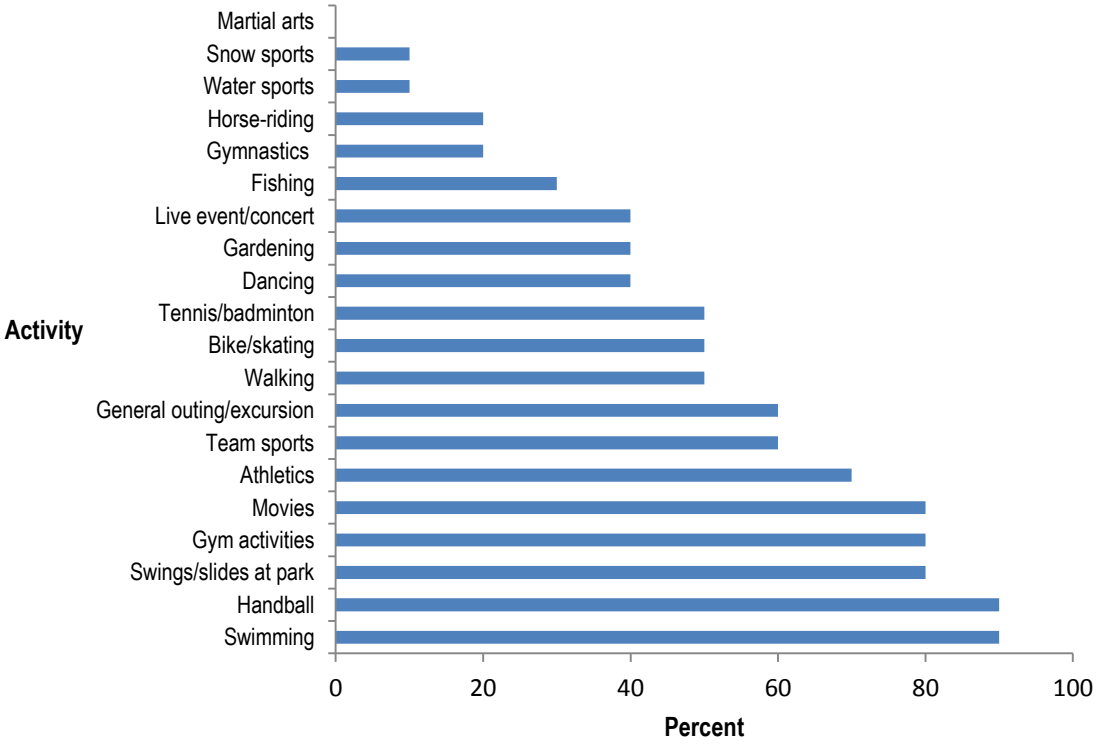

Figure 7.1: *Percentage of children participating in activity (n = 10) from least to most*

Preferred physical activity

Nine activities were rated as being performed less than 50% of the time and for most of these activities children and parents responded that these were activities they would like to be engaged in. These activities were identified as snow sports, water sports, dancing, horse-riding, fishing, martial arts, live events, gymnastics and gardening. The comparison of child and parent responses for preferred activity in terms of whether they would like to do the activity, and how much they would like to be doing it is presented as a percentage agreement between the child and parents (Table 7.3).

Table 7.3: Percentage agreement between child (n = 10) and parent (n = 10) for perception of desire to perform activity not currently performed.

Activity	Desire to do activity			Level of interest in activity					
				A bit			A lot		
	Child (n)	Parent (n)	Agreement (%)	Child (n)	Parent (n)	Agreement (%)	Child (n)	Parent (n)	Agreement (%)
Snow sports	9	9	100	5	5	100	4	4	100
Water sports	4	4	100	4	3	75	0	1	10
Dancing	1	1	100	1	1	100	0	0	100
Horse-riding	5	4	90	0	1	10	5	3	60
Fishing	4	5	80	2	3	66	2	2	100
Martial arts	3	4	75	3	4	75	0	0	100
Live event/concert	4	6	66	0	3	30	4	3	75
Gymnastics	2	5	40	2	3	66	0	2	20
Gardening	0	3	30	0	3	30	0	0	100
Mean (SD)	4 (3)	5 (2)	76 (26)	2 (2)	3 (1)	61 (32)	2 (2)	2 (1)	74 (36)

n = number; % = percentage

Overall the level of agreement was high between the children and their parents in terms of preference of activity. Within these activities, there was 100% agreement between children and parents in terms of choice for snow sports, water sports and dancing. Even though the most common choice of activity (n = 9), was snow sports, the response was equally divided in terms of the desire to engage in the activity. For water sports and dancing, the level of agreement indicated that there was only a small desire to be engaged in the activity.

The only real difference in perception, of preferred choice of activity, was noted for gardening, where 3 parents reported that they thought their child would like to do this activity in contrast to no children choosing it.

Parent's perception of barriers

Parents perceived potential barriers for 16 of the 20 activities (80%). There was a total of 92 responses across the 16 identified barriers and for 75% of these parents identified one or more barriers to participation in an activity. The main barrier was perceived by parents as being the cost of the activity (35%) followed by location (23%), time constraints (14%) and availability (12%) of activities. In the 'other' category (16%), parents identified safety concerns, effect of the child's physical impairment on an activity and, not a culturally relevant activity, as being barriers. In contrast, there were four activities where no barriers to participation were perceived. These activities included going to a local park and playing on swings/slides, unstructured games such as handball, 'statues' and trampoline, gardening and going to the movies.

The perceived barriers to physical activity are separated into categories of frequency of the activity, ie, everyday activities, less frequent (1-2/week) or occasional/holiday activities. The percentage of responses to perceived barriers are presented in Table 7.4 and Figure 7.2

Table 7.4: Number of responses (n = 92) from parents for each category of barrier.

Activity	Barriers					
	Cost	Location	Availability	Time	Other	Total
≥1-2/week						
Walking	0	0	0	0	2	2
Bike/skating	1	3	0	0	1	5
Martial arts	2	1	0	2	2	7
Swimming	3	0	0	0	0	3
Gymnastics	0	2	2	2	2	8
Tennis/badminton/squash	0	0	0	1	0	1
Gym activities	4	0	5	0	0	9
Outing/excursion	0	0	0	2	1	3
Athletics	0	0	0	1	0	1
Team sports	2	1	0	0	1	4
Dancing	3	0	0	1	1	5
						48 (52%)
Occasional/holiday						
Water sports	5	0	1	0	2	8
Snow sports	6	8	1	0	0	15
Fishing	0	2	0	4	0	6
Horse-riding	4	4	2	0	0	10
Live event/concert	2	0	0	0	3	5
						44 (48%)

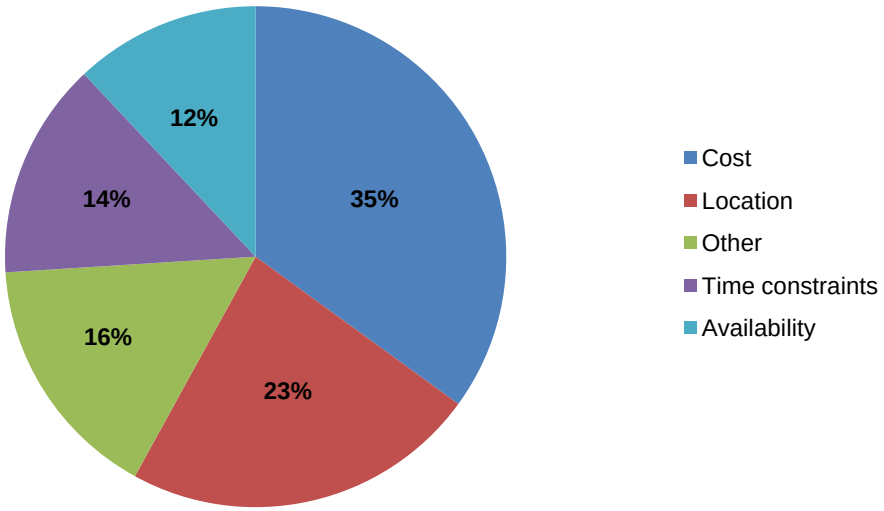

Figure 7.2: *Percentage of responses to perceived barriers.*

DISCUSSION

The procedures used in this study are feasible to investigate preferred choice of physical activity in independently walking children with cerebral palsy and the potential barriers to activity as perceived by their parents. Participants were able to be recruited through the major cerebral palsy service provider in Sydney, the Cerebral Palsy Alliance. Staff from the Cerebral Palsy Alliance identified potential participants who were likely to be interested in participating in the study. In addition, as the testing for the study was conducted close to participant's homes, or in their usual physiotherapy intervention environment, it was considered less likely that there would be issues with retention. In this study, the children and their parents, who had participated in the study reported in Chapter 6, were asked questions in a face-to-face format on their preferred choice of physical activity. However in a future study, telephone interviews or a mailed questionnaire could be used.

The questions asked of the children and their parents were a reflection of the type of physical activities which the child might be engaged in either alone, or with other people, and also within their home or community. All activities, apart from martial arts, were reported as being currently performed by the majority of the children. The children and their parents readily answered the questions about the activities they were engaged in. In addition, the children and their parents did not identify any other physical activities over and above the list of 20 provided. Interestingly, the parents appeared keen to talk about the barriers which were preventing their child being engaged in more physical activities.

This study showed that children with cerebral palsy who are mildly impaired (Level I/II GMFCS) are engaged in a range of physical activities which are representative of age-matched typically-developing children. In addition, the high level of agreement about preferred activities between parents and children showed that the parents were well aware of the types of physical activities their child would like to be engaged in, and also how much they would like to be doing these activities. However, parents perceived many barriers which were either preventing their child from being engaged in an activity, or from performing the activity more frequently.

The common barriers to activity perceived by parents included cost, location, availability, time constraints and safety issues. Cost was perceived as the greatest barrier to activity with 35% of the responses being in this category. The greatest response was seen for activities such as horse-riding, water and snow sports. However, these are activities which have high costs associated with them. Cost may also be a consideration for parents who would like their child to be engaged in a structured activity program, such as gym membership, or YMCA program. Membership costs are often the main economic barrier affecting access to recreation and/or fitness facilities for people with disabilities (Rimmer et al 2004).

Location was perceived as the second largest barrier to activity with 23% of the responses being in this category. Parents commented that even if their child was able to be engaged in activities, such as snow sports and horse-riding, the distances required to travel to these activities made them impractical to consider. Likewise, with bike riding or skating, parents reported that they would have to drive to a park,

or similar area, if their child wanted to ride a bike or skate as there was no suitable area to ride at home.

Time constraints were perceived as the next largest barrier to activity, particularly after school hours, with 14% of the responses being in this category. The majority of children (90%) in the study had siblings, and parents reported they were aware of the need for all their children to have equal opportunities for after school activities. Conflicting time commitments between the child with cerebral palsy, and the requirements of other children in the family, meant that many activities were not able to be considered. In addition, it is also recognised that there is likely to be a greater commitment of time and resources for a child with cerebral palsy than with a non-disabled child (Imms et al 2008). Adding to the time constraint barrier was the fact that most parents in the study had either part or full-time employment. A frequent comment was that it was easier for the child with cerebral palsy to play at home with their siblings, rather than having to spend the time travelling to an activity. The burden of providing opportunities for activities for children with cerebral palsy usually falls on the child's family and therefore, physical activities are more likely to be conducted at home rather than in the wider community (Imms et al 2008).

Availability was perceived as the next largest barrier to activity with 12% of the responses being in this category. Commonly, a lack of available facilities has prevented children with cerebral palsy from participating in community-based activity programs (King et al 2003, Buffart et al 2009). Despite significant development of community facilities in metropolitan areas, activity programs for children with disabilities are often hindered by a lack of suitably trained staff (King

et al 2003). Indeed, several parents from the present study commented that their child would like to attend one of the commercially available gyms. However, they could not find one in their local area where instructors felt they could accommodate their child's needs even though their child was ambulatory and was capable of exercising. Parents expressed their interest in being involved in physiotherapy led exercise programs if they were available. Individually tailored exercise programs, can not only improve levels of daily physical activity in people with motor impairment, but perhaps more importantly, can also maintain the level of activity (van der Ploeg et al 2006).

There were also responses that did not easily fit into categories (16%). Safety issues were identified with activities such as gymnastics, bike riding/skating, and also water sports. These parents acknowledged that they were probably being over protective and tended to avoid any activities which they saw as risk taking. Two parents from the study reported that their child's reduced walking capacity (endurance and speed) often affected their family's choice of outing where walking was influenced by distance or time. Interestingly, one of the parents in the group also revealed that physical activity wasn't a big part of their culture. Their family outings tended to be in the form of large gatherings in a park where people were more likely to sit and talk, rather than engage in more physical pursuits. This is not uncommon in some ethnic groups and recreational activities are known to be influenced by family and cultural beliefs (Dwyer et al 2009b).

Most parents are more likely to want their child to be independent and efficient in self-care and mobility before considering recreational or sporting activities (Chiarello

et al 2010). However, some of the parents from the study commented that they often felt their child wasn't extended enough in their daily physical activities, and that the activities they were doing could be done more frequently.

How can barriers be overcome to increase daily activity?

As the children had mild cerebral palsy and were able to walk, it was expected that they should have been able to undertake as much physical activity as their typically-developing peers. When matched to their siblings, this was found to be true for both the amount and intensity of physical activity (Chapter 6) and also the types of activity (this study). However, the amount of activity for both groups falls short of the daily recommended guidelines. In Table 7.4, the activities have been divided into those which were performed more frequently (1-2x/week), and those which were performed infrequently (occasional/holiday). If the barriers to those activities which are performed frequently can be removed, then physical activity is likely to increase.

Cost was identified as being the largest barrier to children carrying out activities which could easily be incorporated into daily life. For example, children wanted to attend a local gym and this could be addressed by encouraging the gyms to provide sliding-fee gym memberships, or special 'scholarships', for children with disabilities which might enable them to attend more frequently. Likewise, instigating special offers such as a 'try-out' or a 'try before you buy' scheme could be offered to children and their families. Similarly, organisations such as the YMCA have a commitment to social inclusion and run programs such as the Healthy Kids Program and Access & Disability Program. Local community and corporate support could be

fostered by promoting sponsorship for children attending some of these programs or facilities. In addition, initiatives to remove barriers to participation for people with disabilities are being proposed in Australia by the National Disability Insurance Scheme (NDIS) (http://fahcsia.gov.au). The aim of this scheme would be to reform the way services are funded and delivered to people with a disability. Children with cerebral palsy should benefit from this scheme as it is intended that it will encompass all ages and levels of disability and the primary focus is on enabling activity and participation within the individual's community environment.

Another activity which could increase overall physical activity are physiotherapy-led programs. For children who attend organisations such as the Cerebral Palsy Alliance in NSW, there are some initiatives to offer exercise programs run by physiotherapists and exercise physiologists. Children usually attend these programs once a week for approximately one hour during the school term. However, these programs are subject to numbers attending and staff availability and so may not always be offered. Utilising students from physiotherapy and exercise science courses to run these programs as part of their clinical placements could address the issue of staffing availability. This could mean that programs could be offered more frequently and also could be promoted more widely to include children without disabilities.

Even for activities which were available locally, parents identified time constraints which prevented their child from engaging in them more frequently. Therefore, the focus could be on more interactive home-based activities. These activities could be externally monitored to ensure compliance and provide feedback to the child. Devices such as a personal digital assistant (PDA) have been used to examine

physical activity and quality of life in children (Floro et al 2009; Bray et al 2010). These devices can be provided to a child to remind them of activities, provide incentives and track their progress while also enabling competition between other children. Externally monitoring activity programs have been reported to have the best outcomes in terms of compliance and are more likely to promote ongoing physical activity which generalise into other free-living activities (Bania et al 2011, Maher et al 2010, van der Ploeg et al 2006). In addition, combining social contacts with physical exercise will have more benefit than performing activity alone (Buffart et al 2009). Therefore, activities could be devised to enable inclusion of the child's family and friends.

Changing physical activity behaviour is a complex and multi-factorial process in both able-bodied and disabled populations. With children, there is an added complexity of age-related preferred activities and the need to structure activities within the context of play. Current research is exploring the type of activities which are most likely to encourage physical activity and participation in children with cerebral palsy (Van Wely et al 2010, Claassen et al 2011). In addition, focus groups, for both children and their parents, are being implemented to investigate the potential barriers to participation in activities (Van Wely et al 2010, Claassen et al 2011).

For the children in this study it would seem that physical impairment is not the main issue which is stopping them from being engaged in the recommended levels of physical activity. Furthermore, while these children were involved in a range of activities, these were not at a level to promote moderate to vigorous amounts of physical activity. In addition, the perceived barriers often meant that a child was not

able to engage in the activity as frequently as they would like. Therefore, an important focus should be on improving physical activities which have few barriers, such as walking, as this is an activity which could be performed every day and can also be used to achieve moderate to vigorous levels of physical activity.

The frequency and magnitude of the perceived barriers were not examined in-depth in this study. However, these are both issues which could be investigated further. Creating a habit of regular physical activity is challenging for individuals with lifelong disabilities (Cicirello et al 2011). However, this challenge should not be made more difficult by barriers which have the potential to be overcome.

CHAPTER 8

DISCUSSION

EVIDENCE FOR PHYSIOTHERAPY INTERVENTION – Study 1

Where were we in 2008?

Where are we now?

Where do we need to go in the future?

EVIDENCE FOR CARDIORESPIRATORY FITNESS TRAINING – Study 2

Where were we in 2008?

Where are we now?

Where do we need to go in the future?

EVIDENCE FOR PHYSICAL ACTIVITY – Study 3

Where are we now?

Where do we need to go in the future?

BARRIERS TO PHYSICAL ACTIVITY – Study 4

Where are we now?

Where do we need to go in the future?

LIMITATIONS OF THE PROJECT

CONCLUSION

The aim of this thesis was to examine the evidence available for physiotherapy intervention for children with cerebral palsy. Despite advances in medical intervention and rehabilitation, cerebral palsy is the most common activity-limiting condition in children and adolescents worldwide. Likewise, the motor impairments and activity limitations associated with cerebral palsy continue to be the primary focus for physiotherapy intervention. Determining the most effective form of physiotherapy intervention to address motor impairments and activity limitations in order to promote physical activity and enable community participation in children with cerebral palsy, is therefore an important issue for physiotherapists.

In this concluding chapter, the main findings are summarised. Since the studies in this thesis have been conducted over some time, an update is given of how the evidence for physiotherapy intervention for children with cerebral palsy has progressed, and future directions are proposed. The limitations of this thesis are outlined and suggested solutions given.

EVIDENCE FOR PHYSIOTHERAPY INTERVENTION – Study 1

Where were we in 2008?

Study 1 was a summary of systematic reviews which identified 8 systematic reviews examining the effects of 6 physiotherapy interventions for children with cerebral palsy. While the reviews were deemed to be of high quality, the low number of randomised controlled trials within each review prevented meta-analysis. Consequently there is little evidence for use by clinicians to direct their practice. In addition, the evidence identified was not convincing as there were contradictory

results at both the impairment and activity levels and no evidence at the level of participation. For example, the evidence in one review that a physiotherapy intervention was beneficial was contradicted by evidence in another review that it had no effect. The strength of evidence was further undermined by the fact that most of the reviews contained studies of other design as well as randomised controlled trials.

Only one trial in one review reported severity of disability as determined by the Gross Motor Function Classification Scale (GMFCS) (Palisano et al 1997) therefore the similarity of the participants between studies could not be assessed. Furthermore, no conclusions could be made regarding the effect of intervention on the level of severity of cerebral palsy and the lack of significant findings could be attributed to the diverse range of abilities within these participants. Including the most appropriate level for the intervention, ie, choosing Level I and/or II GMFCS, for investigation of walking ability may be more appropriate. A further issue was identified with the choice of outcome measures used particularly at the activity level. For example the Gross Motor Function Measure (GMFM) (Russell et al 1989) was developed as a measure of motor performance but in many studies was frequently used to reflect amount of activity. The failure of many studies to find any carryover to activity may be more reflective of the outcome measures used rather than the intervention.

Where are we now?

Given that it is now 2011, it is timely to review where we are up to compared with 2008 when Study 1 summarising systematic reviews was done. For example, a search of the Physiotherapy Evidence Database (PEDro) shows that since 2008, 47 randomised controlled trials have been added to the database. In addition, there are a further 15 systematic reviews examining physiotherapy intervention in children with cerebral palsy. Of interest to know is whether there is now more, and better quality evidence, than there was in 2008.

Overall, there has been an 8% increase from 4.8 to 5.2 in the mean PEDro score of randomised controlled trials since 2008 and 11% (n = 5) are high quality trials which score of 8 out of 10. In the 15 systematic reviews, it is evident that there are still mixed study designs being examined, and only two reviews have included only randomised controlled trials (Butler et al 2010, Scianni et al 2009). Furthermore, within those two reviews there are small numbers of trials (n = 3, 6) and only one meta-analysis.

Where do we need to go in the future?

Given that 8 is the likely maximum PEDro score achievable because it is not usually possible to blind the therapist or the participants in physiotherapy intervention, the quality of recent randomised controlled trials at a mean PEDro score of 5.2 is encouraging. However, in order to develop clear guidelines to direct physiotherapy practice in children with cerebral palsy, there is a need for better quality research. Unless improving quality is a priority, it is likely the results of systematic reviews

will continue to provide little evidence to direct physiotherapy intervention. Once high quality trials are undertaken, the next step is to combine randomised trials into systematic reviews and perform meta-analyses to determine the highest level of evidence for the effect of physiotherapy intervention. Systematic reviews which only include randomised controlled trials provide the most robust evidence. Of course, high quality evidence is not always available or even possible in some areas of physiotherapy intervention for children with cerebral palsy. In these instances there will always be a place for case-control studies and clinical expertise to guide physiotherapists in their clinical decision-making. However, where high quality evidence is available, this should be used.

Several issues arise from this study which could be applied to future trials. For example, only conducting randomised controlled trials which investigate an intervention versus nothing, or a placebo, may provide more compelling evidence than uncontrolled trials at the level of impairment and activity. Furthermore, one of the challenges is to address physiotherapy intervention across all levels of ability in cerebral palsy. However, evidence of effect of intervention is more likely to be seen if different levels of the GMFCS are examined separately. In particular, those children who are classified as Level I/II and can walk should be examined separately from those children at Levels III-V who require aids for mobility.

To be able to perform meta-analyses, the emphasis in trials needs to be on using a common set of outcome measures which are reflective of the intervention being examined, eg, the 6-minute walk test or the 10-metre walk test. This issue requires further examination particularly when determining activity limitations for children

with cerebral palsy. It is not enough for only common outcome measures to be used, but that the measure is also appropriate for the variable being examined. In 50% (12 out of 24) of the trials in Study 1, the Gross Motor Function Measure (GMFM) (Russell et al 1989) was used to measure activity. Despite the developers describing this as a 'performance measure', it continues to be used as a measure of capacity of children with cerebral palsy. This may in part be due to a recent study (Nordmark et al 2000) which describes the GMFM as a measure of the child's "best ever performance" (Nordmark et al 2000) and is therefore a valid measure of capacity. To date, the 6-Minute Walk Test is the only one which has been validated as a measure of walking capacity in children with cerebral palsy (Maher et al 2008).

EVIDENCE FOR CARDIORESPIRATORY FITNESS TRAINING – Study 2

Where were we in 2008?

In 2008 there were 3 randomised controlled trials which examined cardiorespiratory fitness in children with cerebral palsy and these were reviewed in Study 2. However, there was insufficient data in the trials to perform a meta-analysis. Therefore, the results of the between-group analyses were reported individually. After a short term (2-4 mth) and long-term (8-9 mth) intervention, there was a statistically-significant 20% increase in cardiorespiratory fitness for the short-term intervention and a statistically-significant 34% increase in cardiorespiratory fitness for the long-term intervention. Although these results are positive, there was no evidence of carryover into a reduction in activity limitations.

Where are we now?

Since 2008, there has been one randomised trial which examined the effect of cardiorespiratory fitness training in children with cerebral palsy (Fowler et al 2010). This study included 55 participants, male and female, aged 7-18 yr who had spastic diplegia and were classified Levels I-III Gross Motor Function Classification Scale (GMFCS). Participants were randomised into either a cycling group (n = 27) or a non-cycling control group (n = 28). The experimental group performed cycling on a stationary bicycle 3/wk for 3 mth at an intensity of 70-80% of maximum heart rate whereas the control group had no intervention. The outcome measure for cardiorespiratory fitness was the 600-yard Walk-Run Test, and the outcome measure for activity was the Gross Motor Function Measure (GMFM). A non-significant ($p = 0.24$) between-group mean improvement for cardiorespiratory fitness of the experimental group of 3.1 m/min was reported which represents a 4% increase in fitness. A non-significant between-group mean improvement ($p = 0.23$) for activity of the experimental group of 0.7 points out of 66 was reported which represents a 1% increase in activity.

The non-significant findings found in an additional trial (Fowler et al 2010) perhaps highlight the complexity of conducting research in children with cerebral palsy of varying severity and while growth and development is occurring concurrently. Indeed, there may be a number of ways in which this study differs from the three randomised controlled trials included in Study 2. First, the severity of cerebral palsy is variable and there was a higher ratio of Level III participants to Level I and II. Given that children with Level III have greater motor impairment, it is possible that when their results were pooled with the Level I and II participants, they lowered the

group results since the 600-Yard Walk Test scores for the Level III participants would have been confounded by their motor impairments.

Second, the age range of the participants (7-18 yr) was large. Because children's cardiorespiratory response to exercise is dependent on the maturation of their lungs, it may have been better to have separated the participants into pre and post-adolescent ages and analysed their results separately. Indeed, the authors reported double the number of children in the lower age group (7-11 yr) and therefore, this may have lowered the overall results.

Third, the 600-Yard Walk test is a sub-maximal field test which gives an indirect measure of VO_{2peak} via a prediction of resting heart rate and is seen to be indicative of cardiorespiratory conditioning (Washington et al 1994). However, VO_{2peak} may be under estimated by field tests (Artero et al 2011) and therefore the results could actually have been higher than reported.

All four randomised controlled trials of cardiorespiratory fitness training failed to find any improvement in activity. This may be due to the GMFM being used as a measure activity. Given that the GMFM is a measure of motor performance, and not of motor capacity, it is not a measure which is reflective of cardiorespiratory fitness. Therefore, using a measure such as the 6-Minute Walk Test may have given better results. In conclusion, the level of evidence in 2011 for increasing cardiorespiratory fitness in children with cerebral palsy is little different than it was 3 years ago.

Where do we need to go in the future?

The intervention used in the four trials to improve cardiorespiratory fitness was intense enough to produce a training effect in children with cerebral palsy. To determine categorically if a training effect is possible, it would be helpful if a large adequately powered trial which included a direct measure of cardiorespiratory fitness (VO_{2peak}) was conducted. In addition, to determine if the intervention has an effect at the activity limitation level, it would be useful if measures of activity that reflect fitness, eg, 6-Minute-Walk Test were collected. Furthermore, in the first instance, conducting a randomised trial only of children with cerebral palsy who are Level I/II GMFCS, would provide evidence for effect of cardiorespiratory training without the confounding issues of severe disability.

EVIDENCE FOR PHYSICAL ACTIVITY – Study 3

Where are we now?

In Study 3, the amount of moderate to vigorous physical activity performed by ambulant children with cerebral palsy was examined and compared to their age-matched peers. These children had mild impairments which would theoretically enable them to engage in a normal amount of physical activity. The findings from this study showed that children with cerebral palsy were capable of performing moderate to vigorous physical activity. They were spending a similar amount of time each day in physical activity to their peers. However, the amount of daily physical activity for both groups was less than the recommended guidelines (Department of Health and Ageing 2005) of 60 minutes moderate to vigorous activity.

There are no other studies using accelerometers which compare physical activity with typically-developing peers. However, while this study was being carried out, two studies (Capio et al 2010; Clanchy et al 2011) which examined moderate to vigorous physical activity in children with cerebral palsy using an accelerometer were published. The period of activity measured in both these studies was relatively short, ie, 15 minutes in the Clanchy et al (2011) study, and 22 minutes total in the Capio et al (2010) study. The activity measured was walking, circuit-type activities and free play. In line with Study 3, the findings from these two studies showed that children with cerebral palsy were able to achieve moderate to vigorous amounts of physical activity.

Where do we need to go in the future?

The results from this study have provided some interesting data, but completing the study to the intended cohort of 20 participants would add weight to this data. Since 8 out of 9 children tested on the 6-Minute Walk Test were able to achieve moderate to vigorous levels of physical activity, this could be a starting point to increase their physical activity at home. For example, a simple competition where family members compete against each other to improve the distance gained over the 6-Minute Walk Test might boost the activity levels in all family members. Likewise, varying this activity into other activities which have a component of 'racing the clock' would enhance the motivation to perform more activity. Similarly, pedometer-based gait training has been shown to be effective in improving walking speed in children with cerebral palsy (Hamed & Abd-elwahab 2011). By wearing devices like a pedometer or accelerometer the child may be more motivated to perform physical activity. In

future research, these types of strategies could be tested in a randomised controlled trial in order to see whether they are effective in increasing physical activity.

Of interest, would be to compare children with cerebral palsy throughout Australia for similarities or differences in the amount of their physical activity. A study of a larger cohort could pave the way for some national initiatives on guidelines for physical activity in children with cerebral palsy. Likewise, extending this comparison globally may be an opportunity to develop links with others who are already investigating physical activity in these children. For example, a group of researchers are part of a Dutch government initiative – LEARN 2 MOVE research program (Hielkema et al 2010, Ketelaar et al 2010, Slaman et al 2010). The aim of these researchers is to improve physical activity in children with cerebral palsy through a physical activity stimulation and cardiorespiratory fitness training program. In addition, a research group in Canada are developing a program to promote physical activity and encourage an active lifestyle in children with cerebral palsy (Claassen et al 2011).

BARRIERS TO PHYSICAL ACTIVITY – Study 4

Where are we now?

This study examined the barriers to physical activity in children with cerebral palsy from the parents' perspective. These children had mild impairments which would theoretically enable them to engage in a normal amount of physical activity. The findings from this study showed that children with cerebral palsy were regularly engaged in the type of physical activities which are fairly representative of typically-

developing children. However, parents perceived many barriers which were either preventing their child from being engaged in physical activity, or from performing physical activity more frequently. The cost of activities was perceived by parents as being the greatest barrier to more frequent participation in activities.

Where do we need to go in the future?

For children with cerebral palsy who have mild impairment and are able to walk (Level I/II), there is a need to increase the amount of their physical activity. Specifically there is a need to increase the amount and frequency of their moderate to vigorous activity. Since cost was one of the main barriers preventing these children from performing more regular activities, then exploring the options for reducing the cost of activities which could be performed more frequently, ie, gym membership and local physical activity programs such as those run by the YMCA, is warranted. Similarly, there is a need to implement more physiotherapy-led activity programs for children with cerebral palsy in their communities. Promoting physical activity in all populations is a health priority and for children with cerebral palsy this is an opportunity for activity which is yet to be developed. Furthermore, initiating programs which can be externally monitored for support and progression of activities will most likely increase the motivation of children to perform home-based activities.

LIMITATIONS OF THE PROJECT

In Study 1, only reviews which had randomised controlled trials were to be included, and while the reviews which were selected were deemed to be of high quality, there were only two reviews which contained only randomised controlled trials. Therefore, reviews which reported randomised controlled trials separately were included even though they also contained mixed methods (single-case studies, within group comparisons, cohort studies). Although the author's conclusions were made on studies of mixed design, this problem was overcome to some extent by also examining the results of the randomised controlled trials alone.

In Study 2, there were only three randomised controlled trials which could be included for analysis of cardiorespiratory fitness. Unfortunately, the poor reporting in some trials (in terms of insufficient data provided) meant that a meta-analysis was not possible. However, all the trials reported positive results so it could at least be concluded that fitness training was effective but as a meta-analysis could not be performed the size of the effect could not be determined.

In Study 3, the small numbers of participants mean that the results can only be considered as preliminary. However, it is intended that this study continues with further data collection to enable publication. The model of accelerometer used was not water proof and water-based activity was not able to be captured. This issue was overcome by measuring activity over 7 days to be reflective of the overall amount of activity. Not all the children wore the accelerometer for the 7 days and therefore there was incomplete data. Despite this, there was still 90% of the data which was available to be analysed. Two children did not have respiratory gas analysis as they

were unable to wear the Cosmed K4b2™ mask. However, this was accounted for by using the results from other children with similar exercise test times to equate to similar levels of VO_{2peak} for l/min and ml/kg/min respectively. Finally, it is recognised that it is difficult to encourage children to perform a maximal exercise test to exhaustion. Therefore, their VO_{2peak} results may not be a true reflection of their maximal oxygen uptake potential. However, the moderate correlation between peak heart rate and oxygen uptake during the progressive exercise test suggests that the children had a normal cardiorespiratory response to exercise of increasing intensity.

In Study 4, it is recognised that there may have been potential bias of responses. Bias may have occurred as a result of children and parents endeavouring to be 'good' respondents by over or underestimating the number of activities they were engaged in. In addition, they may have been tempted to give what they saw as the 'right' answer to some of the questions. However, asking the questions separately reduced the likelihood of children and parents overhearing each other's responses. The physical activities chosen were intended to be reflective of a mix of activities which children might commonly be engaged in. Indeed, all but one of the physical activities was reported as being currently performed, or had been performed by the children in the preceding 12 months. In addition, the children and their parents did not identify any other physical activities over and above the list of 20 provided. However in future studies, a broader range of physical activities could be investigated using assessments such as the Children's Assessment of Participation and Enjoyment (CAPE)(King et al 2004). Furthermore, not all barriers to activity might have been addressed in the study. Although parents were given the

opportunity to identify any other barriers which had not been given as examples, there may have been others that they did not want to disclose, eg, personal factors. Finally, the frequency and magnitude of barriers was beyond the scope of this study but could be examined in future studies.

CONCLUSION

Overall, the evidence for physiotherapy intervention for children with cerebral palsy is limited and there is clearly a need for improved quality in research. However, research into cerebral palsy is challenging, not only because of severity of impairments and activity limitations, but also the research is undertaken while growth and development are occurring. High quality research involving children with cerebral palsy requires commitment and adherence by not only the child, but also the family, and at times the child's local community.

Generally, children with cerebral palsy just want to engage in activities alongside their typically-developing peers. The ongoing challenge for physiotherapy intervention for these children is to find the best intervention which will enhance their level of activity and community participation. For those children with more severe motor impairment, this challenge is always more difficult. However, for children who are ambulant and capable of physical activity, it is important that they are given the opportunity to engage in regular activity which is performed at a moderate to vigorous level. Furthermore, overcoming the barriers which are preventing physical activity means that there is a greater chance that children with cerebral palsy will have a more active lifestyle.

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APPENDIX A

CRITERIA FOR SCIENTIFIC QUALITY OF REVIEW ARTICLES

(Hoving et al 2001)

Search methods

1. ***Were the search methods used to find evidence (primary studies) on the primary question(s) stated?***

2 points: Yes; includes description of databases searched, search strategy, and years reviewed. Described well enough to duplicate
1 point: Partially; partial description of methods but not sufficient to duplicate search
0 points: No; no description of search methods

2. ***Was the search for evidence reasonably comprehensive?***

2 points: Yes; must include at least one computerized database search and a search of unpublished or non-indexed literature (for example, manual searches or letters to primary authors)
1 point: Can't tell; search strategy partially comprehensive (for example, at least one of the strategies in the foregoing section were performed)
0 points: No; search not comprehensive or not described well enough to make a judgment

Selection methods

3. ***Were the criteria used for deciding which studies to include in the review reported?***

2 points: Yes; inclusion and exclusion criteria clearly defined
1 point: Partially; reference to inclusion and exclusion criteria can be found in the article but are not clearly defined enough to duplicate
0 points: No; no criteria defined

4. ***Was bias in the selection of articles avoided?***

2 points: Yes; key issues influencing selection bias were covered. Two of three of the following bias avoidance strategies were used; two or more assessors independently judged study relevance and selection using predetermined criteria, reviewers were blinded to identifying features of study (ie journal title, author(s), funding source) and assessors were blinded to treatment outcome.
1 point: Can't tell; if only one of the three strategies above were used.
0 points: No; selection bias was not avoided or was not discussed.

Validity assessment

5. ***Were the criteria used for assessing validity of the studies that were reviewed reported?***

2 points: Yes; criteria defined explicitly.
1 point: Partially; some discussion or reference to criteria but not sufficiently described to duplicate.
0 points: No; validity or methodological quality criteria not used or not described.

6. Was the validity for each study cited assessed using appropriate criteria (either in selecting studies for inclusion or in analyzing the studies that were cited)?
2 points: Yes; the criteria used address the major factors influencing bias (eg population, intervention, outcomes, follow-up)
1 point: Partially; some discussion of the methodological review strategy but not clearly described with predetermined criteria.
0 points: No; criteria not used or not described.

Synthesis
7. Were the methods used to combine the findings for the relevant studies (to reach a conclusion) reported?
2 points: Yes; qualitative or quantitative methods are acceptable
1 point: Partially; partial description of methods to combine and tabulate; not sufficient to duplicate.
0 points: No; methods of combining studies not stated or described.

8. Were findings of the relevant studies combined appropriately relative to the primary question the review addresses?
2 points: Yes; combining of studies appears acceptable.
1 point: Can't tell; should be marked if in doubt.
0 points: No; no attempt was made to combine findings; should be marked if a summary (general) estimate was given anywhere in the abstract, the discussion, or the summary section of the paper, and the method of deriving the estimate was not described, even if there is a statement regarding limitations of combining the findings of the studies reviewed.

9. Were the conclusions made by the author(s) supported by the data or analysis reported in the review?
2 points: Yes; data not merely citations, were reported that support the main conclusions regarding the primary question(s) that the overview addresses.
1 point: Partially
0 points: No; conclusions not supported or unclear.

APPENDIX B

SEARCH STRATEGY FOR CARDIORESPIRATORY FITNESS

Databases: AMED, CINAHL, CDSR, ACP Journal Club, DARE, CCTR, CLCMR, CLHTA, CLEED, EMBASE, Ovid MEDLINE(R) Search Strategy, Web of Science**.**

The search of the databases was optimised by using the terms recommended by the Cochrane Collaboration for the participants (cerebral palsy), and using the terms used in previous Cochrane reviews for the intervention (children, adolescents, cardiorespiratory fitness, exercise [training], physical training, physical fitness, physical activity, aerobic training, exercise tolerance) and outcomes (fitness, activity).

1. muscle spasticity.mp.
2. spastic$.tw.
3. cerebral palsy.mp.
4. cerebral pals$.tw.
5. hemiplegia.mp.
6. quadriplegia.mp.
7. hemiplegi$.tw.
8. monoplegi$.tw.
9. triplegi$.tw.
10. quadriplegi$.tw.
11. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10
12. randomized controlled trial.pt.
13. randomized controlled trials.sh.
14. random allocation.sh.
15. controlled clinical trial.pt.
16. 12 or 13 or 14 or 15
17. (exercise therapy or physical fitness or sports or exertion or locomotion).mp.
18. cardiorespiratory fitness training.mp.
19. ((exercise or circuit or aerobic or cardio* or fitness or physical) adj1 (therap* or train* or program* or intervention* or protocol* or activit* or regime* or group* or class*)).mp.
20. (physiotherapy or locomotion).mp.
21. (sport* or athletic* or leisure activit* or recreation).mp.
22. (jog or jogging or (skate or skating) or (ski or skiing) or soccer or (swim or swimming) or (walk or walking)).mp.
23. (baseball or basketball or bicycling or (cycle or cycling) or boxing or football or golf or gymnastics or hockey or martial arts or mountaineering or tennis or netball or squash or (run or running)).mp.
24. (aquatic or (surf or surfing) or abseiling or (dance or dancing) or rowing or calisthenincs).mp.
25. (exercise tolerance or exercise test or physical endurance).mp. [mp=title, original title, abstract, name of substance word, subject heading word]
26. early ambulation.mp.
27. sports equipment.mp.

28. (isometric contraction or isotonic contraction).mp. [mp=title, original title, abstract, name of substance word, subject heading word]
29. (physical adj3 (exercise$ or therap$ or conditioning or activit$ or fitness)).tw.
30. or/17-29
31. 11 and 16 and 30
32. (child or adolescent).mp. [mp=title, original title, abstract, name of substance word, subject heading word]
33. 31 and 32
34. remove duplicates from 35

DATABASE: PEDro

Search strategy: Advanced
Abstract and Title: Cerebral palsy
Therapy: Fitness training
Subdiscipline: Paediatrics
Method: Clinical trial

APPENDIX C
RECRUITMENT ADVERTISEMENT

The University of Sydney

NSW 2006 AUSTRALIA

Louise Ada
BSc, GradDipPhty, PhD
Associate Professor – Physiotherapy
Faculty of Health Sciences

Cumberland Campus

PO Box 170
Lidcombe NSW 1825
Telephone: 9351 9544
Facsimile: 9351 9278
Email: louise.ada@sydney.edu.au

EXCITING OPPORTUNITY TO VOLUNTEER

Are you:

- **An 8-12 year old with cerebral palsy?**
- **Able to walk independently? (without a walker)**
- **Want to know how active you are?**

We are investigating the amount of physical activity in which children with cerebral palsy engage during a typical week.

Initial data collection will be performed at the Marconi Centre at Prairiewood, the McLeod Centre at Allambie Heights or The University of Sydney Faculty of Health Sciences campus, Lidcombe.

If you are interested in participating, please call Jane Butler for more information on 9351 9265. Please leave a message if I am not there and I will get back to you.

APPENDIX D

EXPLANATION OF PHYSCIAL ACTIVITY STUDY

WHAT IS THE STUDY ABOUT?

- In our study we are looking at physical activity in children with cerebral palsy – how active your child is. We are asking children to participate if they are aged between 8-12 years.

WHAT DO YOU WANT MY CHILD TO DO?

- Two days for testing – approx. ½ hour each time eg 2 Mondays or 2 Tuesdays etc – day, time and location to suit you (usually at either Allambie, Ryde, Prairiewood, Kingswood or Sydney University at Lidcombe)

DAY 1:

- Measurement of joint range, strength and a leg coordination activity
- Walk on a treadmill for as long as they can (usually about 10-15mins) - they will be given a practice session on the treadmill before commencing the test
- While on the treadmill, we would like your child to wear a mask which covers their nose and mouth to see how much oxygen they are breathing – (see picture)
- Wear a monitor on their waist which measures how much activity they are doing – (see picture). They put this on when they wake up and take it off at bedtime. They will wear this for 7 days until they come back to see me.
- Fill in an Activity Diary for 4 days – Friday, Saturday, Sunday, Monday.

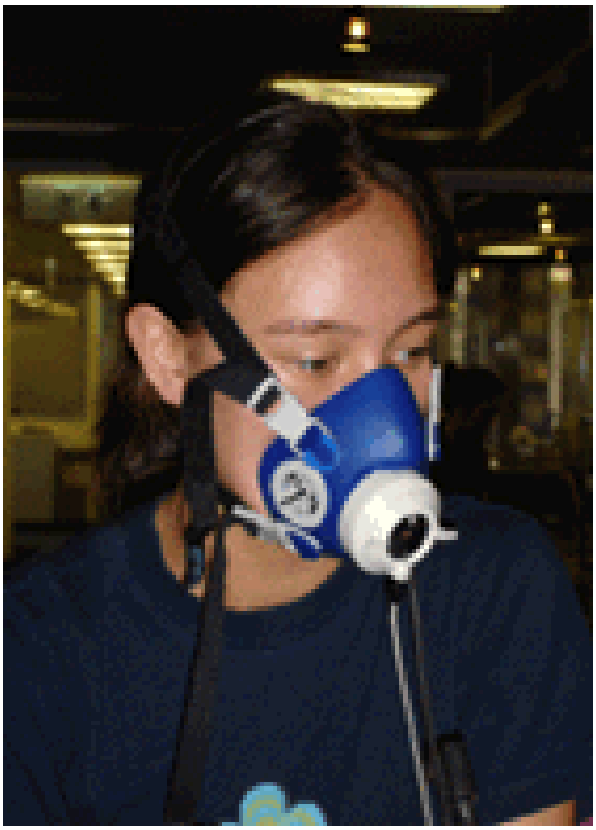
DAY 2: (One week later)

- Return the activity monitor and the Activity Diary and complete a walking test for 6 minutes – this measures how far your child can walk in 6 minutes.
- You and your child will be asked to fill in a questionnaire telling us the type of activities your child likes to do
- We would also like to collect data on the activity monitor only from a sibling if your child has one in the age range? They would only wear the monitor and not participate in the rest of the test.

CONTACT:
Jane Butler
The University of Sydney
Tel 9351 9265
Jane.Butler@sydney.edu.au

There is a more detailed Information Sheet which we will give you to read before committing to the study and we will also explain the study again before beginning any testing.

Mask to measure oxygen while on the treadmill only



Activity monitor to wear for 7 days

APPENDIX E

DATA COLLECTION SHEET – PARTICIPANT

Name:_____ **Date:**_____

Parent/Caregiver:_____

Address:_____

_____**Postcode:** _____

Phone: (H)_____ **(M**):_____

Email:_____

Gender: M/F

DOB:_____**Age:**____

Height:_____ (cm)

Weight:_____ (kg)

CP classification:_____

GMFCS level: I II

CP motor type: Spas Atax Dyst Hypo Unknown Mixed

School year_____

Class_____

Type of school attended: M S DU O

Mobility aids used: y/n home/school/community

Family type: 2 parent single parent

Primary caregiver's place of birth: Aust other

Other comments:

IMPAIRMENTS

MEASUREMENT OF IMPAIRMENT LOWER LIMB(S)

Strength (D/F): **Right:** 1 2 3 4 5

Left: 1 2 3 4 5

Dexterity (15s): **Right** _____ # of taps during LEMOCOT

_____ #/min

Left _____ # of taps during LEMOCOT

_____ #/min

Spasticity (P/F): **V1** **R)** 0 1 2 3 4 **L)** 0 1 2 3 4

V3 **R)** 0 1 2 3 4 **L)** 0 1 2 3 4

Contracture (P/F): **R)** 1 2 3 **L)** 1 2 3
(1=< 90°, 2 = 90°, 3 >90°

ACITIVITY LIMITATIONS

6-MINUTE WALK TEST

Date:

Time:

Pre-test heart rate: **Post-test heart rate:**

# of rests:_____

Number of laps: _____

Distance walked: _____

VO_{2max} TEST

Stage	Time (min)	Treadmill speed (km/hr)	Slope
1			2%
2			2%
3			2%
4			2%
5			2%
6			2%
7			2%
8			2%
9			2%
10			2%

Pre-test heart rate: **Post-test heart rate:**

VO_{2max} _____

PHYSICAL ACTIVITY

Actigraph # :_____

Date on:_____ **Time on:**

Date off: _____ **Time off:**

Comments:

APPENDIX F

DATA COLLECTION SHEET – SIBLING

Parent/caregiver:_____

Child:_____

Address:_____

_____**Postcode:** _____

Phone: (H)_____**(M**):_____

Email:_____

Gender: M/F

DOB:_____**Age:**____

Height:_____ (cm)

Weight:_____ (kg)

School year_____

Class_____

Type of school attended: M S DU O

Family type: 2 parent single parent

Primary caregiver's place of birth: Aust other

Other comments:

Actigraph # :_____

Date on:_____ **Time on:**_____

Date off: _____ **Time off:**_____

Comments:

APPENDIX G
INFORMATION SHEET

The University of Sydney

Discipline of Physiotherapy

Faculty of Health Sciences

ABN 15 211 513 464

LOUISE ADA
ASSOCIATE PROFESSOR

Room O218
Building O Code C42
University of Sydney NSW 2006
AUSTRALIA
Telephone: +61 2 9351 9544
Facsimile: +61 2 9351 9278
Email: louise.ada@sydney.edu.au
Web: www.usyd.edu.au/

PARENT INFORMATION STATEMENT
Research Project

Title: Physical activity in children with cerebral palsy

(1) What is the study about?

You and your child are invited to take part in a research study investigating the amount of physical activity in children with cerebral palsy. The aim of this study is to find out what children with cerebral palsy do in a typical day.

(2) Who is carrying out the study?

The study is being conducted by Jane Butler and will form the basis for the degree of PhD at the University of Sydney under the supervision of Associate Professor Louise Ada.

(3) What does the study involve?

If you agree to participate in this study, you and your child will need to come to either the Discipline of Physiotherapy, University of Sydney, or the Marconi Centre, Prairiewood, or the McLeod Centre at Allambie Heights. Your child will be asked: to walk across a room several times to measure walking performance, a measure of routine physical fitness on a treadmill will be performed, some measures will be done to determine motor ability and your child will be asked some questions about their perception of the amount of physical activity they do each week.

In the second part of the study, your child will be asked to wear a small device around their waist for 7 days (during waking hours) to measure how active they are. The device is lightweight and can be worn comfortably around the waist. The device should be removed for swimming, showering or any occasion where it may come in contact with water. You will be given a sheet with instructions for use of the device and will be shown how to apply and remove the device. In addition, with your permission, one of us may observe your child in the community for 1-2 days (for about six hours) to see what activities they regularly perform.

(4) How much time will the study take?

The initial data collection will take approximately 2 hours.

(5) Can I withdraw from the study?

(a) Being in this study is completely voluntary - you are not under any obligation to consent and - if you do consent - you can withdraw at any time without affecting your relationship with the University of Sydney.

Your decision whether or not to permit your child to participate will not prejudice you or your child's future relations with the University of Sydney. If you decide to permit your child to participate, you are free to withdraw your consent and to discontinue your child's participation at any time without affecting your relationship with the University of Sydney.

(b) Withdrawal from the study will not affect your relationship with receiving services from The Spastic Centre now or in the future.

(6) Will anyone else know the results?

All aspects of the study, including results, will be strictly confidential and only the researchers will have access to information on participants.

(7) Will the study benefit me?

While we intend that this research study may further medical knowledge and may improve physiotherapy practice in cerebral palsy, it may not be of direct benefit to you or your child.

(8) Can I tell other people about the study?

You may tell other people about your involvement in this study.

(9) What if I require further information?

When you have read this information, Jane Butler will discuss it with you further and answer any questions you may have. If you would like to know more at any stage, please feel free to contact Jane Butler Tel 9351 9265 jane.butler@sydney.edu.au

(10) What if I have a complaint or concerns?

Any person with concerns or complaints about the conduct of a research study can contact the Manager, Ethics Administration, University of Sydney on (02) 8627 8175 (Telephone); (02) 8627 8180 (Facsimile) or gbriody@usyd.edu.au (Email).

This study has been approved by The Spastic Centre Human Research Ethics Committee. If you have any complaints or reservations about the ethical conduct of this research you may contact the Ethics Committee on (02) 9479 7200 or ethics@tscnsw.org.au

This information sheet is for you to keep

APPENDIX H CONSENT FORM

The University of Sydney

Discipline of Physiotherapy
Faculty of Health Sciences

ABN 15 211 513 464

LOUISE ADA
ASSOCIATE PROFESSOR

University of Sydney NSW 2006
AUSTRALIA
Telephone: +61 2 9351 9544
Facsimile: +61 2 9351 9278
Email: louise.ada@sydney.edu.au
Web: www.usyd.edu.au/

PARENTAL (OR GUARDIAN) CONSENT FORM

I, ... agree to permit .....……....., who is

aged ..... years, to participate in the research project –

TITLE: **ACTIVITY IN CHILDREN WITH CEREBRAL PALSY**

In giving my consent I acknowledge that:

1. I have read the Information Statement and the time involved for my child's participation in the project. The researcher/s has given me the opportunity to discuss the information and ask any questions I have about the project and they have been answered to my satisfaction.

2. I understand that I can withdraw my child from the study at any time without prejudice to my or my child's relationship with the researcher/s now or in the future.

3. I understand that withdrawal from the study will not affect my relationship with receiving services from The Spastic Centre now or in the future.

4. I agree that research data gathered from the results of the study may be published provided that neither my child nor I can be identified.

5. I understand that if I have any questions relating to my child's participation in this research I may contact the researcher/s who will be happy to answer them.

6. I acknowledge receipt of the Information Statement.

7. I give permission for my child's everyday activities to be observed by one of the researchers YES ☐ NO ☐

.....	.....……
Signature of Parent/Guardian	Signature of Child
.....	.....……
Please PRINT name	Please PRINT name
.....	.....……
Date	Date

APPENDIX I
GRADING OF THE TARDIEU SCALE

Tardieu Scale
This test is performed with the patient in the supine position, with the head in midline.
Measurements take place at 3 velocities (V1, V2, and V3).
Responses are recorded at each velocity as X/Y, with X indicating the 0 to 5 rating, and Y indicating the degree of angle at which the muscle reaction occurs.
By moving the limb at different velocities, the response to stretch can be more easily gauged since the stretch reflex responds differently to velocity.

Velocities:
V1: As slow as possible, slower than the natural drop of the limb segment under gravity
V2: Speed of limb segment falling under gravity
V3: As fast as possible, faster than the rate of the natural drop of the limb segment under gravity

Scoring:
0 No resistance throughout the course of the passive movement
1 Slight resistance throughout the course of passive movement, no clear catch at a precise angle
2 Clear catch at a precise angle, interrupting the passive movement, followed by release
3 Fatigable clonus with less than 10 seconds when maintaining the pressure and appearing at the precise angle
4 Unfatigable clonus with more than 10 seconds when maintaining the pressure and appearing at a precise angle
5 Joint is immovable

Grading is always performed at the same time of day, in a constant position of the body for a given limb. Other joints, particularly the neck, must also remain in a constant position throughout the test and between tests. For each muscle group, reaction to stretch is rated at a specified stretch velocity with two parameters, X and Y

Velocity of stretch
Vi: As slow as possible (minimizing stretch reflex).
V2: Speed of the limb segment falling under gravity.
V3: As fast as possible (faster than the rate of the natural drop of the limb segment under gravity).
Vl is used to measure the passive range of motion. Only V2 or V3 are used to measure spasticity.
Quality of the muscle reaction (X)
0: No resistance through the course of the passive movement.
1: Slight resistance throughout the course of the passive movement with no clear catch at a precise angle.
2: Clear catch at a precise angle, interrupting the passive movement, followed by release.
3: Fatigable clonus (<10 s when maintaining pressure) occurring at a precise angle.
4: Unfatigable clonus (>10 s when maintaining pressure) occurring at a precise angle.

Taken from Patrick E & Ada L (2006)

APPENDIX J

INSTRUCTION SHEET FOR THE *ACTIGRAPH*® ACTIVITY MONITOR

The *Actigraph*® is a device which works like a pedometer to measure the physical activity of your child. This device is very safe. It is a small, light box that is about the size of a matchbox. The device is generally worn under your child's clothes so that it will not be seen.

The *Actigraph*® device needs be worn for seven (7) consecutive days so we will have information on what your child does on a weekday and on weekends. During the period of monitoring you and your child should continue with your usual daily routines.

We have included for you written instructions on when the *Actigraph*® should be worn and how to remove and attach the *Actigraph*® on your child.

When should the Actigraph® be worn?

The *Actigraph*® will be required to be worn from

_____, 2010

till _____, 2010.

The *Actigraph*® should be put on as soon as your child is awake in the morning.

Removal of the belt should only occur when your child is:

- Having a nap or going to bed;
- Having a bath; and/or
- Participating in other water activities such as swimming lessons.

*****IMPORTANT: DO NOT GET DEVICE WET*****

Remember to always put the *Actigraph*® back on your child as soon as possible after waking and/or washing/swimming

Placement of the Actigraph®

When placing the *Actigraph®* on your child:

1. Kneel facing your child (who is standing)

2. Hold the device with the white label facing your child's body and the star sticker visible on the top

3. Place the device slightly above the right hip

4 .Secure the device by closing the safety clasp on the adjustable stretch belt
NB: the belt should be firmly in place but should not cause any discomfort to your child

IMPORTANT: DO NOT GET DEVICE WET!

Should any problems arise while you are wearing the *Actigraph®* please contact one of the researchers immediately.

Louise Ada	**or**	**Jane Butler**
Tel: 9351 9544		**Tel: 9351 9265**
Mob: 0409 032 554		**Mob: 0418 233 190**

APPENDIX K
ACTIVITY RECORD BOOK

PHYSICAL ACTIVITY STUDY – THE UNIVERSITY OF SYDNEY

ACTIVITY RECORD BOOK

Name:_____

We would like you to keep a record of your activity for 4 days while you are wearing the Actigraph. We want you to do this so that we can know what you were doing while you were wearing the Actigraph.

Starting on: Friday morning_____

Finishing on: Monday evening _____

Instructions for filling in each day of the activity record are on the following pages.

Day 1: _____ Day: Friday

When were you active?	What did you do?	Were you puffed?
Before school		No A bit A lot
How did you get to school?	Walk/bike/bus/car/other	No A bit A lot
Recess		No A bit A lot
Lunch time		No A bit A lot
Physical Education or sport		No A bit A lot
How did you get home from school?	Walk/bike/bus/car/other	No A bit A lot
After school		No A bit A lot

Did you wear the Actigraph all day? Yes No

If no, what time did you take it off?_____

How long was it off?_____

Why? eg swimming, shower/bath, other_____

Day 2: _____ Day: Saturday

When were you active?	What did you do?	Were you puffed?
Before lunch		No A bit A lot No A bit A lot No A bit A lot No A bit A lot
After lunch		No A bit A lot No A bit A lot No A bit A lot No A bit A lot

Did you wear the Actigraph all day? Yes No

If no, what time did you take it off?_____

How long was it off?_____

Why? eg swimming, shower/bath, other_____

Day 3: _____ Day: Sunday

When were you active?	What did you do?	Were you puffed?
Before lunch		No A bit A lot No A bit A lot No A bit A lot No A bit A lot
After lunch		No A bit A lot No A bit A lot No A bit A lot No A bit A lot

Did you wear the Actigraph all day? Yes No

If no, what time did you take it off?_____

How long was it off?_____

Why? eg swimming, shower/bath, other_____

Day 4: _____ Day: Monday

When were you active?	What did you do?	Were you puffed?
Before school		No A bit A lot
How did you get to school?	Walk/bike/bus/car/other	No A bit A lot
Recess		No A bit A lot
Lunch time		No A bit A lot
Physical Education or sport		No A bit A lot
How did you get home from school?	Walk/bike/bus/car/other	No A bit A lot
After school		No A bit A lot

Did you wear the Actigraph all day? Yes No

If no, what time did you take it off?_____

How long was it off?_____

Why? eg swimming, shower/bath, other_____