Chapter 1

Introduction
Physical activity

Physical activity is recommended for its beneficial effects on mental and physical health in the general population, by for example the reduction of mortality, prevention of osteoporosis, and improved health-related quality of life.\(^1\) It is defined as ‘any bodily movement produced by skeletal muscles that requires energy expenditure’,\(^2\) and expressed in terms of duration, intensity, and frequency. Physical activity is important for children, since regularly loading the bones and muscles contributes to an optimal development of the musculoskeletal system and physical fitness during growth.\(^3,4\) Thereby, a physically active lifestyle in childhood increases the chance for an active lifestyle in adulthood.\(^5\) Regular physical activity is especially important for children with a physical disability who experience activity limitations, such as children with cerebral palsy.

Cerebral palsy

Cerebral palsy (CP) is the most common cause of a physical disability in pediatric rehabilitation and defined by Bax et al. as ‘a group of disorders of the development of movement and posture, causing activity limitations, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behavior, and/or by a seizure disorder’.\(^6\) There are multiple causes for the brain disturbances, which can occur during either the prenatal, perinatal, or postnatal period till the first birthday, resulting in a very heterogeneous group of disorders.\(^7\) To be diagnosed as having CP, clinical symptoms of the brain dysfunction by a central nervous motor disorder, that are causing activity limitations, are obligatory. In the Netherlands the latest data are from 1977-1988 showing a prevalence of 2.44 per 1000 live births.\(^8\) However, more recent data obtained between 1999 and 2002 in South Sweden demonstrate a slight decrease in the prevalence to 2.18 per 1000 live births.\(^9\)

The Surveillance of Cerebral Palsy in Europe (SCPE) agreed on distinguishing three main types of motor disorders: spastic, dyskinetic, and ataxic CP.\(^10\) The SCPE describes these main types as follows: 1) Children with spastic CP have a velocity-dependent
increase in muscle tone at one side (unilateral) or both sides of the body (bilateral), and may demonstrate pathological reflexes, and synergistic movements, 2) Children with dyskinetic CP demonstrate involuntary repeating movements of body segments (e.g. head, or arms) during rest or activities, and 3) Children with ataxic CP have problems with muscle coordination, resulting in performing movements with deviating force, rhythm, and accuracy. Since these types of CP can coexist, children are classified based on the predominant type of the motor disorder. The majority of children with CP (about 80%) demonstrates the spastic subtype.

The severity of CP is categorized by the extent to which the motor disorders result in limitations in activities by using the five-level Gross Motor Function Classification System (GMFCS). The GMFCS classifies children who walk without limitations as GMFCS I, children who walk with limitations as GMFCS II, children who walk with a hand-held mobility device as GMFCS III, children who use wheelchair transportation as GMFCS IV, and children who cannot move around independently as GMFCS V. This thesis is focused on children with unilateral and bilateral spastic CP who are classified as GMFCS I-III.

Figure 1. The international classification of functioning, disability, and health (ICF-CY) model.
Consequences of spastic cerebral palsy

To categorize the impact of the CP on the child’s functioning, the International Classification of Functioning, disability, and health for Children and Youth (ICF-CY) is used.\(^{12}\) The ICF-CY distinguishes the levels of body functions and structure, activity and participation, as well as recognizes their mutual interaction and the interaction with personal and environmental factors (Figure 1). In the ICF-CY terminology, activity is defined as ‘the execution of a task’ in the broadest sense. Mobility is one component of ‘activity’ and describes moving by changing body position or location or by transferring from one place to another, by carrying, moving or manipulating objects, by walking, running or climbing, and by using various forms of transportation.\(^{12}\) The terms ‘mobility capacity’ and ‘mobility performance’ are used to distinguish between ‘what the child can do’ from ‘what the child does do’, respectively. A frequently used synonym for mobility performance is ‘physical activity’. In this chapter the ICF-CY model is used to show how the spastic motor disorder may negatively influence the child’s physical activity level.

At the level of body functions and structure the spastic motor disorder is characterized by muscle dysfunction, which can be divided in reduced passive joint range of motion, increased muscle stiffness, or disturbances in muscle activation.\(^{13}\) Muscle activation can be exaggerated, with signs of abnormal muscle activity, positive or excess signs (e.g. spasticity, increased tone, muscle co-contraction, involuntary synergies, abnormal reflexes) or reduced, with negative or deficit signs (e.g. reduced selective motor control, reduced dexterity).\(^ {13}\) An important feature of the disturbed muscle function in children with CP is a reduction of voluntary muscle strength.\(^ {14}\)

Children with CP also show lower levels of aerobic and anaerobic capacity than typically developing children.\(^ {15-17}\) The causes of the reduction in aerobic and anaerobic capacity are unclear yet. Possibly, it is a direct consequence of the muscle dysfunction, in particular the reduced voluntary muscle strength. It may also be an indirect consequence of the mobility problems of children with CP, possibly resulting in physical inactivity and deconditioning. The term ‘fitness’ is used to define voluntary muscle strength, aerobic capacity, and anaerobic capacity together.
Children with CP in GMFCS I-III experience problems with mobility, such as walking, running, and climbing stairs. Furthermore, most of these children demonstrate a higher energy cost during walking compared to typically developing children, probably as a result of the muscle dysfunction. The higher energy cost of walking in combination with the lower maximal aerobic capacity results in a higher physical strain of walking than typically developing children, possibly resulting in an earlier fatigue. A higher physical strain has been associated with reduced physical activity of adults with CP. It is suggested that this high physical strain negatively impacts physical activity, as illustrated by children with CP taking less strides per day (further referred to as ‘walking activity’), and showing a lower level of physical activity than typically developing children.

Figure 2. Physical activity for people with a disability (PAD) model. Reprinted with permission.
Besides the reduced fitness and mobility problems of children with CP, personal and environmental influences may also contribute to a decreased level of physical activity.\textsuperscript{18,25,26} Bar-Or has indicated that, among children with a disability or a chronic disease, parents may pamper their child with CP for performing too strenuous activities, they may have fear for injuries, children may have problems with social interactions, or there may be a lack of awareness of the importance of being physically active.\textsuperscript{25} Rimmer et al. have demonstrated similar issues in their ‘disability-associated low energy expenditure deconditioning syndrome (DALEEDS) model’, but added that people with a disability may have the perception that they need to save energy to prevent fatigue, resulting in reduced physical activity.\textsuperscript{26} The ‘Physical Activity of people with a Disability (PAD) model’ (See Figure 2) provides a schematic overview of the personal (e.g. health condition, self-efficacy) and environmental factors (e.g. accessibility of the home, social environment) that may influence the intention of people with a disability to be physically active.\textsuperscript{27}

\textbf{Vicious cycle of deconditioning}

Although CP is a non-progressive disorder, mobility capacity is not necessarily stable over time.\textsuperscript{28} Some children who walk independently in childhood use walking aids during adolescence.\textsuperscript{29,30} As a consequence of the muscle dysfunction and mobility limitations, children with CP often lack opportunities for physical activity and exercise which, in combination with the CP itself, restrict an adequate development of their muscle strength as needed for a healthy development. As a result, increases in muscle strength fall short of the inevitable increases in the child’s bodyweight and height, leading to secondary impairments that negatively influence independent walking and regular engagement in physical activity. These include the mobility limitation itself, as well as pain, fatigue, overweight, and over-use injuries.\textsuperscript{26,31} In addition, the reduced fitness of children with CP in itself may lead to mobility limitations, which in turn may result in more sedentary behavior. This decrease in physical activity further reduces fitness, increasing the risk for a vicious cycle of deconditioning, and further deterioration of mobility.\textsuperscript{25,26,32} This may also have negative effects on the child’s participation, mental health, and future health care use.\textsuperscript{26} The promotion of physical activity and fitness in children with CP is suggested to break down this vicious cycle.\textsuperscript{25,31,32}
Previous studies have demonstrated that positive effects on fitness can be achieved in children with CP after lower-extremity strength training, or aerobic and anaerobic training. However, effects were not maintained after the training ended. With regard to improving physical activity, two studies among children with CP were found that evaluated the effect of either a counselling program or an aerobic training program. No significant effects were determined on physical activity, however, both studies suggested that their interventions may be beneficial for improving physical activity in children with CP based on trends. Thereby, positive effects on self-reported physical activity have been shown after counseling in adults with a disability. These previous findings suggest that interventions aimed at breaking down the vicious cycle of deconditioning should not only focus on improving fitness, but also on helping child and parents to change towards a more physically active behavior. This may be operationalized by helping families how to deal with the personal and environmental barriers that limit physical activity. Additionally, practicing activities in the daily situation (e.g. the child’s home) instead of in the physiotherapy practice setting may positively influence physical activity, since it is suggested to decrease the gap between mobility capacity and mobility performance. Based on this information the physical activity stimulation program ‘LEARN 2 MOVE 7-12’ has been developed for children with CP, in which fitness training, counseling, and home physiotherapy were combined in order to improve physical activity.

**Embedding**

The thesis ‘Physical activity in children with spastic cerebral palsy’ has been performed at the VU University Medical Center Amsterdam, within the scope of the national research program ‘LEARN 2 MOVE’ (see also www.cp-research.nl/LEARN2MOVE). LEARN 2 MOVE evaluates the effects of age-specific interventions for children and adolescents with cerebral palsy on performance of activities. The present thesis is focused at children with spastic cerebral palsy in the age of 7-12 years.

**Aim and outline of the thesis**

The primary aim of this thesis is to determine the effectiveness of the LEARN 2 MOVE 7-12 physical activity stimulation program for children with CP aged 7-12 years. The
secondary aim of this thesis is to better understand the characteristics of the walking activity of children with CP. The outline of this thesis is as follows: Chapter 2 describes the characteristics associated with walking activity on schooldays and on weekend days among Dutch children with CP. Chapter 3 compares the walking activity of children with CP and typically developing children between the Netherlands and the United States, while Chapter 4 compares the walking activity and its intensity between children with CP and typically developing children in the Netherlands. The rationale, content and study protocol of the LEARN 2 MOVE 7-12 physical activity stimulation program are described in Chapter 5. Chapter 6 addresses the effectiveness of the LEARN 2 MOVE 7-12 program on physical activity, mobility capacity, fitness, and attitude towards sports. Chapter 7 addresses the effectiveness of the LEARN 2 MOVE 7-12 program on social participation, self-perception and quality of life. In the general discussion in Chapter 8, the main findings, practical implications, methodological issues, and future directions following from this thesis are provided.

References


