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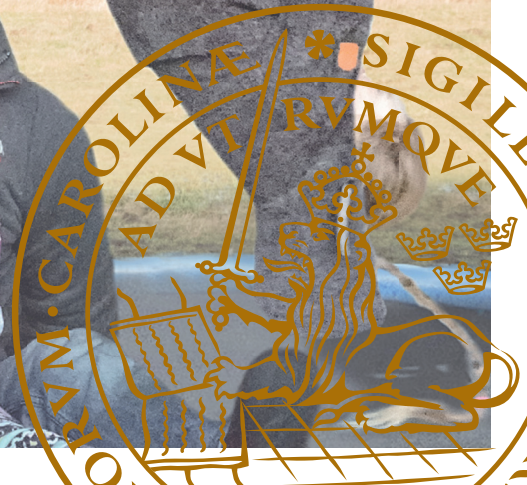
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# Participation in physical activities and sedentary behaviour among children with physical disabilities

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# Participation in physical activities and sedentary behaviour among children with physical disabilities

Katarina Lauruschkus



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DOCTORAL DISSERTATION

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To be defended at Health Science Centre, SSSH-salen on Friday the  
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*Faculty opponent*

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Oslo University Hospital

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Title : Participation in physical activities and sedentary behaviour among children with physical disabilities	
<p><b>Abstract</b></p> <p>Participating in physical activity promotes physical and mental health. Children with physical disabilities are not as physically active as other children. They tend to participate in lower intensity physical activity, choose more passive activities with less variety and have more sedentary time. The overall aim of this thesis was to gain knowledge about participation in physical activities, to identify associated factors for school and leisure time and to explore the perceived experiences of the children and the parents about possible facilitators or barriers associated with physical activities. A further aim was to evaluate the feasibility of Physical Activity Referrals (PAR) and outcome measures for children with physical disabilities and its effectiveness on increasing participation in physical activity and decreasing sedentary behaviour. The thesis is comprised of three studies with children with various gross- and fine motor, communicative and cognitive functions. In Study I (Paper I) a total population of 364 children and adolescents with cerebral palsy (CP) aged 7-17 years was studied by using cross-sectional data from the National Quality Registry CP follow-up programme (CPUP). In Study II qualitative interviews with 16 children with CP, aged 8-11 years (Paper II), and with 25 of their parents (Paper III) were performed and analysed by content analysis. In Study III (Paper IV), 14 children with physical disabilities, aged 7-12 years, participated in PAR together with 24 of their parents. The intervention was based on a written agreement of self-selected physical activities, motivational interviewing (MI), goal-directed outcome measurements Canadian Occupational Performance measure (COPM) and Goal Attainment Scaling (GAS) and assessments including Gross Motor Function Measure (GMFM-66), physical activity monitors, the International Physical Activity Questionnaire (IPAQ) and other questionnaires at baseline, and after 8 and 11 months. Children and adolescents with more severe motor limitations, with intellectual disability and with thinness seem to have the greatest needs for interventions to increase their participation in physical activities and reduce sedentary behaviour. Children discussed that they want to be asked about their wishes and needs, and get the opportunity to try self-selected physical activities with individualised support, as well as to have fun with family and friends, have someone to do the activity with and enjoy the sensation of speed. Environmental factors could be either facilitators or barriers, while pain, fatigue and the perception of not being good enough were considered barriers. The parents experienced several challenges when their child wanted to be physically active: parents wanted to protect their child while also pushing them towards independence. They wanted competent persons to be available to assist their child in participating in the physical activities and support them in finding friends to be active with. Family culture and attitudes affect children's motivation for being physically active and should be taken into account. In Study III the families were compliant and there were no dropouts; there were positive evaluations of PAR. The families learned more about the accessibility and locations of physical activities, and the children got the opportunity to try their self-selected activities with individualised support. Several children made new friends through engaging in self-selected group activities. In addition, most children were able to maintain the physical activity levels; the outcome scores for GMFM-66, COPM and GAS were maintained for some and increased for most children. Modifications by synchronising physical activity measures into the same time periods and to give a feedback to the families close in time to the baseline measures are recommended. In this thesis the children with the greatest needs of a more active lifestyle have been identified, and we have listened to their voices and talked with them and their parents before designing an individualised intervention. Families with different ethnical, cultural and socioeconomic backgrounds participated in the studies. PAR promotes an active lifestyle by increasing participation, motivation and engagement in physical activities and social participation. The intervention involves both everyday and organised habitual physical activities. To our knowledge, this is the first study about PAR in children with disabilities that measures objective and estimated physical activity. The results suggest that PAR is feasible and effective for children with physical disabilities.</p>	
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Katarina Lauruschkus



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Stay positive, be active,  
and look to the future.

Marc Twynholm





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# Abstract

Participating in physical activity promotes physical and mental health. Children with physical disabilities are not as physically active as other children. They tend to participate in lower intensity physical activity, choose more passive activities with less variety and have more sedentary time.

The overall aim of this thesis was to gain knowledge about participation in physical activities, to identify associated factors for school and leisure time and to explore the perceived experiences of the children and the parents about possible facilitators or barriers associated with physical activities. A further aim was to evaluate the feasibility of Physical Activity Referrals (PAR) and outcome measures for children with physical disabilities and its effectiveness on increasing participation in physical activity and decreasing sedentary behaviour.

The thesis is comprised of three studies with children with various gross- and fine motor, communicative and cognitive functions. In Study I (Paper I) a total population of 364 children and adolescents with cerebral palsy (CP) aged 7-17 years was studied by using cross-sectional data from the National Quality Registry CP follow-up programme (CPUP). In Study II qualitative interviews with 16 children with CP, aged 8-11 years (Paper II), and with 25 of their parents (Paper III) were performed and analysed by content analysis. In Study III (Paper IV), 14 children with physical disabilities, aged 7-12 years, participated in PAR together with 24 of their parents. The intervention was based on a written agreement of self-selected physical activities, motivational interviewing (MI), goal-directed outcome measurements Canadian Occupational Performance measure (COPM) and Goal Attainment Scaling (GAS) and assessments including Gross Motor Function Measure (GMFM-66), physical activity monitors, the International Physical Activity Questionnaire (IPAQ) and other questionnaires at baseline, and after 8 and 11 months.

Children and adolescents with more severe motor limitations, with intellectual disability and with thinness seem to have the greatest needs for interventions to increase their participation in physical activities and reduce sedentary behaviour. Children discussed that they want to be asked about their wishes and needs, and get the opportunity to try self-selected physical activities with individualised support, as

well as to have fun with family and friends, have someone to do the activity with and enjoy the sensation of speed. Environmental factors could be either facilitators or barriers, while pain, fatigue and the perception of not being good enough were considered barriers. The parents experienced several challenges when their child wanted to be physically active: parents wanted to protect their child while also pushing them towards independence. They wanted competent persons to be available to assist their child in participating in the physical activities and support them in finding friends to be active with. Family culture and attitudes affect children's motivation for being physically active and should be taken into account. In Study III the families were compliant and there were no dropouts; there were positive evaluations of PAR. The families learned more about the accessibility and locations of physical activities, and the children got the opportunity to try their self-selected activities with individualised support. Several children made new friends through engaging in self-selected group activities. In addition, most children were able to maintain the physical activity levels; the outcome scores for GMFM-66, COPM and GAS were maintained for some and increased for most children. Modifications by synchronising physical activity measures into the same time periods and to give a feedback to the families close in time to the baseline measures are recommended.

In this thesis the children with the greatest needs of a more active lifestyle have been identified, and we have listened to their voices and talked with them and their parents before designing an individualised intervention. Families with different ethnical, cultural and socioeconomic backgrounds participated in the studies. PAR promotes an active lifestyle by increasing participation, motivation and engagement in physical activities and social participation. The intervention involves both everyday and organised habitual physical activities. To our knowledge, this is the first study about PAR in children with disabilities that measures objective and estimated physical activity. The results suggest that PAR is feasible and effective for children with physical disabilities.

# Abbreviations

BMI	Body Mass Index
CFCS	Communication Function Classification System
CI	Confidence Interval
COPM	Canadian Occupational Performance Measure
CP	Cerebral Palsy
CPUP	Cerebral Palsy Follow Up Programme (CPUP) and quality registry
FCS	Family-Centred Service
GMFCS-E&R	Gross Motor Function Classification System Expanded and Revised
GAS	Goal Attainment Scaling
GMFM-66	Gross Motor Function Measure, 66-item
ICF/ICF-CY	International Classification of Functioning, Disability and Health/ ICF-version for Children and Youth
IPAQ	International Physical Activity Questionnaire
MACS	Manual Ability Classification System
MI	Motivational Interviewing
PA	Physical Activity
PAR/PAP	Physical Activity Referral/Physical Activity on Prescription
PE	Physical Education
PIMD	Profound Intellectual and Multiple Disabilities
PD	Physical Disability
POR	Proportional Odds Ratio
RCT	Randomised Controlled Trial
SCT	Social Cognitive Theory
SD	Standard Deviation
TTM	Transtheoretical Model
VAS	Visual Analogue Scale
WHO	World Health Organisation



# Original papers

This thesis for the doctoral degree is based on the following papers referred to in the text by their Roman numerals I-IV:

- I Lauruschkus K, Westbom L, Hallström I, Wagner, P, Nordmark E. Physical activity in a total population of children and adolescents with cerebral palsy. *Research in Developmental Disabilities*. 2013; 34:157-167.
- II Lauruschkus K, Nordmark E, Hallström I. “It’s fun, but...” Children with cerebral palsy and their experiences of participation in physical activities. *Disability and Rehabilitation*. 2014; Early Online:1-7.  
Doi:10.3109/09638288.2014.915348.
- III Lauruschkus K, Nordmark E, Hallström I. Parents’ experiences of participation in physical activities for children with cerebral palsy – protecting and pushing towards independency. Submitted.
- IV Lauruschkus K, Hallström I, Westbom L, Tornberg, Å., Nordmark E. Participation in physical activities for children with physical disabilities: Feasibility and effectiveness of physical activity referrals. Submitted.

Papers I and II have been reprinted with the permission of the journals.

# Thesis at a glance

Study	Aims	Methods	Results
I	To describe the participation in physical activity of children with CP at school and during leisure time and to identify characteristics associated with physical activity	N=364 children 7-17 years in the total population with CP in the Skåne region  Descriptive statistics and proportional odds ratios, using cross-sectional data from the CPUP	Severe gross motor limitations led to low participation in PE at school and to low participation in regular physical leisure activity. Intellectual disability was associated with low frequency of physiotherapeutic interventions.
II	To explore the experiences of children with CP regarding their participation in physical activities, and to describe facilitators and barriers	N=16 children with CP 8-11 years  Individual and focus group interviews analysed with content analysis	Children with CP want to be physically active with others, have fun and enjoy the sensation of speed. Pain, fatigue and the accessibility of activities and locations should be considered.
	To explore how parents of children with CP experience their child's participation in physical activities and to identify facilitators and barriers for being physically active and reducing sedentary behaviour	N=25 parents of the 16 children with CP  Individual and focus group interviews analysed with content analysis	Parents desire competent persons to supporting participation in physical activities and to find friends to be physically active with. Family culture and attitudes affect their child's motivation for being physically active.
III	To evaluate PAR for children with physical disabilities according to feasibility and the impact on physical activity and sedentary behaviour	N=14 children with physical disabilities, 7-12 years  N=24 parents of the 14 children with physical disabilities Multiple cases to assess PAR, MI, COPM, GAS, GMFM-66, questionnaires, IPAQ, logbook, accelerometer, time-use diary, analysed with descriptive statistics	PAR was feasible and effective without any drop-out rate. The children participated in 1-3 self-selected physical activities, the ones in group activities made new friends. Their physical activity was, on average, 90 minutes at a moderate to vigorous level daily (< 30 minutes to > 3 hours), measured by an accelerometer. The outcome scores of GMFM-66, COPM and GAS increased for most children.



# Introduction

There is a general consensus that participating in physical activities enhances people's health, and that it is important to obtain an active lifestyle in childhood. Health is defined as a 'state of complete physical, mental and social well-being and not merely the absence of disease or infirmity' (WHO, 1948). This definition has not been modified since 1948, which has caused discussions worldwide. The critics have proposed a definition change, and that health should be considered 'the ability to adapt and to self-manage' (Huber et al., 2011).

However, health is important for our wellbeing, and global and national public health goals promote physical and psychological health and disease prevention. Prerequisites for good health are affected by living conditions, the individual's own choices and lifestyles. The overall goal, in order to substantially reduce deaths and disease burden worldwide according to the WHO's Strategy on Diet, Physical Activity and Health (2004), is to promote and protect health through healthy eating and physical activity (PA). Long-term combined efforts of many stakeholders, both public and private, are required to change dietary habits and patterns of PA. According to the Global Strategy effective actions are needed, with close monitoring and evaluation of their impact, at global, national, regional and local levels.

Levels of physical inactivity have increased worldwide, and physical inactivity is the fourth leading risk factor for global mortality. Globally, a third of the adult population is not physically active enough. The WHO recommends a supportive environment for increased levels of PA and claims that doing some PA is better than doing none. They also state that PA has health benefits across all age groups (WHO, 2010a). PA has been associated with an increased life expectancy and decreased risk of cardiovascular disease. PA produces overall physical, psychological and social benefits (AHA, 2014).

Children with disabilities and their families are very similar to children without disabilities and their families. Since families are the central setting for each child, offering opportunities for participation, they are essential for any community (LaForme Fiss et al., 2013). It can be challenging to find opportunities for children with disabilities to participate, and parents use various coping strategies in order to

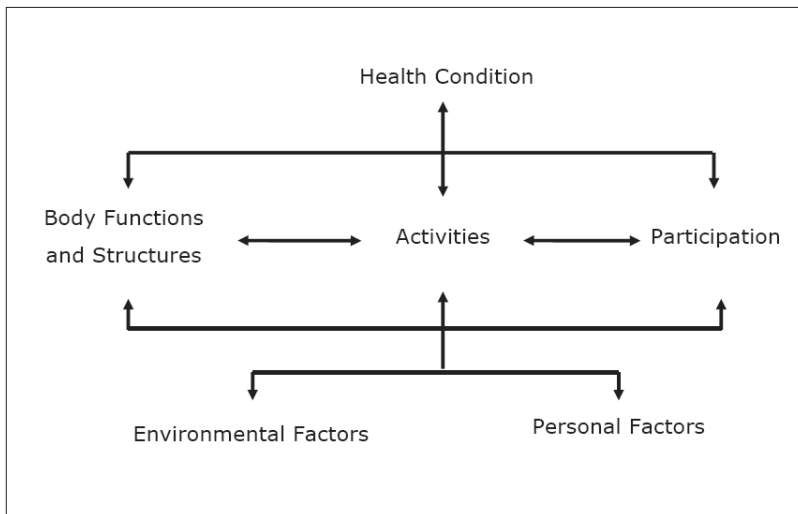
achieve this both within the family unit and in their wider community (King et al., 2006; Palisano et al., 2011). Although research has been done to explore the participation of children with disabilities in leisure activities and physical activities (Fauconnier et al., 2009; Palisano et al., 2011), there is a lack of participation research about children with different motor, cognitive and communicative functions and about children from different sociodemographic and cultural backgrounds (Kolehmainen et al., 2011). Research is also needed about personalised interventions that address each child's individual preferences, wishes and conditions and that include the child and their parents in the intervention processes. The purposes of this thesis were to explore the participation in PA in children with physical disabilities and from various backgrounds and to evaluate the feasibility and effectiveness of an intervention for increased participation in PA and decreased sedentary behaviour.

## Conceptual framework

The International Classification of Functioning, Disability and Health (ICF) (WHO, 2001) and its version for Children and Youth (ICF-CY) (WHO, 2007) are multidimensional frameworks for measuring and classifying the concepts of health, functioning and disability at both individual and population levels. They are classifications of health and health-related domains, including a list of environmental factors as the functioning and disability of an individual occurs in context. The WHO developed the models to provide a common language for people, caregivers and organisations to communicate people's functions and life situations (WHO, 2001; WHO, 2007; Adolfsson et al., 2010). The ICF and ICF-CY are further developments of the earlier International Classification of Impairments, Disabilities, and Handicaps (ICIDH) (WHO, 1980), based on a paradigm change from a biomedical perspective, with physical impairment causing disability in daily life and the focus being on 'normalising' or 'curing' the clients, to a social perspective, with social and physical barriers in the environment causing disability and the focus being on adapting the environment. Based on a combination of biomedical and social dimensions of health, the ICF was established as a biopsychosocial conceptual model. The ICF integrates body function and structure, activity and participation with personal and environmental factors. The ICF-CY and from the ICF developed 'F-words' constitute overarching conceptual frameworks for discussions about the participation in physical activities for children with physical disabilities.

## ICF-CY

The ICF and ICF-CY focuses on health rather than on the consequences of disability. The concepts of body function and structure, activity and participation interact dynamically with each other and with personal and environmental factors as shown in Figure 1. The ICF-CY combines the Convention on the Rights of the Child developed by the United Nations (UN, 1990) with the ICF, covering developmental characteristics of children 0-17 years of age (Simeonsson et al., 2003 and 2010; WHO, 2007). The relation between health status and developmental changes from infancy to adolescence is focus of the ICF-CY (Bult et al., 2011). By identifying impairments, activity limitations and participation restrictions as well as the effects of personal and environmental contextual factors, the ICF-CY can be used for identifying primary goals and evaluating the effects of interventions in paediatric physiotherapy (Darrah, 2008).



**Figure 1.** The ICF/ICF-CY model (WHO, 2007; p. 17)

### The 'F-words'

Grounded in the ICF, the 'F-words' in childhood disability were developed by Rosenbaum and Gorter (2012). The idea was to find an appealing way to integrate the ICF into advocacy regarding children with disabilities and their families, clinical services and research. For this, the authors claimed that further development was needed with respect to the ongoing discussions about the WHO definition of health.

As mentioned above, Huber et al. (2011) proposed a change from a ‘state of complete physical, mental and social well-being and not merely the absence of disease or infirmity’ (WHO, 1948) to using the definition of ‘health as the ability to adapt and to self-manage’ (Huber et al., 2011), when facing social, physical, and emotional challenges.

The ‘F-words’ are fitness, function, friends, family, fun and future. In accordance to the ICF-CY, there is no hierarchy of implied importance in these concepts (Rosenbaum & Gorter, 2012):

*Fitness* is used to personalise the domain *Body Structure and Body Function* in the ICF framework. The authors claim that the fitness of children with disabilities has been neglected until recently even though they are in need of improved fitness. They are less fit than they should be, and also when compared to children without disabilities.

*Function* is used to personalise the domain *Activity*, as function according to the authors refers to what people do. A distinction should be made between the child’s capacity – what they can do when at their best, and their performance – what they ordinarily do. As performance improves with practice, children should first learn to do activities in their own way before developing specific skills.

*Friends* personalise the domain *Participation*, but are also relevant as the child’s *Personal Factors*. Friends and facilitating friendships should be included as an important component of child development in our contacts with the families. The quality rather than quantity of peer relationships are described as important, and ways to encourage, empower and enhance children’s opportunities for developing friendships should be considered.

*Family* is used to personalise the domain *Environmental Factors* and the family represents the essential environment of children. Family-centred services address the concerns about challenged parental health, when taking grandparents’ and siblings’ voices into account. The family – parents, siblings, grandparents and other important persons – is essential for the child’s wellbeing, and therefore various supports and resources specifically for families should be considered when helping them to make informed decisions.

*Fun* personalises the domain *Personal Factors*, but it is also relevant for *Participation*. The authors feel that the answer on how to increase participation in children with disabilities is to find out what they want to do. Fun increases participation and should be used to build children’s confidence, competence and capacity.

*Future* is according to the authors what child development is all about. Service providers and parents should consider their child’s future. The child and their parents

should be asked about their expectations and dreams for a possible future, and not been told what is impossible.

## Children with physical disabilities

In the Convention on the Rights of the Child the UN (1990) have proclaimed that childhood is entitled to special care and assistance and consider that all children should have equal dignity and rights. In addition, all actions relating to the child should focus on the child's best interests, and each child's right to life includes their physical health as well as their spiritual, moral and social development. Each child should actively participate in society, have a full life, have the right to express their own views and be considered in all matters that affect them.

Disability is an umbrella term for impairments, activity limitations and participation restrictions (WHO, 2011). In their definition, the World Report on Disability refers to the negative aspects of a disabled person's contextual factors, including both environmental and personal factors (WHO, 2011). Similarly, the United Nations Convention states that people with disabilities can have long-term physical, mental, intellectual or sensory impairments. These impairments may hinder their equal participation in society, particularly when considering other barriers (UN, 2006). The Equality and Human Rights Commission (1995) definition of disability is: 'a physical or mental impairment which has a substantial and long-term adverse effect on the ability to carry out normal day-to-day activities'. These definitions of disability support the biopsychosocial health paradigm that disability is not a personal attribute and individuals with similar impairments may have different experiences of limitations in their everyday living.

The Convention on the Rights of the Child (UN, 1990) declares in Article 23 that 'a mentally or physically disabled child should enjoy a full and decent life, in conditions which ensure dignity, promote self-reliance and facilitate the child's active participation in the community'. The disabled child's eligibility to and extension of assistance shall be encouraged and ensured. In addition, the special needs of a disabled child shall be recognised and designed to ensure that the each child has 'effective access to and receives education, training, health care services, rehabilitation services, preparation for employment and recreation opportunities in a manner conducive to the child's achieving the fullest possible social integration and individual development, including his or her cultural and spiritual development.' Children with disabilities also have the right to be treated with respect. In the 1970s and 1980s a

non-categorical approach to chronic conditions, including the variation and different factors that affect health, was commonly used (Pless & Pinkerton, 1975). Children with chronic illness have the same basic needs of services as all children, and limitations of daily functioning are related to impaired health rather than to specific causes of diseases, according to the non-categorical approach. This approach has received renewed attention and is described as making it possible to construct health outcome models assessing outcomes both between and within conditions (Ronen et al., 2011).

Physical disabilities (PD) as a chronic illness cover permanent motor impairments in children as a result of congenital or early acquired injury to or illness in the nervous, muscular or skeletal systems, which is described in the Swedish Habilitation and Assistive Technology Services (2011). Additional conditions such as limitations in intellectual and communicative functions, hearing and visual impairments and epilepsy are common in children with PD. The largest group of children with PD is represented by children with cerebral palsy (CP) with an estimated prevalence of 2.0 to 2.5/1000 children (Westbom et al., 2007). There are many other PD, such as neuromuscular disorders with an estimated prevalence of 6.3/10000 children (Darin & Tulinius, 2000), or spina bifida with an estimated prevalence of 3/10000 children in Sweden (Nikkila et al., 2006). Children with profound intellectual and multiple disabilities (PIMD) have intellectual impairments combined with profound physical and sensory impairments and also commonly have medical complications (Nakken & Vlaskamp, 2007). Children with PIMD are often included in the group of children with CP, as this group involves a wide range of disorders. There is little knowledge about the opportunities for PA and an active lifestyle for children with PIMD, as well as how cultural and socioeconomic backgrounds might impact on this.

## Cerebral palsy

As a disorder, cerebral palsy is complex and heterogeneous and after decades of considerations the following definition was developed in 2006: *CP describes a group of permanent 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, perception, cognition, communication and behaviour, by epilepsy and by secondary musculoskeletal problems* (Rosenbaum et al., 2007, p. 9)

This definition allows for a common international terminology, as does the classification of CP-subtypes according to the Surveillance of Cerebral Palsy in Europe (SCPE). CP-subtypes are classified at 4-7 years of age based on the dominant neurological sign. Spastic CP can be divided into the subtypes Unilateral Spastic CP (USCP) and Bilateral Spastic CP (BSCP). The other CP-subtypes can be classified as dyskinetic, ataxic or non-classifiable CP (SCPE, 2000). Gross motor function, manual ability and communication function in children with CP can be classified in the classification systems GMFCS-E&R, MACS and CFCS (Compagnone et al., 2014), as described in the following three sections.

### *GMFCS-E&R*

The gross motor function for children and adolescents with CP aged 0-18 years can be classified according to the Gross Motor Function Classification System Expanded and Revised (GMFCS-E&R), which is a five level classification system (Palisano et al., 2007). Children whose gross motor function is classified at level I can generally walk without restrictions but tend to be limited in some of the more advanced motor skills. Children with level V are generally very limited in their ability to move around themselves even with the use of assistive technology. The focus of GMFCS-E&R is on usual performance in the home, at school and in community settings, rather than on capability. The potential impact of environmental factors such as travelling distances to school and around the community as well as personal factors such as energy demands and social preferences for methods of mobility are taken into account. In a Swedish context the Swedish translation of GMFCS-E&R can be used (Lundkvist & Nordmark, 2007).

### *MACS*

The Manual Ability Classification System (MACS) is used to classify and describe self-initiated ability to handle objects in important daily activities, for example during play and leisure, as well as the need for assistance or adaptation in order for children and adolescents with CP to be able to perform manual activities in everyday life. MACS is a classification system with five levels for children and adolescents aged 4-18 years where level I corresponds to 'handles objects easily and successfully' and level V means 'does not handle objects and has severely limited ability to perform even simple actions'. Both validity and reliability of MACS are described to be good (Eliasson et al., 2006).

## *CFCS*

The communication function for persons with CP in all ages can be classified according to the Communication Function Classification System. The CFCS consists of five descriptive levels for everyday communication performance where level I describes the child being an 'effective sender and receiver with unfamiliar and familiar partners' and level V describes a child that is 'seldom effective sender and receiver even with familiar partners' (Hidecker et al., 2011). While originally developed for use with persons with CP, the CFCS is now being used to describe communication performance of individuals with any disability ([www.cfcs.us](http://www.cfcs.us)).

All children with CP or CP-like symptoms in Sweden are offered participation in the CPUP (CPUP, 2014) which is described in the following section.

## **CPUP**

CPUP is a follow-up surveillance programme for people with CP and since 2005 a National Quality Register, implying that all counties in Sweden participate in CPUP. Other Scandinavian countries as Norway, parts of Denmark and Iceland, as well as parts of Australia and Scotland also participate in the programme. CPUP was started in 1994 as a collaboration between the paediatric orthopaedics and the child and youth habilitation centres, in the two Southern Swedish counties Skåne and Blekinge and was aimed at preventing the occurrence of hip dislocation and severe deformities by means of continuous and standardised surveillance and combined with treatment at an early stage, if necessary, and thereby optimising the functional ability and quality-of-life of persons with CP. In 2014 a 20-year follow-up study of CPUP of a population-based prevention programme was published showing that hip dislocation could be prevented and the number of children who develop severe contractures, windswept-deformity and scoliosis could be reduced (Hägglund et al., 2014). Physiotherapists, occupational therapists and paediatricians are using assessment and evaluation forms developed for their profession and children with CP, their families as well as the participating professionals consider CPUP to lead to improved cooperation and a more uniform and overall improved healthcare system in the country. Besides information on joint range of motion, the assessment form for physiotherapists contains information such as muscle tone, pain, motor function and information about the frequency of PA, leisure activities and physical therapy interventions (CPUP, 2014).



## Family

The definition of the nuclear family consisting of a mother, father and children is no longer adequate considering that divorce, remarriage and cohabitation are common practices and accepted in our society. Families can be described as complex constructs, and an understanding of issues such as ethnicity, culture, socioeconomic status, education and values are needed for clinical interventions (Dunst & Trivette, 2009; LaForme Fiss et al., 2013). All members of a family influence each other. The most central part of each child's ecology during childhood is their family, and family ecology refers to interaction and interdependence among individual family members and their environment (LaForme Fiss et al., 2013). As family ecology – including family strengths, expectations, interests, supports and resources – influences children's activity and participation in daily life (King et al., 2006) and including their social and leisure time activities (Palisano et al., 2011), it should be considered when providing services to children with disabilities (Imms et al., 2008; LaForme Fiss et al., 2013). Therefore, children must be seen in their family context and cannot be viewed as an isolated system (Bronfenbrenner, 1986). In this thesis, the term family is used in Study II and III and is defined by each child and their parents. Parents are biological, adoptive or foster home parents. Siblings, grandparents and cousins are the most frequent named members of the extended family, thus other important adults or a dog are occasionally included by a child.

Parents of school-aged children with CP experience high levels of distress, emotional and financial burden and time constraints, which are associated with the child's behaviour difficulties and functional limitations (Majnemer et al., 2012). Majnemer et al. suggested that the family's ability to adapt to having a child with a physical disability is essential for the parents' physical and psychological health as well as their child's well-being and participation. Parents of children with more severe impairments report a poorer quality of life, which may depend on more parental distress (Davis et al., 2012). Children with profound intellectual and multiple disabilities (PIMD) are less engaged in family activities compared with children without disabilities (Axelsson et al., 2013). In these families much effort is spent on adapting family living patterns to the child's functioning. Socioeconomic factors such as family income and the parents' education are negatively associated to the child with PIMD's engagement. A low diversity of family activities are seen in families with children with PIMD, and less physically demanding activities such as watching movies are preferably chosen instead of housekeeping activities or playing outside with their siblings (Axelsson et al., 2013). Parents of children with CP experience different needs over time according to their child's interventions, and they need time

to build a collaborative relationship to their child's therapists (Kruijsen-Terpstra et al., 2014). The demands of everyday life, resources provided to parents, attitudes in the community and culture have a major impact on the parental experiences. Parents of children with PD have many strategies to support their child and they feel that they have to advocate and educate others about their child's needs (Piskur et al., 2012).

Parents coming from diverse ethnic groups often experience challenges when receiving healthcare for their child with a disability due to language obstacles, discrimination and having little knowledge of healthcare services. This implies a need for culturally sensitive care where health providers try to understand and appreciate the families' values, beliefs and goals in order to plan and deliver effective healthcare (Lindsay et al., 2012).

## Physical activity

Physical activity promotes good health and disease prevention, but has also been identified as the fourth leading risk factor for global mortality and causes an estimated 3.2 million deaths globally. PA is defined as any bodily movement produced by skeletal muscles that require energy expenditure. PA for children include play, transportation, chores, recreation, sports, exercises or physical education, performed in the context of family, school and community activities (WHO, 2010a). The global daily recommendations for children aged 5 to 17 years consist of at least 60 minutes of PA at a moderate to vigorous intensity in order to improve cardiorespiratory, muscular fitness and bone health, and to reduce cardiovascular disease risk. Additional health benefits are provided when the amounts of PA are greater than 60 minutes. Vigorous-intensity activities should be incorporated, including those that strengthen muscle and bone such as running or jumping, at least 3 times per week.

The intensity of PA varies between people, depending on each individual's previous exercise experience and their relative level of fitness. Low-intensity PA requires a low amount of effort and barely accelerates the heart rate. Moderate-intensity PA requires a moderate amount of effort and noticeably accelerates the heart rate, whereas vigorous-intensity PA requires a large amount of effort and causes rapid breathing and a substantial increase in heart rate (WHO, 2010a).

The recommendations of PA apply to all children, including those with disabilities. However, as 'bodily movement produced by skeletal muscles resulting in increased energy expenditure' also happens in dyskinesia and spasticity, the PA definition may

sometimes be difficult to applicate. Those movements are not voluntarily and may lead to high PA levels, which should be considered in interventions.

The National Guidelines for Methods of Preventing Disease have been developed by the Swedish National Board of Health and Welfare (2011). They provide recommendations for methods of disease prevention by supporting individuals in their efforts to change unhealthy lifestyle habits. Unhealthy lifestyle habits such as tobacco use, excessive alcohol consumption, low levels of PA and unhealthy eating habits were identified, which are similar to the factors identified by WHO (2010a). The Public Health Agency of Sweden (2014) has identified 11 target areas to create social conditions for good health on equal terms for the entire population, where PA is one of them. Biological and social factors, as well as habits and relationships, affect PA according to the description of the Public Health Agency. Overall structural factors, such as an individual's living conditions and lifestyle, social networks and relationships are described as having an effect on each other.

### **Habitual physical activity**

PA can be performed through domestic, occupational, commuting or leisure activities, although domestic and occupational activities are part of daily tasks and have other purposes besides being physically active. Daily commuting, which implies active transportation such as walking or cycling to school and leisure PA are commonly planned, and exercises and sports are particularly dependent on personal preferences. Exercises and sports can be described as bodily movement that is planned, structured and performed to improve physical fitness (Caspersen et al., 1985). Occupational and commuting PA can be described as everyday PA, such as when the child is cycling to school. All activity during transportation, at school, household or leisure time on moderate or vigorous intensity is health-enhancing PA.

In recent research, regular PA, which is increasingly described as habitual PA, consists of activities such as leisure activities and organised and spontaneous sports or games that are performed daily as a habit (Bania et al., 2011; Carlon et al., 2013). The majority of independently ambulant children and adolescents with unilateral CP do not perform sufficient habitual PA to meet the PA recommendations (Mitchell et al., 2014).

## **Cardiorespiratory fitness**

Physical fitness is a set of attributes that people are considered to either have or can achieve, and it relates to the ability to perform PA (Caspersen et al., 1985). Cardiorespiratory fitness is a health-related component of physical fitness and described as the ability to supply oxygen during sustained PA to the circulatory, respiratory and muscular systems. Cardiorespiratory fitness can be expressed in metabolic equivalents (METs) or maximal oxygen uptake ( $VO_2$  max) measured by exercise tests such as cycle ergometer. One MET is defined as the energy cost of sitting quietly and is equivalent to a caloric consumption of  $1\text{kcal}\cdot\text{kg}^{-1}\cdot\text{h}^{-1}$ . Cardiorespiratory fitness is described as a sensitive and reliable measure of habitual PA and is associated with morbidity and mortality in both men and women independently of other risk factors (Lee et al., 2010).

PA and cardiorespiratory fitness are both independently associated with lower cardiovascular risk and should be considered when estimating that risk for each individual (Ekblom-Bak, 2010).

## **Sedentary behaviour**

Regardless of meeting global recommendations for children of 60 minutes PA per day, sedentary behaviour has been found to increase the risk for several common health diseases and premature mortality (Hamilton et al., 2008). Sedentary behaviour is defined as ‘any waking behaviour characterised by an energy expenditure  $\leq 1.5$  metabolic equivalents (METs) while in a sitting or reclining posture’. The MET is a physiological measure expressing the energy cost of activities, against a reference to the metabolic cost of rest. Activities that require 1.0-1.5 METs are considered to be sedentary behaviour as the recruitment of larger muscle groups is not needed (Innes & Darrah, 2013; Trembley, 2012; Verschuren et al., 2014b).

Adolescents and young adults with CP in GMFCS-E&R levels I-II spend more time sedentary and are less physically active than people without CP, which suggests that this group may have an increased risk for health problems due to less favourable physical behaviour (Maher et al., 2007; Nooijen et al., 2014). Ambulatory children with CP with reduced moderate to vigorous PA and increased sedentary behaviour have an increased risk for elevated blood pressure values (Ryan et al., 2014). Children with spasticity or dystonia may have increased muscle activity in sitting postures and therefore the deleterious health effects of sitting positions might not apply to these children (Innes & Darrah, 2013). However, Verschuren et al. (2014b) have shown

that children with spastic CP across all GMFCS-E&R levels had energy expenditures higher than 1.5 METs during standing, while the energy expenditure in sitting with or without support was lower than 1.5 METs. These findings imply that the definition of sedentary behaviour can be applied for children with spasticity and that changing children's position to a standing position was light activity and reduced sedentary behaviour. The authors suggest that integrating standing work stations in classrooms and using standing desks or standing frames for children of all GMFCS-E&R levels may contribute to the accumulation of light activity.

## **Physical Activity Referrals**

Although there is evidence for the health benefits of PA, promoting PA is a challenge for the healthcare systems as they have to translate the evidence into effective and naturally integrated daily PA activities. Prescribing evidence-based treatment that is known to be most effective and with the fewest side effects is traditionally used in medicine. This approach of using written prescriptions for PA has become common in several of the Western countries. In Sweden, Physical Activity on Prescription (FaR or PAP) or Physical Activity Referral (PAR) has become an accepted concept where the prescription is adjusted to the individual and corresponds to conventional treatment. The prescription can be a simple or comprehensive written suggestion of an activity, with or without a supportive structure from the prescriber or an activity leader. The prescription, or the referral, normally refers people to structured facility-based activities, such as attending gyms, or to self-administrated community-based activities, such as cycling (Swedish National Institute of Public Health, 2010). Individualised PAR was found to increase PA levels for six months or longer and influenced cardiovascular risk factors positively (Kallings et al., 2008; Kallings et al., 2009). The evaluation of effectiveness of PAR implemented in routine primary health care in 6300 persons during a 2-year period showed increased PA and decreased inactivity as well as increased habitual PA (Leijon et al., 2009).

## **Motivational Interviewing**

Motivational Interviewing (MI) explores people's own arguments for change and was developed for those with drinking problems (Miller, 1983). MI is a person-centred, directive therapeutic way where the interviewer enhances readiness for change by supporting people to explore and resolve ambivalence. The interviewer tries to evoke expressions of a person's reasons, desire and ability as well as the need for change.

This 'change talk' includes responding with active listening by the interviewer. MI can be used independently or integrated into an intervention (Hettema et al., 2005). Using MI in combination with PAR to help people change their lifestyles by generating interest and motivation and by preparing and helping a person to initiate change is recommended by the Swedish National Institute of Public Health (2010).

## Participation

One dimension of participation is to physically be there, but it also includes what one is doing when in that environment. This is what the ICF and ICF-CY have defined as a person's involvement in a life situation (WHO, 2001; WHO, 2007). This definition includes involvement in everyday activities such as leisure, school and household activities for children. Being active and involved in freely chosen activities are important for socialising with peers, exploring personal interests and enjoying life (Simpkins et al., 2005). The degree and the child's subjective experience of engagement and the social aspects of being there are other dimensions of participation (Maxwell et al., 2012). Being a part of something, having a social connection, and control have all been described as important issues for people with disabilities (Hammel et al., 2008). The engagement in social situations is commonly described as social participation. Social participation in different contexts increases children's skills to interact and to play with others (Bedell & Dumas, 2004; Hjelle & Vik, 2011). Children may consider themselves to be participating in activities and to be engaged without actually performing the activities, which is an interesting view on participation (Pereira et al., 2010).

### **Participation of children with physical disabilities**

Children with PD participate in activities less frequently than children without disabilities (Imms et al., 2008; Kang et al., 2014), which causes less opportunities for building friendships and may result in feeling socially isolated (Frostdad & Pijl, 2007). The amount of participation is associated with gross motor and communicative function and with the environment (Bult et al., 2011). The findings of a multi-centre cross-sectional European study has shown differences in frequency of participation in everyday life between countries, which might depend on environmental factors (Michelsen et al., 2009). The authors claimed that the use of adaptations, assistance or environmental adjustments can enhance participation. Palisano et al. (2009) found

that adolescents and young adults with CP aged 13-21 years participate in more activities outside of their home and with friends or other children than children with CP aged 6-12 years. This is an important aspect of social participation and age needs to be considered when planning interventions. The authors even found gross motor function classification (GMFCS-E&R) to be decisive for participation in activities outside of the home with a higher percentage of children, adolescents and young adults in levels I, IV and V and a lower percentage in levels II and III. Physiotherapists should take the role as consultants for accessibility and assistive devices and as advocates for inclusive environments according to the authors (Palisano et al., 2009). Participation of children and adolescents with disabilities can be increased by measuring and promoting meaningful outcomes in the individual, family, and environmental components (Carey & Long, 2012).

### *Participation in physical activities*

Children with CP/PD tend to participate in lower intensity PA, to choose more passive activities with less variety and to have more sedentary time. Participating in PA promotes physical and mental health for children with PD. An active lifestyle and increased participation in habitual PA are recommended for promoting the health of children with PD as CP, and replacing sedentary time with light PA might be a beneficial way to reach health-enhancing PA (Verschuren et al., 2014a). In interviews with adolescents with CP and their parents environmental and personal factors were either positive or negative factors for PA participation, whereas limitations related to their disability such as pain, fatigue and muscle power functions were seen as barriers (Shimmell et al., 2013). In interviews with adults with disabilities, with architects, fitness professionals and city planners several factors affecting PA participation were identified: the built and natural environment; economic issues; emotional, psychological and equipment barriers; information-related barriers, laws and policies; professional knowledge, education and training issues; attitudes towards people with disabilities; availability of resources (Rimmer et al., 2004). Motivation, enjoyment, self-efficacy, lack of interest, and low outcome expectation are described as personal factors associated with participation in PA and activity adherence, in addition to age, race and attitude towards exercise (Feehan et al., 2012; Fowler et al., 2007; Lui & Hui, 2009). Disability and health variables such as pain and body weight, programme content and environment effects such as family support, accessibility of exercise equipment and costs all contribute to the disparity in PA participation (Lui & Hui, 2009).

### *Participation in leisure activities*

According to the participation domain of the ICF-CY participation in leisure activities consists of play, sports, culture, crafts, hobbies and social activities (Bult et al., 2011). Leisure activities can be described as freely chosen activities that are performed when the child is not involved in self-care or at school. They are defined as everyday activities in all sporting, entertainment, learning and religious expressions for children (King et al., 2003; Majnemer, 2009). In order to explore personal interests, socialise with peers, develop skill competencies and enjoy life, it is essential to be actively involved in freely chosen leisure activities (Simpkins et al., 2005). Leisure activities can be structured or unstructured. Structured activities are organised and commonly overseen by adults, and unstructured activities are spontaneous and unsupervised (Fletcher et al., 2003). According to the UN convention on the rights of the child (1990) participation in leisure activities is a human right for every child and contributes to children's physical, mental and social well-being and to their quality of life (Shikako-Thomas et al., 2014). Children with physical disabilities participate in fewer leisure activities and would prefer to engage in informal activities compared to children and adolescents without disabilities. They participate in more passive, home-based activities and spend more time with their families than with friends (King et al., 2003; King et al., 2010). Girls with PD might be at risk for being overlooked in recreation programmes because of community attitudes and a lack of staff awareness (Anderson et al., 2005). The authors recommend advertising available activities, education for family, peers and professionals and the use of female role models to reach out to girls. Exploring leisure pursuits for children with PD and their families and promoting engagement in leisure opportunities should be considered a part of health and well-being (Shikako-Thomas et al., 2014).

## Intervention approaches

In addition to the traditional biomedical evaluation of a disorder, child and parent issues are distinct and need careful exploration (Ronen et al., 2011). The ICF is described as a useful framework for understanding and categorising health and function. The authors suggest adding the important dimensions of quality of life to the ICF framework in order to gain information about children's and parents' perceptions and valuations of their well-being.



Different approaches or strategies have been developed for planning and designing interventions for children with PD. When aiming to enhance the level of participation among persons with disabilities the transtheoretical model (TTM) or the social cognitive theory (SCT) are approaches that can be used (Kosma et al., 2002; Lui & Hui, 2009). TTM is a contemporary and integrative motivational theory for behaviour change in a framework consisting of five stages: precontemplation, contemplation, preparation, action and maintenance. The stages can be dynamic or stable, meaning that even if adherence in one stage lasts for a long time, there is always an openness to change. Verschuren et al. (2013) also described stages of change in children with CP's physical activity behaviour: pre-intention, intention and action. The focus is on initial motivation for a change. In this model, the dimension of maintenance is missing, which might be essential when working for lifestyle changes such as physical activity behaviour. Becoming, being and staying physically active are described as important approaches (Claassen et al., 2011). The constructs of SCT are goals guiding specific behaviour, outcome expectancies, perceived self-efficacy, facilitators and barriers (Lui & Hui, 2009). The different approaches might complement each other while focusing on specific aspects.

### **Family- and client-centred approach**

Family-Centred Service (FCS) is described as best practice for children with disabilities on the basis that the parents know what is best for their child (Rosenbaum et al., 1998). The family is engaged as equal partners in rehabilitation services. Each family should be seen as unique with the strengths and needs of every family member considered during the rehabilitation process. The focus is on client-centeredness which is aimed at enhancing and optimising capabilities of the child through improving each family's opportunity for participation (King et al., 2004). The child's and their parents' voices are needed when planning interventions and setting goals.

#### *Context therapy*

Darrah et al. (2011) described context therapy for children with CP as a new intervention approach, where focus is more on adapting the environment or the task than on the child's impairment. While a child-focused approach is aimed at changing components of body function and structure, a context-focused approach consists of these principles: promoting functional performance, identifying periods of change, identifying and changing the primary constraints, and providing opportunities for practice (Law et al., 2007). Both approaches focus on the child's function, and when

comparing child-focused and context-focused interventions for children with CP both approaches have shown to be effective (Law et al., 2011). Law et al. have suggested that the frequency of an intervention is a critical component of a successful intervention, and further studies are needed to identify dose-response relations concerning the amounts of treatment and changes in functional abilities.

### *Family-professional collaboration*

The components of family-professional collaboration include mutual respect and trust, open communication and shared decision-making as well as considering family beliefs, needs and preferences according to interventions (An & Palisano, 2013; Palisano et al., 2012). The recommended strategy for this approach is a 4-step process that begins with determination of both the family and the therapist towards a mutually agreed-upon goal. Secondly, shared planning develops an intervention plan tailored to the child's and their family's needs. The third step is the shared implementation. Together, the parent and therapist implement the intervention after modifying the intervention plan if needed. The fourth step consists of a shared evaluation of child and family outcomes which determines the effectiveness of the intervention and the goal achievement.

King et al. (2014) described the importance of building a collaborative relationship between the families and the therapist for effective communication and family engagement. Therapist strategies may start with trying to understand the family situation and then building a collaborative relationship. Tailoring practice can be seen as part of building a collaborative relationship and therapists should ensure that parents understand the whole process.

# Aims

The overall aim of this thesis was to explore the participation in physical activity in children with physical disabilities such as cerebral palsy (CP). Furthermore, the aim was to evaluate the feasibility and effectiveness of an intervention to increase participation in PA and decrease sedentary behaviour.

The specific aims were:

- To describe the participation in PA of children with CP at school and during leisure time and to identify characteristics associated with PA.
- To explore how children with CP experience participation in physical activities, and describe facilitators and barriers.
- To explore how parents of children with CP experience their child's participation in physical activities and to identify facilitators and barriers for being physically active and reducing sedentary behaviour.
- To develop and evaluate a Physical Activity Referral (PAR) for children with physical disabilities according to its feasibility and how it impacts on participation in PA and sedentary behaviour.



# Methods

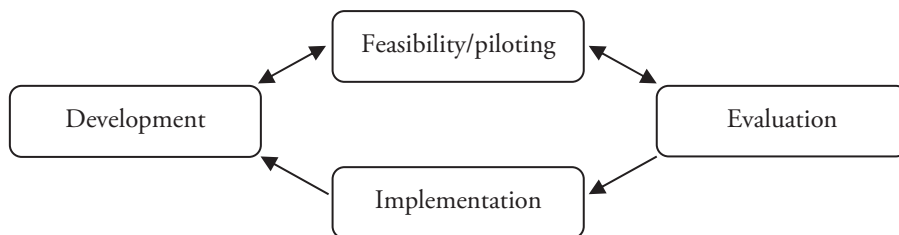
This thesis includes three studies and four papers. Study I resulted in Paper I and Study II resulted in two papers (Paper II and III); Study III resulted in Paper IV. The design of the intervention included in the feasibility study (Study III) is based on the results of Study I and Study II.

## Design

In Study I the total population of children with verified CP aged 7-17 years living in the Skåne region in southern Sweden was studied using cross-sectional data from the National Quality Registry CPUP. Study II had an inductive qualitative design where 16 children aged 8-11 years with CP and 25 of their parents were interviewed. In order to obtain a deeper understanding, the children and the parents were asked to participate in a focus group or in individual interviews. Together with their parents the children decided whether they wanted a proxy (parent or a personal assistant advocating the child). Both children and parents were offered a language interpreter during the interview if needed. In Study III multiple cases were used to assess the feasibility and effectiveness of PAR for 14 children aged 7-12 years with physical disabilities within a real-life context, including 24 of their parents participating in the study. The intervention contained multiple questionnaires and outcome measures that were assessed at baseline and after 8 and 11 months in order to evaluate its feasibility and its utility for a lifestyle change.

Research for children with PD and their families aiming to achieve a more active lifestyle can be seen as a complex intervention. Interventions in health care are associated with several methodological and practical problems (Craig et al., 2008). The Medical Research Council's framework for the development and evaluations of randomised controlled trials (RCTs) for complex interventions to improve health (MRC, 2000) and its revised form (Craig et al., 2008) is used as a methodological framework in this thesis. Complex interventions are usually described as interventions that contain several interacting components (MRC, 2000; Craig et al., 2008). There

is a wide range of complexity and it can be difficult to distinguish between simple and complex interventions. As it is challenging to evaluate complex interventions a phased approach to the development and evaluation is proposed to help researchers define where they are in the research process. The main elements of this process are: Developing an intervention, feasibility/piloting, evaluating, reporting and implementing. The process in the updated and extended guidance (Craig et al., 2008) is more flexible and less linear as shown in Figure 2, i.e. reporting should be seen as an important element of each stage in the process. As best practice the systematic development of the complex intervention is described by using available evidence and appropriate theory. The intervention should be tested in a feasibility and/or pilot study in order to then be exploratory and finally definitive evaluated. The process of implementation of the results consists of dissemination, monitoring and long term follow-up.



**Figure 2.** The development and evaluation process of complex interventions

## Context of the studies

All participants lived in the Skåne region in southern Sweden when they participated in the studies. Approximately 1.2 million inhabitants were living in the county Skåne in 2013 (Statistics Sweden, 2014). The study participants came from different places in the Skåne region; some lived in cities, others in rural areas. In Study I, a total population of children and adolescents with CP were included. In Study II, the individual interviews took place according to each child's preference; seven children were interviewed in their homes, three children at their local child and youth habilitation centre and one child at school. The focus group with five children was conducted at a child and youth habilitation centre. All individual interviews with the parents were conducted in the parents' homes or at a child and youth habilitation centre. Fourteen parents participated in four focus group interviews; according to the parents' wishes or for practical reasons one focus group consisted of six participants,

two of three, and one of two. Three focus group interviews took place at a child and youth habilitation centre, and one at a hospital. In Study III, the families participated in the intervention that took place in their homes, at their local child and youth habilitation centre, in the neighbourhood or where the self-selected activities were offered.

## Study populations

Characteristics of the participating children with PD, mainly CP, are presented in Table 1.

In Study I the age range was 7-17 and the participants are described as children and adolescents. In Study I the total population of children and adolescents with CP in the Skåne region consisted of 364 participants. The age interval of 7-17 years of age at the latest CPUP assessment by the child's physiotherapist before the inventory in January 2011 was chosen as children are school-aged at 7 years and should participate in physical education at school. In addition, children's gross motor function is almost fully developed at the age of 7 years (Hanna et al., 2009) and a decreasing passive range of motion and increasing weight gains have been noticed for several children with CP after school start at the age of 6-7 years (Nordmark et al, 2009; Westbom et al, 2010). The upper age limit before 18 years of age is when most of the adolescents leave the Child and Youth Habilitation Services.

In Study II and III the participating children were up to 12 years old. The studied age interval was chosen because younger school children are more physically active and less sedentary before the transition to adolescence (Corder et al., 2014), and an intervention started before adolescence might influence future PA and sedentary behaviour in terms of staying or becoming physically active according to each child's needs. The parents in Study II and III had different cultural backgrounds regarding ethnicity, gender, language, socioeconomic status and geographical area.

In Study II 16 children with CP aged 8-11 years with various gross motor and cognitive functions were interviewed. Fifteen children were living together with their biological parents; one child was living in a foster home. Twelve children were living in families together with both parents and four with one parent. Four children used augmentative and alternative communication in the form of sign language interpretation with various picture systems such as concrete photos or the graphical language of Bliss Symbolics (Chung et al., 2012; Hammond & Baily, 1976) during the interview. One child had both a language interpreter on the phone and a parent as

a sign language interpreter; one child had only a language interpreter. The five children participating in the focus group had various gross motor functions and none had a need for an interpreter. Nine married or cohabiting couples, six mothers and one father agreed to participate in the study. Twenty-three of the participants were biological parents and two were foster home parents. Eleven parents were born abroad; five of whom requested a language interpreter for the interviews. In Sweden, the social security system provides for personal assistance to facilitate social participation and equality of living conditions and five parents in the present study were employed as personal assistants to their children.

In Study III 14 children with PD (11 with CP and 3 with other PD), aged 7-11 years at baseline, with various gross and fine motor, communicative and cognitive functions were included in the study. In addition, there were 24 parents to the 14 children participating in the study. Nine of the parents were born abroad, and two of them requested a language interpreter for the intervention. Eleven parents were employed as personal assistants for their child.

### **Recruitment for Study II and III**

An invitation letter with information about the respective study was sent by an administrative assistant at the Child and Youth Habilitation Services. In Study II all parents of children with CP aged 8 to 11 (N= 117) received the letter in January to June 2012; in Study III all parents of children with physical disabilities aged 7 to 12 years (N= 347) received the letter in September 2013. The information was given to each child and their parents, including an easy-to-read child-appropriate version.

In Study II a reminder was sent to all parents who had not answered after two weeks. The parents of 26 children answered, and 10 of them declined to participate. The first author contacted the parents of the 16 children for verbal information. In addition, 25 parents to the 16 children chose to participate in the study.

In Study III information was enclosed in the letter which stated that participants would be selected in order to get a maximum variation of different perspectives of PAR. This information was included in case there was an overabundance of families wanting to participate in the study. Six out of 14 included children were recruited through an inspiration day in November 2013, where they could try different PA together with siblings and parents. The parents to 17 children were recruited through the invitation letter with information and 8 of those 17 children were included. Altogether 14 children and 24 of their parents participated in Study III.



**Table 1.** Characteristics of the children in Study I-III

	Study I (N=364) n (%)	Study II (N=16) n (%)	Study III (N=14) n (%)
Gender			
Female	144 (39.6)	9 (56.3)	7 (50.0)
Male	220 (60.4)	7 (43.7)	7 (50.0)
Age (years)			
7-11	180 (49.5)	16 (100.0)	14 (100.0)
12-17	184 (50.5)		
GMFCS-E&R <sup>a</sup> level			
I	158 (43.4)	6 (37.5)	3 (21.4)
II	71 (19.5)	5 (31.3)	3 (21.4)
III	28 (7.7)	2 (12.5)	2 (14.3)
IV	59 (16.2)	2 (12.5)	4 (28.6)
V	48 (13.2)	1 (6.2)	2 (14.3)
MACS level <sup>b</sup>		-	
I	135 (37.1)		7 (50.0)
II	69 (19.0)		2 (14.3)
III	52 (14.3)		3 (21.4)
IV	53 (14.6)		1 (7.1)
V	47 (12.9)		1 (7.1)
Missing information	8 (2.2)		
Cognitive level <sup>c</sup>			
No mental retardation	210 (57.7)	9 (56.3)	8 (57.1)
Mild mental retardation	67 (18.4)	5 (31.3)	2 (14.3)
Moderate to profound mental retardation	87 (23.9)	2 (12.5)	4 (28.6)
Augmentative and alternative communication	-	4 (25.0)	4 (28.6)
Language interpretation	-	2 (12.5)	1 (7.1)
Parents' ethnic origin	-	N=25	N=24
Sweden		14 (56.0)	15 (62.5)
Europe, others than Sweden		3 (12.0)	3 (12.5)
Outside Europe		8 (32.0)	6 (25.0)

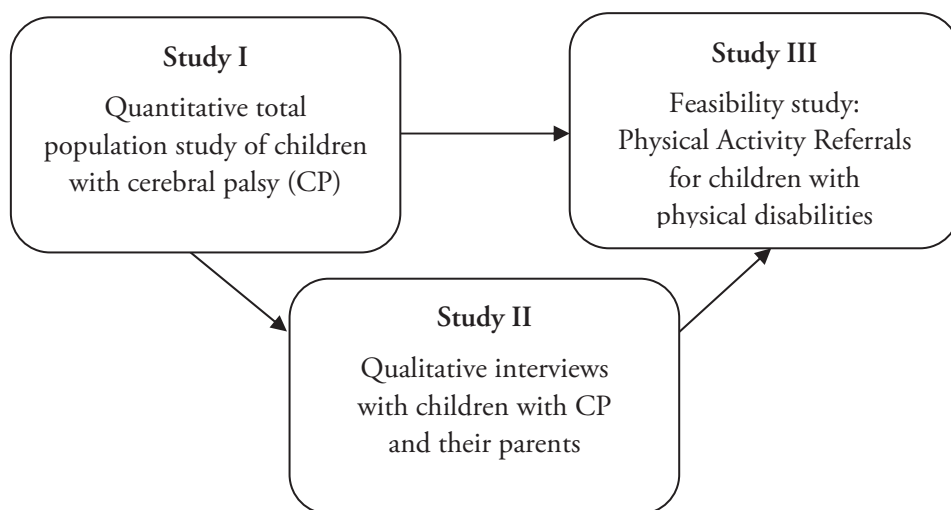
<sup>a</sup>Gross Motor Function Classification System Expanded and Revised

<sup>b</sup>Manual Ability Classification System

<sup>c</sup>International Classification of Diseases, (WHO, 2010b): ICD-10 codes for mental retardation (intellectual disability)

## Procedures and data collection

This thesis started with a quantitative study (Study I) in order to gain knowledge on types of PA and their frequency among children with CP. Qualitative interviews with children and their parents (Study II) then added the dimension of learning about their experiences of PA and sedentary behaviour. Based on the results of Study I and Study II the intervention included in the feasibility study (Study III) was developed (Figure 3).



**Figure 3.** Study process

### Study I

The first study was a population-based observational study with a cross-sectional design which used data from the CPUP database. The data reported by physiotherapists in this study contained information regarding the level of gross motor function classified at the latest assessment for CPUP before 1 January 2011. Further data reported by physiotherapists for the database contained information regarding the frequency with which the child or adolescent actively and regularly participated and performed physical activities in PE at school. The child's local occupational therapist established and reported the manual ability level classified at the latest assessment for CPUP before 1 January 2011. The paediatrician determined the CP-diagnosis and subtype and also collected information about gender, epilepsy,

visual and hearing impairment, anthropometrics and cognitive function for the CPUP-database.

## Study II

In Study II interviews were carried out from March to June 2012. Sixteen children and 25 parents were offered the choice of whether to participate in a focus group or in an individual interview which enabled participation for all participants.

Eleven children chose to participate in individual interviews and five children in a focus group. The individual interviews were conducted by the first author and lasted between 30 and 50 minutes. However, the focus group with five children, conducted at the child and youth habilitation centre, took 65 minutes and was moderated by the first author of Paper II, with the second author as an observer. Refreshments and informal chatting took place prior to each interview in order for the children to become more comfortable and relaxed.

Three parents chose to be interviewed individually, and four couples chose to be interviewed in pairs. When being interviewed as couples, both mothers and fathers were asked to answer the questions individually. All individual interviews were conducted by the first author of Paper III either in the parents' homes or at a child and youth habilitation centre and lasted between 40 and 75 minutes. They were all moderated by the first author, three of them with the second author, and one with a doctoral student as an observer and lasted between 90 and 120 minutes. Both the individual and focus group interviews with the parents started with the parents giving a short presentation of themselves and their child.

In order to get a better understanding of what 'physical activity' meant to each child and to each parent, several minutes were spent discussing their own experiences and habits regarding physical activities.

The children were asked to narrate their perceived experiences of PA in different environments – including at home, during leisure time, at school and at their child and youth habilitation centre. To maintain a playful manner throughout the interview and to facilitate talking about their experiences, pictures with different activities and/or small play figures were used while talking (Figure 4). The parents talked about how they perceived their child's experiences of participation in PA in the different environments. They were also asked to consider what they felt were facilitators and barriers to their child being physically active.



**Figure 4.** Examples of toy figures, pictogram and pictures used in Study II

During the interviews, additional questions were asked which were aimed at encouraging further responses and more reflective narration such as, ‘Can you tell me more about this?’ They were also asked ‘What do you mean?’ or ‘Can you give me an example?’ The procedure was the same in individual and focus group interviews, except that the participants in the focus groups were discussing and reflecting together. In addition to the interview, the parents filled in a sociodemographic questionnaire, giving characteristics of themselves and their child with CP. Information about gross motor and cognitive function was provided according to the parents’ perceived opinion. All interviews were audio tape-recorded and transcribed verbatim by the first author.

### Study III

The intervention period was from November 2013 to September 2014. The intervention included a written agreement, PAR, between each child, their parents and the physiotherapist, based on Motivational Interviewing (MI), the Canadian Occupational Performance Measure (COPM) and Goal Attainment Scaling (GAS). The intervention was assessed through the use of sociodemographic, clinical, PA and costs and time spent questionnaires and by a questionnaire to evaluate each part of the intervention. In addition, the Gross Motor Function Measure (GMFM-66) was performed; physical activity and heart rate monitors and time-use diaries were used. The physiotherapists who did the assessments and supported the families during the intervention were the first and last author of Paper IV and a project assistant.

#### *PAR*

The written personalised and individual agreement was about self-selected physical activities which could be used to enhance each child's habitual PA and reduce sedentary behaviour (Figure 5).

Motivational interviewing (MI) (Hettema et al., 2005; O'Halloran et al., 2014) was used to identify each child's and parents' desires and readiness for change. MI was integrated into the intervention and used throughout the study period by the physiotherapist as a counselling and guiding method towards a more active lifestyle.

The Canadian Occupational Performance Measure (COPM) (Law et al., 2005), adapted for children (Cusick et al., 2007), is an individualised outcome measure with good validity, good reliability and that is sensitive to change. COPM was used to identify important physical activities for each child and to capture the child's self-perception of performance in everyday living over time. Performance problems, concerns and issues according to physical activities were identified and the performance and satisfaction levels in self-care, productivity and leisure were ranked and rated from the child's perspective on a Visual Analogue Scale (VAS) 1-10 by using a scoring card. Performance and satisfaction scores were reassessed after 8 and 11 months to detect changes.

Goal Attainment Scaling (GAS) (Kiresuk and Sherman, 1968; King et al., 1999; Turner-Stokes, 2009) is a validated method of scoring the achievement of goals that were set. Each child had their own outcome measure, developed collaboratively with the child, their parents and the physiotherapist (Palisano, 1993). The extent to which the individual goals were achieved was scored by the child and its parents on a 5-point scale ranging from -2 to +2. Baseline was set at -2; the expected level of attainment

after the intervention was at 0 and the most favourable outcome at +2. In the agreements -2 on the scale was named 'baseline', -1 was named 'better than baseline', 0 was the 'goal', +1 was named 'better than expected', and +2 'much better than expected'. The goals according to GAS were continuously followed up and evaluated after 8 and 11 months as a main outcome measure. Some children changed their self-selected PA or they wanted to adjust their goal during the intervention, which was done in collaboration with the child, their parents and the physiotherapist.

<p>Agreement about Kim's physical activity</p> <p><b>Choice for physical activity 1:</b> 'Alla-kan-gympa' (gymnastics group)</p> <p><b>Reason for physical activity:</b> Reduce sedentary time <input checked="" type="checkbox"/>    Increase daily activity <input type="checkbox"/>    Have more energy <input type="checkbox"/>    Get more fit <input type="checkbox"/> Become stronger <input type="checkbox"/>    Increase endurance <input type="checkbox"/>    Become smoother <input type="checkbox"/>    Maintain health <input type="checkbox"/>    Feel better <input checked="" type="checkbox"/> Have fun <input checked="" type="checkbox"/>    Be with friends <input checked="" type="checkbox"/>    Do something together with family <input type="checkbox"/>    Other: Sleep better <input checked="" type="checkbox"/></p> <p><b>Start date:</b> 14th January 2014      <b>Follow up:</b> June 2014 Number of days/week: 1    Minutes/session: 60 min    Estimated intensity: Low – moderate</p> <p><b>Goal with the physical activity:</b> To participate in gymnastics to music together with other children 1x/week</p> <p><i>Baseline:</i> Participates in no physical activity with other children <i>Better than baseline:</i> Participates in gymnastics to music with other children 1x/month <i>Goal:</i> Participate in gymnastics to music together with other children 1x/week <i>A little better than expected:</i> Participates in gymnastics to music with other children 1x/week, occasionally sleeps better <i>Much better than expected:</i> Participates in gymnastics to music with other children 1x/week, sleeps better in general</p> <p>Date: Signature child: _____ Signature parents: _____ Signature physiotherapist: _____</p>
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**Figure 5.** Example of a written agreement, presented by using a pseudonym

Individually designed logbooks (Figure 6) enabled each child to document how often they participated by using self-selected stickers, by ticking a smiley which corresponded with their enjoyment, and by writing down individual comments.

My self-selected activities							
		<i>Cycling 3x/week</i>					
		<i>Swimming 1x/weekend</i>					
	Monday 20 January	Tuesday 21 January	Wednesday 22 January	Thursday 23 January	Friday 24 January	Saturday 25 January	Sunday 26 January
I have participated in my activity							
I enjoyed it							
Comments							

**Figure 6.** Example of an individual logbook

### *Questionnaires*

Characteristics of each child and their parents were recorded in sociodemographic and clinical questionnaires, including age, gender, type of physical disability and classifications of the child’s gross motor function, manual ability, communication and cognitive function. The Gross Motor Function Classification System Expanded and Revised (GMFCS-E&R), the Manual Ability Classification System (MACS) and the Communication Function Classification System (CFCS) are five level classification systems for children with CP, where level V implies the most severe function limitations (Palisano et al., 2007; Eliasson et al., 2006; Hidecker et al., 2011; Compagnone et al., 2014). The cognitive function was classified according the International Classification of Diseases (ICD-10) into the groups of: no mental retardation, mild mental retardation and moderate to profound mental retardation

(WHO, 2010b). The parents filled in the sociodemographic questionnaire during the first home visit with support of the physiotherapist if necessary. The physiotherapist then filled in the clinical questionnaire with verbal responses from the child and their parents.

The frequency of physical activities at school and leisure time and the physiotherapeutic interventions of the child were recorded according to the questionnaire used in the National Quality Registry and CP Follow-Up Programme (CPUP, 2014) by the physiotherapist through interviewing the child and their parents. To estimate the time the child spent being physically active with light, moderate and vigorous intensity and how much time the child spent sedentary over the past seven days, the International Physical Activity Questionnaire (IPAQ) was used and filled in by the physiotherapist in collaboration with the child and their parents (Booth, 2000). In a multi-country reliability and validity study the results showed that the IPAQ measurement properties were comparable to other established self-report PA measures (Craig et al., 2003).

A questionnaire was designed to evaluate each part of the intervention on a VAS, where 1 represents 'very difficult/bad' and 10 'extremely easy/good'. This questionnaire was filled in by the physiotherapist in collaboration with the child and their parents at 8 and 11 months. In addition, questionnaires were designed including the costs of the self-selected PA and time spent there, and included costs and time spent for travelling to the activity. These questionnaires were filled in by both the parents and the physiotherapists.

#### *Gross Motor Function Measure*

The Gross Motor Function Measure 66 (GMFM-66) was used for all participating children. GMFM-66 is a 66-item clinical measure designed and validated to assess gross motor function in children with CP and to measure changes over time (Avery et al., 2013; Hanna et al., 2009; Russell et al., 2013). The GMFM-66 assessments were made by the first author and the project assistant. The assessments were video-recorded and independently scored from the videos by two of the physiotherapists. The scores were translated and calculated with the Gross Motor Ability Estimator (GMAE-2) scoring software for the GMFM-66, where the total score, CI and percentiles were calculated.

#### *Physical activity monitors and time-use diaries*

Physical activity monitors – accelerometers – are feasible and useful to measure habitual PA in adolescents with CP in all gross motor function levels (Gorter et al., 2012). Capio et al. (2010) validated the uniaxial accelerometer (MTI) for ambulatory



children with CP. The validity of the accelerometer ActiGraph (Actigraph, USA) as a objective measure of PA for ambulatory children and adolescents with CP was confirmed by Clanchy et al. (2011).

As this study included children who did not walk, the triaxial ActiTrainer (Actigraph, USA) was chosen for its ability to measure activity in lying, sitting or standing position over a 7-day period. Cut-points for activity levels developed for children at  $\leq 100$  counts/minute for sedentary time (ST), 101-499 counts/minute for light intensity (LPA),  $\geq 500$  counts/minute for moderate intensity (MPA) and  $\geq 4000$  counts/minute for vigorous intensity (VPA) were used (Freedson et al., 2005). The children wore the ActiTrainer on their hips during all waking hours for a 7-day period according to best practices (Gorter et al., 2012; Pfeiffer et al., 2009; Trost et al., 2000). In addition, they had a heart rate monitor around their chest during the 7-day period (Figure 7).



**Figure 7.** Accelerometer and heart rate monitor

Time-use diaries with a time-geographical approach (Ellegård, 1999) were filled in by the child, with support of a parent or personnel at school if necessary, to learn how the children used their time and space in their everyday life. Information about all activities that the child participated in during one week day and one weekend day were recorded. Additionally, what physical intensity the activities were classified as, and where, with whom and in what mood the child was doing the activities was also collected.

## Data analyses

In Study I and III Excel 2003 and the software SPSS<sup>TM</sup> version 19.0 (Study I) and 22.0 (Study III) were used in the statistical analyses. Descriptive statistics were used for presenting the child's and the parents' demographic and clinical data in all three studies. In Study I the type of PA and the frequency of participation in PA at leisure and school, and in Study III the measurements were described by using descriptive statistics (Björk, 2010).

In Study I the analysis of the three components of PA, PE, leisure time and physiotherapeutic intervention, the corresponding scales measuring the frequency of PA were treated as ordinal. Consequently, the evaluation of the effect of characteristics GMFCS-E&R, MACS, age group, gender, BMI category, epilepsy and cognitive function level on the frequency of PA was carried out using proportional odds regression. Proportional odds were chosen in order to keep the ordinal scale of the outcomes that were analysed. The estimates produced are referred to as proportional odds ratios (POR). An estimate above one indicates an increasing effect on the frequency of PA; the greater the estimate, the greater the effect. An estimate below one indicates a decreasing effect; the smaller the estimate the greater the decrease in frequency. All statistical tests are two-sided and P-values below the standard value of 0.05 were considered statistically significant.

In Study II content analysis was used to understand the underlying meaning of the interview texts with the children and with their parents (Downe-Wamboldt, 1998; Graneheim & Lundman, 2004). The verbatim transcribed interviews were read several times, with the aim of the study in mind, by all three authors of Paper II and III in order to obtain a sense of the content. From all interviews, meaning units relating to the children's experiences of PA were extracted. A meaning unit was described as a constellation of sentences relating to the same central meaning. Taking the whole context into consideration, the meaning units were condensed and designated as codes. After determination of similarities and differences among the codes, sub-categories were abstracted from all codes sharing a commonality. Agreement about codes and sub-themes was achieved after a process of reflection and critical discussion among all authors, moving between the whole and the parts of the texts. During the whole process, from planning and conducting the interviews, to all steps of the analysis, all of the authors, who had longstanding experience of working with children with disabilities or chronic diseases, discussed their pre-understanding. This also meant that they reflected about the ways in which the pre-understanding might affect the analysis, abstraction and interpretation. In addition, the results were discussed in a multidisciplinary research group.

In Study III the estimated and measured PA levels through IPAQ, accelerometer ActiTrainer and time-use diaries, and the outcome measures of COPM, GAS and GMFM-66, were described by using descriptive statistics. When using the accelerometer ActiTrainer children with  $\geq 5$  hours of monitoring time on  $\geq 4$  days were included for analyses (Gorter et al., 2012); in a second step, and according to the recommendations of Trost et al. (2000), children with  $\geq 5$  hours of monitoring time on  $\geq 2$  days were included.

## Ethical considerations

All studies were approved by the Regional Ethical Review Board at Lund University, Sweden: LU-443-99 (Study I), LU-Dnr 2011/350 (Study II), and LU-Dnr 2013/521 (Study III).

In Study II the children gave their assent and their parents gave written, informed consent before the interviews took place in accordance with the World Medical Association (WMA) Declaration of Helsinki (2008). The children and their parents were guaranteed confidentiality and the right to discontinue the interview at any time, and they agreed to the interviews being tape-recorded.

In Study III the children gave their assent and all parents submitted written, informed consent at the first home visit in accordance with the Declaration of Helsinki (WMA, 2013). The families who wanted to participate but were not selected for this study were informed that they were welcome to participate in a future large scale study if the results of the present study were promising. This information was included in the invitation letter that all families received. The participating children and their parents were guaranteed confidentiality and the right to discontinue their participation in the study at any time, and they agreed to being video-recorded as part of the GMFM-66.

Children, and particularly those with disabilities, are a vulnerable population and ethical issues need to be considered in any kind of research. As basic ethical principles in studies on children their autonomy, beneficence, non-maleficence and justice has been identified (Merlo et al., 2007).

### Autonomy

Each child's autonomy is related to respect for the child and their freedom to make decisions and their right to receive information (Merlo et al., 2007). Each child should be informed about the study aims, methods and possible risks and benefits. In addition, the children in Study II and III were encouraged, whenever possible, to communicate their assent for participating in the study. However, all children might not fully understand the research information due to their age or impairment-related physical and psychological development. Therefore, that their parents also gave written informed consent for their child's study participation.

## **Beneficence and non-maleficence**

The scientific benefits as well as possible benefits for the participating children (beneficence) are supposed to outweigh potential risks, harm or inconveniences (non-maleficence) and their well-being should be secured (Merlo, 2007; WMA, 2013). The interviews with the children (Study II) lasted not longer than one hour and were performed in a playful manner whenever possible and according to the child's interests. In Study II and III some time was spent in the beginning to get to know each other in order to minimise inconvenience. Any possible inconvenience was considered to be minor and the results of the interviews and the intervention study can be of value to the planning of a future randomised controlled trial.

## **Justice**

The participants were recruited by respecting the principles of non-discrimination (Beauchamp, 1994). No child, adolescent or parent was excluded because of ethnicity, gender or socioeconomic factors. In Study I a total population was included and in Study II all children were included whose parents sent in the letter of interest. In Study III the parents who wanted their child and themselves to participate but were not selected for this study were informed that they were welcome to participate in a future large scale study.

# Results

The results are presented according to the overall aim of this thesis, which was to explore the participation in physical activities among children with CP and other PD. The main focus is based on the specific aims of the studies which describe and explore participation in PA in different contexts and identify facilitators and barriers to participation both quantitatively and qualitatively (Study I and II). In Study II the interviews with the children revealed ten sub-categories containing facilitating and inhibiting factors and generated two main categories. Based on whether they were considered facilitators or barriers for PA the sub-categories were then compiled into the categories. From the interviews with the parents one main category emerged from five subcategories (Table 2). In addition, we evaluated the feasibility and effectiveness of an intervention aimed at increasing participation in physical activities and decreasing sedentary behaviour (Study III).

## Participation in physical activities in different contexts

Children participate in physical activities in different contexts, and in this thesis the following contexts have been explored: in or outside the child's home, in their neighbourhood or where an organised leisure PA took place, and at their school. Physiotherapeutic interventions might include PA and children with PD might be physically active in groups organised at their child and youth habilitation centre. This context was also explored in Study I and II.

### Findings from CPUP-data

The participants in Study I were 364 children and adolescents with CP aged 7-17 years. Half of them participated in habitual physical activities during their leisure time, with differences between GMFCS-E&R levels: 65% of the children and adolescents in level I participated in a habitual leisure activity compared to 21% with the most severe impairments in level V. The most frequent activities in all gross

motor levels were swimming and horse riding. Children and adolescents in GMFCS-E&R level I had the greatest variation in these types of activity. Children and adolescents in GMFCS-E&R level III were significantly less likely to participate in regular PA in leisure time than children and adolescents in level I.

In total, 87% actively participated in PE at school. However, differences were seen in GMFCS-E&R levels and only 50% of the children and adolescents in level V participated in PE at school.

In the total group, six out of ten children and adolescents had some kind of physiotherapeutic intervention and differences were seen in GMFCS-E&R levels, where children and adolescents with more severe gross motor function limitation received most physiotherapeutic interventions. Children and adolescents with an intellectual disability received less physiotherapeutic interventions than those without.

## **Experiences of the children and their parents**

The 16 children with CP aged 8-11 years in Study II were naturally physically active during leisure time or at school. PA within the family or during leisure time were often initiated by the parents but were often in accordance with the child's wishes. They participated in school activities, such as school sports and excursions, without question but expressed that they had less little say in the choice of activity. Competitive games could be exciting and fun, but could also make the child feel as if they were singled out. The children enjoyed physiotherapeutic interventions when they involved playing or training at the child and youth habilitation centre together with the physiotherapist or other children, especially when they had a say about the activities. Adjusting assistive devices or performing measurements often had to be done, but the children said that they were used to it.

All 25 parents of the 16 children experienced difficulties in finding appropriate physical leisure activities for their child, which would preferably be within a reasonable distance from where they were living. Their family life was affected because the parents had to arrange the ordering and adjustments of assistive devices, home programmes with stretching or exercising, and transportation for the child to different activities or medical assessments and treatments. The parents wanted competent personnel and group leaders for their child's PA, both during leisure time and at school. The child's local child and youth habilitation centre was seen as a resource for getting support and treatment for their child and sometimes for themselves, but they asked for more help in meeting other children similar to their own, including meeting other parents at the child and youth habilitation centre.

## Facilitators and barriers

In Study I and II facilitators and barriers to participation in physical activities were identified and in Study II also to reduce sedentary behaviour.

### Findings from CPUP-data

In addition to the characteristics presented in Table I, thinness was seen in 10% of the 364 participants and obesity in 9%. Visual impairment was reported in 14%, hearing impairment in 3% and 32% had epilepsy. Of all characteristics included in the POR calculations, according to GMFCS-E&R gross motor function proved to be the strongest.

Thinness was a statistically significant characteristic for low participation in regular PA in leisure time. Moderate to profound mental retardation showed a tendency to low odds for PA in leisure time whereas no obvious associations were seen between age group, gender, MACS or epilepsy and regular physical leisure activity.

Age proved to be a characteristic affecting participation in PE at school; children between 7 and 11 years of age had a statistically significant higher chance of participation in PE at school than the older children. Severe functional limitation was significantly associated with low participation; children and adolescents with GMFCS-E&R level V had statistically significant lower odds for participating in PE at school than their peers with GMFCS-E&R level I. A statistically significant higher participation in PE at school was seen in obese children and adolescents compared to those without obesity. No obvious associations were seen between gender, MACS, cognitive function or epilepsy and the odds for PA in PE at school.

Although the proportion of children receiving physiotherapeutic intervention was highest in GMFCS E&R level III (93%) the chance for receiving physiotherapeutic intervention increased with the severity of gross motor function limitation, GMFCS E&R level V with the highest POR, when the other characteristics were included in the analysis. Manual ability also influenced the odds. Children and adolescents with MACS level II or III received more physiotherapeutic intervention than children and adolescents with MACS level I. Intellectual disability reduced the odds for receiving physiotherapeutic intervention with a statistically significant reduction for children with moderate to profound mental retardation. No significant associations were seen between age group, gender, obesity, thinness or epilepsy and the odds for receiving physiotherapeutic intervention.

## Experiences of the children and their parents

Both the children and their parents were aware that PA was good for the child. The children wanted to be asked what PA they enjoyed and to get the opportunity to try self-selected activities whilst getting the right support in all contexts but especially in PE at school. They wanted to have fun with their family and friends when being physically active, and to enjoy the sensation of speed.

Parents asked for competent personnel and persons to be available to their children's ability to participate in PA. Having friends to be physically active with was seen as a facilitator, but for children who had difficulties making friends it was described as a barrier towards participating in PA. Parents wanted help with getting friends for their children. Family culture formed by the parents' attitudes, ethnical background and previous life experiences proved to be both facilitators and barriers for their child's motivation to be physically active.

Whereas the children often thought in terms of their present experiences, the parents often looked into the future and worried about the challenges that their child might face. The sub-categories based on the main categories are presented in Table 2.

Table 2. Categories and sub-categories emerged from the interviews

Children's experiences	Children's experiences	Parents' experiences
Being physical active, because...	Being physical active, but ...	Protecting and pushing towards independence
Enjoying the feeling	Getting tired and experiencing pain	Belonging and taking space in the family
Being capable	Something being wrong with my body	Important persons facilitating and hindering
Feeling of togetherness	Being dependent on others	Friends important but hard to get
Being aware it is good for me	Not being good enough	Good for the body but challenging
Using available opportunities	Missing available opportunities	Availability and opting out possibilities



## Physical Activity Referrals

The feasibility of PAR and its effectiveness on sedentary behaviour and participation in PA were examined for 14 children with PD and their parents in Study III. Each child participated in 1-3 self-selected physical activities. Seven children who tried new physical activities in a group made new friends and, according to the intervention evaluation, all 14 improved their abilities. Several families chose family activities such as going swimming at the weekends or improving everyday activities like cycling to school.

### Feasibility

There were no dropouts in the study and all families fulfilled their participation in the intervention throughout the study period. The acceptability of PAR was generally good according to the evaluation questionnaire. The families scored the overall experience of PAR high. The children enjoyed scoring the COPM themselves or with the help of their parents and filling in the logbooks as well as performing the GMFM-66. Writing the agreement, GAS and writing in the time-use diaries was accepted. COPM was experienced as a helpful assessment by the parents in identifying important activities and in detecting changes in both the performance and satisfaction for each child. Completing the questionnaires was experienced as neither good nor bad by most children and parents. However, completing the IPAQ with support of the physiotherapist was experienced by the parents as being difficult to score. The parents found it hard to know when their child was physically active in a low, moderate or vigorous level or how much sedentary time the child had during individual days or throughout a 7-day period. The families were also concerned with using the physical activity monitors. The children were not bothered by wearing the accelerometer on the hips, whereas the heart rate monitor often became loose and slipped down a lot. The parents intervened and tried to support their child with wearing the accelerometer and heart rate monitor. However, the children felt that their peers were looking at them because of the accelerometer and two children refused to use them at all.

### Effectiveness

The effectiveness for increased participation in PA and decreased sedentary behaviour of PAR was evaluated. According to the logbooks, COPM and GAS, all children

increased their frequency and duration of participation in physical activities from baseline. COPM and GAS scores increased from baseline. According to GAS, some goals had to be adjusted during the intervention; the goal was either already achieved after a short time or the child changed the selected activity from one to another one. The GMFM-66 was performed by all children at baseline. At 8 months eleven children performed the measurement; two children had recently undergone unplanned surgeries followed by movement restrictions and one child declined due to lack of time. The outcome score of the GMFM-66 improved in six children, and two of the four children that maintained their score scored 100% at baseline.

The families spent between 17 and 100 hours for the intervention and between one and 34 hours for travel, depending on the self-selected activity. Participating in a group activity once a week as their self-selected or participating several times a day in transfers from sitting to standing activity can explain these differences. In addition, the costs varied among the families depending on potential fees for the activity and any purchased equipment. Time spent and travel costs for the physiotherapist varied but not to the same extent as the families. Some families opted out certain activities because of travel distances, inconvenient time for the family or costs.

The IPAQ for each child was completed by all parents with support of the physiotherapist at baseline and at 8 months during the home visits. Thirteen families wrote a time-use diary for two days at baseline and 11 families at 8 months. The estimated time for each child in the different physical activity levels (sedentary time, light, moderate and vigorous intensity) varied between baseline and 8 months, as well as between the estimations in the time-use diaries and the IPAQ.

Twelve children accepted wearing the physical activity monitor ActiTrainer for one week at baseline and 11 children at 8 months. Eight children were physically active in moderate-vigorous levels for more than 60 minutes per day at both assessments. The children spent most of their daytime sedentary, up to 3 hours a day in light PA, and their time spent daily in moderate PA varied between the children from less than half an hour to more than 4 hours; vigorous PA was rarely seen in this group. The median for the 12 children at baseline was 84 minutes daily for moderate-vigorous PA and for the 11 children at 8 months the median was 106 minutes.

All families got a feedback after 11 months about their child's estimated and measured PA levels as shown in two examples (Table 3 and Table 4).

Table 3. Estimated and objective measurements of physical activity, example 1

	ActiTrainer <sup>a</sup>	ActiTrainer	Time-use diary <sup>b</sup>	Time-use diary	IPAQ <sup>c</sup>	IPAQ
	December 2013 (min/day)	April 2014 (min/day)	November 2013 (min/day)	April 2014 (min/day)	November 2013 (min/day)	May 2014 (min/day)
ST <sup>d</sup>	344 min (ca 5.5 h)	411 min (ca 7 h)	883 min (ca 14.5 h)	1028 min (ca 17 h)	240 min (4 h)	240 min (4 h)
LPA <sup>e</sup>	105 min (ca 1.5 h)	96 min (ca 1.5 h)	LPA + MPA: 538 min (ca 9 h)	LPA + MPA: 413 min (ca 7 h)	540 min (9 h)	540 min (9 h)
MPA <sup>f</sup>	271 min (ca 4.5 h)	178 min (ca 3 h)			120 min	120 min
VPA <sup>g</sup>	5 min	5 min	20 min	0 min	0 min	0 min

<sup>a</sup> Physical activity monitors, accelerometers (ActiTrainer, Actigraph, USA), excluding sleeping time;

<sup>b</sup>Including sleeping time; <sup>c</sup>International Physical Activity Questionnaire, excluding sleeping time;

<sup>d</sup>ST: sedentary time; <sup>e</sup>LPA: light physical activity; <sup>f</sup>MPA: moderate physical activity;

<sup>g</sup>VPA: vigorous physical activity; min=minutes; h=hour

The PA monitor, accelerometer ActiTrainer, was used during the daytime and the results are presented in all PA levels: sedentary time, light PA, moderate PA and vigorous PA. The time-use diaries were designed so the participant could fill in all activities during a 24-hours period and were used to estimate the PA levels for each activity, whether it was sedentary, light/moderate or vigorous PA. The IPAQ was used during the daytime including all estimated PA levels.

In example 1 (Table 3) the child was physically active in light and moderate intensity at baseline for 6 hours according to the ActiTrainer, for 9 hours according to the time-use diaries and for 11 hours according to the IPAQ. After 8 months the child's PA in moderate intensity decreased 1.5 hours according to the ActiTrainer, the PA in light and moderate intensity decreased 2 hours according to the time-use diaries and did not change according to the IPAQ.

Table 4. Estimated and objective measurements of physical activity, example 2

	ActiTrainer <sup>a</sup>	ActiTrainer	Time-use diary <sup>b</sup>	Time-use diary	IPAQ <sup>c</sup>	IPAQ
	December 2013 (min/day)	May 2014 (min/day)	December 2013 (min/day)	May 2014 (min/day)	December 2013 (min/day)	June 2014 (min/day)
ST <sup>d</sup>	694 min (ca 11,5 h)	562 min (ca 9.5 h)	1038 min (ca 17 h)	1210 min (ca 20 h)	180 min (3 h)	180 min (3 h)
LPA <sup>e</sup>	61 min	111 min (ca 2 h)	LPA + MPA: 295 min (ca 5 h)	LPA + MPA: 90 min (1.5 h)	600 min (10 h)	600 min (10 h)
MPA <sup>f</sup>	27 min	51 min			60 min	60 min
VPA <sup>g</sup>	1 min	0 min	8 min	180 min	30 min	60 min

<sup>a</sup> Physical activity monitors, accelerometers (ActiTrainer, Actigraph, USA), excluding sleeping time;

<sup>b</sup>Including sleeping time; <sup>c</sup> International Physical Activity Questionnaire, excluding sleeping time;

<sup>d</sup>ST: sedentary time; <sup>e</sup>LPA: light physical activity; <sup>f</sup>MPA: moderate physical activity;

<sup>g</sup>VPA: vigorous physical activity; min=minutes; h=hour

The child in example 2 (Table 4) was physically active in light and moderate intensity at baseline for 1.5 hours according to the ActiTrainer, for 5 hours according to the time-use diaries and for 11 hours according to the IPAQ. After 8 months the child's PA in light and moderate intensity increased with 1.5 hours according to the ActiTrainer, decreased with 3.5 hours according to the time-use diaries and did not change according to the IPAQ.

The child in example 1 met the recommendations for PA by a wide margin of 4.5 hours moderate to vigorous PA at baseline and 3 hours after 8 months measured with the accelerometer. In Example 2 the child did not meet the recommendations, but increased its moderate to vigorous PA from 28 minutes at baseline to 51 minutes after 8 months.

# Discussion

## Methodological considerations

Research with children with disabilities as a vulnerable and heterogeneous group needs careful and thorough considerations. In this thesis the results of Study I and II contributed to the design and development of an intervention to increase participation in PA and reduced sedentary behaviour in this population, which is reported in Papers I-III. In Study III the feasibility and effectiveness of the intervention was studied and the results are reported in Paper IV. The studies in this thesis constitute the development and feasibility/piloting stages and report on each of these components according to the guidance for developing and evaluating complex interventions. The different interacting components involve development, feasibility/piloting, evaluation, reporting and finally the implementation of a complex intervention (MRC, 2000; Craig et al., 2008). Aiming to improve health for children with PD through a more active lifestyle and reducing sedentary behaviour is a complex and lengthy process where each component is important and needs to be thoroughly carried out. The three studies in this thesis consist of the components developing and feasibility/piloting including reporting in Papers I-IV.

The study results will determine whether a feasibility study is sufficient or if a series of studies may be required to refine the design prior to a full-scale evaluation (Craig et al., 2008). Difficulties such as problems of acceptability, compliance and recruitment need to be anticipated in the development and feasibility stages. The acceptability of the different components of the intervention PAR in Study III varied according to the evaluation questionnaire, with high scores for the COPM, GMFM-66 and GAS and lower scores for the heart rate and PA monitors and the IPAQ. It was difficult for the parents to know the PA level for their child with PD, and a feedback from the heart rate and PA monitors close in time to the baseline would probably affect both the estimation of PA for the IPAQ and the motivation for the intervention. The absence of a drop-out rate implies high compliance.

The design of the intervention to include MI as a main component of PAR aimed at encouraging a lifestyle change provided that the child and their parents had a desire

for change (Swedish National Institute of Public Health, 2010). The MI change talk was based on the child's and their parent's readiness for change. Our findings suggest that it was important to be aware of whether the child and their parents were not fully informed and were undecided, whether they had decided to engage in physical activity behaviour but had not yet started to act, or whether they had already acted out their intentions regarding physical activity. These three stages of change have been described in another study as the pre-intention stage, intention stage and action stage (Verschuren et al., 2013). The parents in Study III chose to actively participate in the study and it can be assumed that they were motivated and might have been in the intention or the action stage. Their children, however, were in all different stages, which became obvious when discussing the agreement based on MI, COPM and GAS. COPM proved support the MI change talk by identifying activities that were important for the child, which increased the child's motivation for participating in the physical activities. Several children enjoyed rating the importance, performance and satisfaction of the activities themselves. Engagement and motivation were found to be important for adolescents with CP who were physically active, and participation in activities was a key factor for motivation (Shimmell et al., 2013). These findings together with the findings in Study III show the importance of using MI and COPM as components of the intervention.

Recruiting participants for Study II and III was challenging. Out of the 117 children in Study II which met the inclusion criteria only 16 children with 25 of their parents chose to participate. In Study III 347 children met the inclusion criteria and 23 of them decided to participate; through strategic selection 14 children and 24 of their parents were included in the intervention. Difficulties in recruiting participants in disability research and for randomised controlled trials (RCT) in health research are well known (Nicholson et al., 2013; Porter et al., 2014).

Fundamental mechanisms for a successful recruitment process have been identified: education, facilitation, audit and feedback, time, organisational support and culture (Porter et al., 2014). Participants and clinicians and all others involved should receive sufficient information about the planned intervention which includes possible benefits and risks. In Study II and III all participants and the involved clinicians and administrative personnel received extensive and repeated information. In Study III the children's ordinary physiotherapists received, in agreement with the families, continuously information during the intervention period. All children who met the inclusion criteria in Study III were invited to an Inspiration day where they together with their parents and siblings tried different PA and talked to the researchers. The Inspiration day was facilitating for the recruitment process as six families were recruited there.

Participants and clinicians should be ensured that the time and effort required is minimised and that any disruption of their daily lives because of the intervention would be minimal, while the clinicians' referral processes should be as simple and efficient as possible (Porter et al., 2014). In Study II the children and their parents were reassured that they only had to attend one interview and that the individual interviews with all participants would not take more than one hour. The focus group interview with the children was limited to one hour whereas the focus group interviews with the parents were limited to 1.5-2 hours in agreement with the parents. These time limitations might have contributed to some discussions being missed. The interviewer tried to minimise this risk by summarising what has been said and asking the participants if they wanted to add anything. In the focus groups the observer ensured that all participants had the possibility to say what they wanted to say. In Study III the time and effort required was far more than in the interviews for both children and parents as the intervention covered a time period of 11 months. All meetings with the families were held at times that suited them, and all parties involved were able to decide on the time-frame for each meeting. Family support was offered according to their wishes. This could be through e-mails, text messages or meeting in person, which is in accordance to the findings in another study (Lindqvist et al., 2014b). The children's ordinary physiotherapists were involved through booking the locations for the GMGM-66 at the local habilitation centre, and they could choose to attend these sessions.

The researcher contacted the families in Study II and III within several days of the families voicing their interest to participate in the study. The information letter was sent by an administrative assistant so as not to burden the clinicians at the Child and Youth Habilitation Services. The importance of minimising the period between referral by a clinician and contact by the researcher is considered to be important (Porter et al., 2014). In Study II and III ethical approval was needed before participant recruitment could begin. As neither Study II nor Study III was a RCT the number of participants needed was limited, even though a variation among them was desired. We can confirm that time is a vulnerable recruitment factor, and the results of a review show that time often is underestimated in the planning for recruitment in interventions, particularly RCTs (Porter et al., 2014).

Organisational support in the recruitment process is necessary, where this support includes granting and facilitating access to the study population. Clear access procedures should be agreed at the outset, including sufficient resources for the employees involved in the recruitment process. Organisational support also means that the research has been identified as an organisational priority (Porter et al., 2014). In Study II and III information about the studies were given to the Child and Youth

Habilitation Services. An agreement was reached about the recruitment process in Study II and III, that the involvement of the professionals in this service providing organisation should be minimised, to avoid an increased burden for them. The culture of organisations and professions affects the recruitment process: a culture that values the importance of research and is open to the potential of the planned intervention helps to facilitate the recruitment process (Porter et al., 2014).

Even though recruitment was carefully considered in Study II and III the recruitment of the participants was difficult as there was only a small amount of interested people. In a future RCT all aspects need to be considered once again by taking into account the difficulties which occurred and by modifying the recruitment process. Sufficient time, organisational culture and support in the recruitment process are important aspects to consider.

The researchers chose outcome measures in Study III which were considered to be relevant for the research questions and the different components of PAR showed to be feasible. The IPAQ and the time-use diaries, however, were difficult and time consuming to fill in, as the children and their parents were uncertain how they should estimate the PA intensity of their different activities. The results of the different PA measures illustrate these difficulties as they do not match (Table 3 and Table 4.). Several of the children felt that the heart rate and PA monitors were uncomfortable to wear. As these objective measures of PA are considered to be important outcomes for the intervention and the children had to wear the monitors during two limited periods of one week, enabling factors should be looked for. The heart rate monitor was loose and often slipped down. This might be prevented by the supplemental application of braces or by using a more child-appropriate model. The parents and several of the children found it interesting and motivating to get feedback of the analyses of the objective and estimated PA measurements after 11 months; compliance with wearing the heart rate and PA monitors during the second period might have improved had the families got this feedback prior to the second period. In addition, the objective and estimated PA measurements should be synchronised so that the PA for the same weeks are measured. The time aspect should not only be considered for the recruitment process, but also for the intervention. Individual endurance capacity of the children might imply the need for spreading the assessments out onto more than two sessions. The choice of outcome measures is crucial when designing the evaluation of an intervention. Appropriate measures can be chosen through careful development works and good theoretical understanding (Craig et al., 2008), which we intended to do though the three studies in this thesis.



The parents were asked about the time they spent at the self-selected PA, the activity costs as well as the travel time and costs in Study III. The results of this questionnaire are supposed to contribute to the basis for a health economic analysis of modified PAR in a RCT setting. Assessing cost-effectiveness is important in complex interventions, as this will make the results more useful for decision makers (Craig et al., 2008). A health economic evaluation is aimed at examining the usefulness, effectiveness and availability of a health promotion programme, and to provide information about how to allocate resources (Drummond et al., 2005). The two components costs and consequences of a health promotion programme are included in a health economic analysis.

The studies included in this thesis were considered to be sufficient for continuing with the stages of evaluating and implementation. Some components of the intervention PAR for children with PD are recommended to be modified such as synchronising the objective and estimated PA measurements and giving the families a feedback from the PA measurements in good time prior to the second period. When doing so it might be possible to use a modified PAR in a RCT setting without more piloting.

Both quantitative and qualitative approaches were used in this thesis as the methods may complement each other with different strengths and limitations (Polit & Beck, 2014). For evaluating the quality of the studies in this section, the terms validity and reliability are addressed for the quantitative instruments and the terms trustworthiness, credibility, dependability, transferability and confirmability for the qualitative approach (Lincoln & Guba, 1985; Polit & Beck, 2014).

## **Validity and reliability**

Validity is the degree to which an instrument measures what it is intended to measure. Reliability refers to the consistency with which an instrument measures an attribute. An instrument is reliable if its measurements accurately reflect the measures of the attribute. Validity and reliability of an instrument are not totally independent, as an instrument only can be valid if it is reliable. However, an instrument can be reliable without being valid (Polit & Beck, 2014).

*Internal validity* refers to the extent to which the outcome is explained by the independent variable and the extent to which alternative factors affected the outcome (Polit & Beck, 2014). In Study I three contexts of PA were described as leisure time, PE at school and physiotherapeutic interventions. The CPUP registry data could not be used to investigate whether the recommendations of 60 minutes of PA daily

(WHO, 2014) were achieved or not as only the frequency of participation in PA and not duration and intensity were investigated. POR were used for analysing data in this study, and comparisons was made within the same subgroups, such as comparing boys to girls that have the same GMFCS-E&R level, age, MACS level, stature, and cognitive level, either with or without an epilepsy diagnosis. A limited number of characteristics were selected for examination in the POR analyses in order to enable statistical precision. GMFCS-E&R levels and not CP-subtypes were chosen because GMFCS classification is reliable, valid and adopted worldwide (Westbom et al., 2007). In addition to GMFCS-E&R levels MACS levels, age groups, gender, obesity/thinness, epilepsy and cognitive function were selected, while neither visual nor hearing impairment was included. The selected independent variables were values to be of importance for variations in PA according to literature review and clinical experience.

In Study III COPM, GAS, a personalised logbook, GMFM-66, a PA questionnaire with questions used in the CPUP, IPAQ, heart rate and PA monitors and time-use diaries were used to assess the outcomes. The instruments were chosen due to their validity and reliability; however, they have not yet been used in the context of PAR for children with PD and not in this combination.

*External validity* refers to the extent to which the results can be generalised (Polit & Beck, 2014). In Study I a total population was included, implying high external validity. The validity of the CPUP database has been found to be good, providing for both the availability of a second opinion about the classification of CP and CP subtypes and an active search for undiagnosed CP among all children with impairments (Westbom et al., 2007). In Study III only 14 of 347 children participated, which might be a hindrance for external validity while limiting the generalisation of the results. Even though variation in the participants was great as shown in Table 1, this limitation implies the need for a full-scale evaluation of the intervention PAR as a RCT. Replication of a study is relevant to external validity (Polit & Beck, 2014). Using data from the National Quality Registry CPUP in Study I provides good potential for replication as a total population is represented and the analysis was described stepwise. In Study III the procedure of the intervention was carefully described, which might imply a possibility for replication. However, this was a feasibility study and, to our knowledge, the first study about PAR for children with PD including MI and the assessments we chose. This limits the replicability and indicates the need for further research.

*Reliability* might be threatened by the procedure used in Study III as several assessments were performed in the children's homes. On the other hand, being at

their home made the children and their parents more comfortable and might have encouraged their motivation and engagement for participating in the study. The GMFM-66 was performed at the child's local habilitation centre to ensure a more standardised environment. In addition, the GMFM-66 was filmed and scored independently by two physiotherapists. Efforts were made to reduce the threats to reliability by educating all three physiotherapists in the methods of MI. All instruments used in Study III were tested in advance on other people.

## **Trustworthiness**

In Study II a qualitative approach was applied to illuminate the children's and parents' experiences of a certain phenomenon, which was the child's participation in PA; content analysis was used for the analyses (Graneheim & Lundman, 2004). Both individual interviews and focus group interviews according to the participants' wishes were conducted. Focus groups were used to yield qualitative data in a focused discussion aimed at trying to understand differences in perspectives (Krueger, 2009). A response from one participant could trigger responses from other participants. The interaction in the focus group with children became livelier when using pictures and play figures.

Interviewing young children is difficult as there is the risk that they might respond how they feel the interviewer wants rather than how they feel, and they can have a short attention span. Pictures and small play figures were used to stimulate the children's discussions and they seemed to enjoy these narrations. The individual interviews took place in a comfortable and not-threatening environment with the possibility to have a proxy with them according to each child's preference. During the interviews the interviewer checked her understanding by summarising discussions and adding follow-up questions. At the focus group there was both a moderator and an observer; the observer helped to ensure that the moderator understood what the children (Paper II) or their parents (Paper III) were saying and ensured that everybody had the possibility to say what they wanted to talk about.

It is important to demonstrate rigour when describing the analysis and findings of a qualitative study to achieve trustworthiness; the aspects of trustworthiness are described as credibility, dependability, transferability and confirmability (Polit & Beck, 2014).

## *Credibility*

Credibility refers to confidence of truth of data and how well the processes of analysis are focused on the research question (Graneheim & Lundman, 2004; Polit & Beck, 2014). Having a variety in the selection of participants increases the credibility. The procedure of data collection and the analysis process is described carefully and stepwise in Paper II and III. The recruitment process in Study II was challenging and it is likely that the parents who responded were especially interested in PA, which might have affected the results of the interviews. In Paper II there was a great variation among the children and included boys and girls from both rural and urban areas; the children lived in various family constellations and had various limitations in gross motor and cognitive function. Some children communicated non-verbally or were not native Swedish speakers; interpreters including sign language interpreters and language interpreters were used for several interviews. It is challenging to involve children with disabilities in research, and those with non-verbal communication are rarely involved (Bailey et al., 2014). In the Studies II and III we tried to support the involvement of all children, including of those with non-verbal communication. In Paper III the variation among the children's parents was also great according to gender, living area, socioeconomic and ethnical backgrounds; 11 of 25 parents were not native Swedish speakers. Several participants in Study II, described in Paper II and III, chose to use a language interpreter. The use of an interpreter may have caused the language to be interpreted in a variety of ways or create possible misunderstandings. Additionally, the interpreter might not understand the child, parent or interviewer correctly or might interpret the language more than translate verbatim. The interviewer carefully explained the study to the interpreter and about the need of verbatim translation; in addition, the interviewer cross-checked the answers by asking the same questions with different formulations. However, that was the only possibility to interview these participants and the authors later discussed these difficulties. Including a proxy during the interview (Paper II) introduced the risk that the adult wanted to talk instead of the child. The interviewer clarified that the interview was meant to illuminate the child's perception, and that the proxy did not need to comment on or correct anything the child said. The parent's experiences were also highlighted and reported on in Paper III. The way of recruiting participants might have resulted in a large number of motivated parents who wanted to participate, whereas the interest and motivation of child participants might have been more varied. The variation among all participants, both among the children and their parents, contributed to the interesting and comprehensive results, especially as including such a great variation of people's ethnic origin is rarely seen in qualitative studies.

The sub-categories and categories (Paper II and III) covered the data well; all meaning units fit into the final analysis. There was on-going dialog among all co-authors which resulted in an agreement about the way the data was labelled and sorted. All authors have experiences of working with children with PD, which obviously affected their pre-understanding. During the whole process, from planning to interviewing and including all steps of the analysis, all authors considered how their pre-understanding may affect the analysis, abstraction and interpretation.

### *Dependability*

Dependability refers to the change of data over time and the researcher's decisions during the analysis process (Polit & Beck, 2014). In Paper II and III the same opening question was used for all children respectively their parents and the semi-structured interview guide ensured that all areas were covered, which increased dependability. There were discussions within the research group during the whole process to verify respective positions. All three co-authors read all interviews and conducted the steps for analysing the material together. In addition, a PhD-student from the research group participated in some of the discussions.

### *Transferability*

Transferability refers to whether the findings can be transferred to other settings or groups (Lincoln & Guba, 1985). Graneheim & Lundman (2004) claim that the authors may give suggestions but it is up to the reader to judge the transferability. By being as distinct and clear as possible when describing data collection, participants in their various contexts, the process of analysis and the findings of Study II, the researchers tried to facilitate the transferability. The variation in the participants was impressive even though there were only 16 children and 25 parents.

### *Confirmability*

Confirmability refers to the objectivity of the data and the interpretations (Polit & Beck, 2014). The co-authors' different backgrounds and pre-understandings might reduce the risk for subjective analyses of the interviews. To confirm the results quotations from different children (Paper II) and their parents (Paper III) were used when describing the findings, which verified that the results were taken from the interviews.

## General discussion of results

This section will discuss the main results within the frame of the ICF-CY and through highlighting the 'F-words' *fitness, function, friends, family, fun* and *future* (Rosenbaum & Gorter, 2012). These main findings involve context-based participation in PA of children with PD including the exploration of facilitators and barriers. Another important contribution was a feasibility and effectiveness evaluation for the intervention PAR when used with children with PD.

### Context-based participation

Children with PD participate in PA in different contexts, and in this thesis these contexts, which include home and community settings for leisure time activities, school and the child and youth habilitation centre, have been explored and described. In addition, a context-focused approach has been used in Study III through identifying the stage of change in which each child and their parents were, through identifying and changing the primary constraints in the environment if needed, and through providing opportunities to engage in their self-selected PA and promote functional performance (Law et al., 2007). The focus of context therapy is on adapting the environment or the task rather than on the child's impairment (Darrah et al., 2011). PAR is an individualised intervention and each child, their parents and the physiotherapist are in collaboration when developing the written agreement about the self-selected PA in Study III. Environmental adaptations were considered and performed whenever necessary.

The amount of support the child needed from the physiotherapist for their self-selected PA was considered by the child, their parents and the physiotherapist and was adapted to the individual needs of the child during that period. This type of individual approach seemed to be successful and could coincide with Law's et al. (2011) suggestion that the frequency of intervention is a critical component of a successful intervention, while dose-response relations which relate to the amounts of treatment and changes in functional abilities still need to be identified. These dose-response relations might have been identified through the individualised context-based approach in Study III including ongoing evaluations and adjustments of the self-selected PA by using MI and GAS.

The family represents the essential environment for children (Rosenbaum & Gorter, 2012). This was carefully considered in the PAR intervention by collaborating with the child and their parents and by engaging with them as equal partners in the

intervention (Bailey et al., 2014). Family-centred services (FCS) as best practice is based on the understanding that parents know what is best for their child (Rosenbaum et al., 1998). In Study III this view is extended by trusting in each child's ability to have their own opinion. In the FCS, however, each family should be seen as unique with their own strengths and the needs of every family member should be focus in a client-centred approach (King et al., 2004). Socioeconomic status has an influence on habitual PA and sedentary behaviour in children and should be considered in any approach (Drenowatz et al., 2010). PAR has both a context-focused and an individualised approach. The two approaches address the 'F-words' of *family* and *function* (Rosenbaum & Gorter, 2011), and both seem to be necessary for PAR.

Mutual respect and trust, open communication and shared decision-making are some of the components of family-professional collaboration (An & Palisano, 2013), which were used for PAR in Study III. The written agreement was developed in collaboration with the child, their parents and the physiotherapist. King et al. (2014) described that building a collaborative relationship between the families and the therapist for effective communication and family engagement is important. However, increasing participation levels in PA among children with PD, behaviour changes are needed. While the transtheoretical model (TTM) is a motivational theory for behaviour change which consists of different stages (Kosma et al., 2002), the constructs of social cognitive theory (SCT) are goals guiding specific behaviour, outcome expectancies, perceived self-efficacy, facilitators and barriers (Lui & Hui, 2009). These different approaches might complement each other whilst they focus on specific aspects, and in Study III both approaches were applied. Through MI the different stages of change for both children and parents were identified. MI and COPM were used for goal setting and to discuss specific behaviour, outcome expectancies, perceived self-efficacy, facilitators and barriers for each child included in the study.

In Study I we described the participation in PA of children and adolescents with CP aged 7-17 years in their contexts leisure time and PE at school as well as their physiotherapeutic interventions. The variation in the types of leisure time PA increased with increasing gross motor function. Only half of the children and adolescents with the most severe motor limitations participated in PE at school while those with intellectual disabilities received the least physiotherapeutic interventions. Children and adolescents with more severe gross motor function limitation received the most physiotherapeutic interventions. These results show that the contexts of PA are important when planning interventions. In Study II the findings from the interviews confirm the importance of having different contexts for children to participate in PA. School was considered the most challenging context according to

the interviews, where there was a risk for the children to be singled out and where the parents felt they lacked influence. Parents wanted competent personnel and group leaders for their child's PA, both at leisure time and at school. Parents also wanted support for their children to make friends.

## Facilitators and barriers

The results of Study I showed that children and adolescents with more severe motor limitations, with intellectual disability and with thinness seem to have the greatest needs for interventions to help increase their participation in PA and decrease their sedentary behaviour. As these results mostly address the children's body structure and body function, the 'F-word' *fitness* might be appropriate. A child with thinness, implying a BMI of less than minus two SD, might not be in a condition to exercise and rather be tired and lack energy. In these children the treatment of thinness should be prioritised before increasing their PA. It may be expected that children and adolescents with more severe motor limitations have the greatest needs for increased PA and reduced sedentary behaviour, which have been shown in other studies (Jahnsen et al., 2003; Kaewutan et al., 2014; Parkes et al., 2010).

In Study II the children stated that they were physically active because they enjoyed the sensation of speed, felt capable or had someone with whom to be active with, which addresses the 'F-words' *fun*, *function* and *friends*. They wanted to be asked about their needs and get the opportunity to try self-selected activities and receive individualised support (*fun*, *family*, *function*, and *friends*). This is similar to other studies with adolescents without disability, which highlighted the importance of empowering and engaging adolescents when promoting health and PA (Lindqvist et al., 2014a; Lindqvist et al., 2014b).

Environmental factors (*family*) could be either facilitators or barriers, while pain, fatigue or feelings of inadequacy were barriers (*function* and *fitness*). In another study, recurrent musculoskeletal pain and mental health problems were found to contribute to reduced participation for children with CP (Ramstad et al., 2012). Focus on pain and fatigue management is needed as these factors are associated to deteriorated walking function in adults with CP (Opheim et al., 2009; Riquelme & Montoya, 2011). In a recent study the importance of both self-reported and proxy-reported mental health is pronounced as the child's perspective provides different information from the parents' perspective (Ramstad et al., 2014). In Study II both the children and their parents were interviewed which contributed to having multiple viewpoints. Perceived barriers seem to differ for children, parents and health professionals



according to the findings of study from the Netherlands (Jaarsma et al., 2014). While the children in Study II were positive towards participation in PA, several challenges were experienced by the parents when their child wanted to be physically active: the parents wanted to protect their child while they also wanted to push them to be more independent, which addresses the 'F-word' *future*. The parents want competent persons to be available to support their child in participation in PA and help them to find friends to be physically active with (*function* and *friends*).

Providing effective strategies and resources for increasing PA among adolescents with disabilities in community-based sports and recreation programmes are described as important responsibilities for paediatric rehabilitation and healthcare professionals (Rimmer & Rowland, 2008; Shields et al., 2012). Differences in participation patterns for children with disabilities have been found between Scandinavian countries and the Netherlands, where Scandinavian children participated more frequent in leisure activities than Dutch children (Ullenhag et al., 2012). To explain these findings, the different education systems were discussed, with 4 % of children with special education needs attending special schools in Norway and Sweden compared to 62 % in the Netherlands. Children with obesity in the CP population (Study I) participated more often in PE at school, implying that efforts were made in school to meet their needs of PA.

Family culture and attitudes affect children's motivation for being physically active and should be taken into account (*family*). Different cultural backgrounds might reflect on how parents perceive of their child's needs and whether they push them to participate in PA or whether they are more protective. The findings of another study with parents of obese children show that limited knowledge of PA guidelines, low priority for PA, girls' dislike for PE, and the cost of PA might be barriers to healthy lifestyles within and between ethnic groups (Rawlins et al., 2013). However, all parents in Study II and III were interested in PA, which might be due to their voluntary participation in a study about PA. Assumptions of homogeneity or labelling within groups should be avoided, even though attention should be paid to individual family culture and attitudes (Callanan & Waxman, 2013). This critical approach is also discussed in another study, which described the predominance of cultural difference theory in science education and its uncritical use without attention to its limitations, such as the disregard of possible intra-group differences (Carlone & Johnson, 2012). In Study II and III limited Swedish language skills were found to reduce access to information and also the parents' ability to support their child with participating in PA. This was also shown in a study with children from different ethnic backgrounds with severe disabilities (Pickl, 2011). An enhanced awareness of

language limitations might lead to better use of language interpreters when discussing, planning and performing an intervention with non-native speaking families.

## Physical Activity Referrals

PAR seems to be feasible and effective for children with physical disabilities, where the referral consists of a written agreement between each child, their parents and a physiotherapist combined with MI and individualised support. MI as an independent intervention has not shown to increase PA (Hollis et al., 2013); however, when integrated in other treatments MI was useful for increased PA in adults with chronic health conditions (O'Halloran et al., 2014). In Study III MI was used as one component of the intervention.

The engagement of the children and parents, their motivation and the child's social participation are important for lifestyle changes. This addresses the 'F-words' *fun*, *family* and *friends* and is in accordance to the findings of Majnemer et al. (2008). As the intervention involves both everyday physical activities and organised physical activities according to individual preferences, opportunities and facilities, PAR might be a possible way of promoting an active lifestyle. Time and availability of self-selected activities, competent leaders, and other children to be friends with as well as overall costs for the activity are important factors towards participation and cover the 'F-words' *function*, *friends* and *future*. In addition, availability of assistive devices for leisure time and help with adjusting those assistive devices are required for children with more severe disabilities (*function* and *fitness*). PAR was occasionally time-consuming for the families and costs varied among the families, depending on their child's self-selected PA.

The assessments at baseline, the ongoing evaluation with feedback and adjustments of the self-selected PA, and the assessments, feedback and evaluations after 8 and 11 months were accepted and motivating factors for the children's participation in PA. All children increased frequency and duration of their participation in PA from baseline according to the logbooks, COPM and GAS. Additionally, several children improved their gross motor function according to the GMFM-66.

The estimated and objectively measured time spent in the different PA levels including sedentary time, light, moderate and vigorous intensity increased for many children but differed among the time-use diaries, the IPAQ and the accelerometer. The parents found it difficult to know when their child engaged in these different PA levels and it was interesting for the children and their parents to get feedback about their objectively measured PA. As they only got this feedback after 11 months, it had

neither an impact for their estimated PA at 8 months nor could it increase their motivation during the intervention. Receiving feedback closer to the baseline would probably affect both the estimation of PA and the motivation for the intervention. The results from the accelerometer showed that the average for moderate-vigorous PA was 1.5 hours every day, with a variation from less than half an hour to more than 4 hours for the individual child. The children were sedentary most of the day with about 3 hours of light PA.

PAR consisting of the components used in Study III was effective for increasing or maintaining an active lifestyle for the participating children during the intervention period.

# Conclusion and clinical implications

Two components according to the guidance for developing and evaluation of complex interventions were carried out in this thesis with the aim for children with PD to achieve a more active lifestyle and reduce their sedentary behaviour. The development of the intervention PAR is reported in Paper I-III, the feasibility/piloting in Paper IV.

The children with the greatest needs for a more active lifestyle have been identified in Study I as those with more severe motor limitations, with intellectual disability and with thinness. The focus for children with PD who meet the recommendations of 60 minutes moderate-vigorous PA daily, as shown in Study III, should be on maintaining this active lifestyle by participating in PA that they enjoy and want to participate in. Motivation, engagement and social participation are crucial factors for children with PD's participation in everyday and organised habitual PA. It is essential to conduct PAR for children with PD in collaboration with the children, their parents and a physiotherapist. Each child's preferences, wishes and other thoughts on participation in PA are important and guiding for PAR. Children with PD want to try self-selected activities, and individual support for each child through using MI and considering their different ethnical, cultural and socioeconomic family backgrounds seems to help PAR be successful. The timeframes and availability of self-selected activities, costs, having competent personnel and the opportunity to make friends are important factors towards higher participation rates. In addition, the availability of assistive devices for leisure time activities and help in adjusting those assistive devices are required for children with more severe disabilities.

To our knowledge, this is the first study about PAR with children with PD where objective and estimated PA is measured. To improve the practicality and likelihood of implementation, splitting the assessment meetings with the families into shorter meetings and seeing that PA measures are performed during the corresponding time periods are recommended modifications. The results suggest that PAR is feasible and effective for children with PD. The developed intervention PAR might be implemented and further evaluated in the regular Child and Youth Habilitation Services.

# Future Perspectives

Further research of PAR for children with PD is needed in order to carry out the missing components according to the guidance for developing and evaluating complex interventions. The missing components include evaluating the intervention and implementation. Modifications developed from the feasibility study should be implemented and the developed PAR for children with PD needs to be evaluated, preferably in a randomised controlled trial in a multi-centre setting.

The potentials and limitations of the developed PAR for children with PD according to its impact on participation in PA, on sedentary behaviour and on social participation need to be further evaluated. How can an adequate support of the children with PD and their parents to become or stay physically active and less sedentary be incorporated in the children's daily life such as at school? As participation in PA at school has been found challenging, replacing sedentary time with light intensity PA such as standing with or without standing devices are important to evaluate. The accessibility of PA in the community and assistive devices for PA with individual adjustments also needs more attention in future research.

Other future perspectives are how to meet the children's and parents' wishes for support through competent personnel for the child's participation in PA and in getting friends. How can standardised and participation based outcome measures that provide both feedback and feedforward information as those used in Study III such as COPM, GAS and GMFM-66 be integrated into the standard services in order to increase the competence among the physiotherapists with respect to their time and routines? As MI has shown to be an important component of PAR, further research is needed to evaluate whether MI or other interview-techniques are best suited for promoting an active life-style. Physiotherapists should be educated in MI or another interview-technique.

The potentials and limitations of the developed PAR for children with PD according to its impact on PA intensity and sedentary time with the use of objective and estimated measurements need further evaluation. As maintaining an active life-style during the adolescence is a challenge, the developed PAR intervention needs to

continue and be evaluated during many years. In addition, health economic analysis should be made to show the costs and costs savings for the individual and society.

Working with the studies included in this thesis has raised new questions. What information do the data from CPUP comprise from other countries with different cultures, social conditions and health care organisations? How can the impact of gender and environmental factors according to participation in PA be addressed? How can interdisciplinary research such as designing and developing joyful mobile computer games contribute to strengthen self-efficacy, increase social participation and enhance friendship among all children with disabilities in spite of age, gender, and the severity of gross motor, manual, communicative and cognitive function?

# Summary in Swedish

Fysisk aktivitet är en förutsättning för god fysisk och psykisk hälsoutveckling. Barn med rörelsenedsättning som cerebral pares (CP) är mindre fysiskt aktiva och samtidigt mer stillasittande än barn utan rörelsenedsättning och har svårt att uppnå WHO:s rekommendationer för fysisk aktivitet. För barn och unga mellan 5 och 17 år rekommenderas minst 60 minuter fysisk aktivitet varje dag samt en minskning av stillasittande tid. Aktiviteterna bör inkludera både måttlig och hög intensitet och vara så allsidiga som möjligt för att ge kondition, muskelstyrka, rörlighet, snabbhet, koordination samt kortare reaktionstid. Det saknas kunskap om vilket stöd som barn med rörelsenedsättning och deras föräldrar behöver för ökad delaktighet i fysiska aktiviteter samt vilka behov och önskemål de har och vilka möjligheter och hinder de upplever i samband med fysisk aktivitet. En metod för att främja fysisk aktivitet är Fysisk aktivitet på Recept (FaR), som har visat lovande resultat för en aktivare livsstil för vuxna. Det saknas dock forskningsbaserad kunskap om hur barn med rörelsenedsättningar kan bli mer regelbundet fysiskt aktiva, mindre stillasittande och huruvida FaR för en aktivare livsstil är tillämpligt för dessa barn.

Det övergripande syftet med avhandlingsarbetet var att utforska faktorer och processer som påverkar tillgänglighet och delaktighet i fysisk aktivitet och stillasittande hos barn med rörelsenedsättning, där CP är den vanligaste diagnosen. Ett ytterligare syfte var att utvärdera huruvida FaR är tillämpligt för barn med rörelsenedsättning och vilken effekt det har på barnens stillasittande och deltagande i fysiska aktiviteter. Tre studier som resulterat i fyra artiklar ingår i avhandlingsarbetet.

I första studien (artikel I) beskrevs frekvensen av deltagandet i fysiska aktiviteter på fritid och skolidrott samt fysioterapeutiska insatser för en totalpopulation bestående av 364 barn och unga med CP i åldern 7-17 år. Även faktorer som påverkar deltagandet i fysiska aktiviteter identifierades. Uppgifterna hämtades från det nationella kvalitetsregistret och uppföljningsprogrammet för CP (CPUP). Resultaten visade att barn med de största grovmotoriska svårigheterna deltog i minst omfattning i skolidrott, medan måttliga grovmotoriska svårigheter ledde till låg delaktighet i fysiska fritidsaktiviteter. Barn i åldern 7-11 år samt barn med fetma deltog i större utsträckning i skolidrott, medan undervikt (för låg vikt i förhållande till

kroppslängden) minskade delaktighet i fysiska fritidsaktiviteter. Frekvensen av fysioterapeutiska insatser ökade ju större barnens grovmotoriska svårigheter var, däremot erhöll barn med intellektuella funktionsnedsättningar färre fysioterapeutiska insatser. Kön och epilepsi visade inga samband med deltagande i fysiska aktiviteter. Den andra studien bestod av kvalitativa intervjuer med 16 barn i åldern 8-11 år med CP och med 25 av deras föräldrar (artikel II och III). I den tredje studien (artikel IV) deltog 14 barn i åldern 7-12 år med rörelsenedsättningar och 24 av deras föräldrar. Barnen som deltog i Studie II och III hade olika diagnoser och svårighetsgrad av motorisk, kognitiv och kommunikativ funktion samt olika etniska, kulturella och socioekonomiska bakgrunder.

Intervjuerna analyserades med kvalitativ innehållsanalys och deltagarna fick välja att delta i individuella eller fokusgruppsintervjuer. Resultaten av intervjuerna med barnen visade att de var fysiskt aktiva för att de tyckte om känslan av fart, att de kände att de klarade av att göra aktiviteterna och/eller hade någon att göra aktiviteten tillsammans med. Omgivningsfaktorer kunde vara både främjande och hämmande, där smärta, trötthet eller att inte vara tillräckligt bra var hindrande faktorer. Resultaten av föräldrantervjuerna visade vilka utmaningar föräldrar upplevde att de stod inför när deras barn ville vara fysiskt aktiva. De ville å ena sidan skydda sina barn från alla svårigheter men kände å andra sidan behov av att pusha barnen mot självständighet. Varje familj hade olika inställning, attityder och motivation till att delta i fysisk aktivitet och minska stillasittandet. Faciliterande faktorer för föräldrarna var kompetent personal och stöd för barnen att få kompisar att vara fysiskt aktiva med.

I interventionen ingick en skriftlig överenskommelse (FaR) baserat på motiverande samtal (MI), Canadian Occupational Performance Measure (COPM) och Goal Attainment Scaling (GAS). Utöver detta ingick grovmotoriska bedömningar enligt Gross Motor Function Measure (GMFM-66) och skattning av fysisk aktivitet (International Physical Activity Questionnaire, IPAQ). En tidsdagbok fylldes i under två dagar i samma vecka som barnen använde en accelerometer för att objektivt mäta fysisk aktivitet. Bedömningarna gjordes vid interventionens start, efter 8 och 11 månader. Alla barn och föräldrar fullföljde interventionen. Kunskap om tillgänglighet av fysiska aktiviteter och lokaler ökade, barnen fick möjligheten att själva välja aktiviteterna de deltog i och de fick det stöd de önskade. Barnen som deltog i gruppaktiviteter fick nya vänner, alla barn uppfyllde delvis eller helt sina mål och 6 barn förbättrade sina grovmotoriska förmågor. I genomsnitt var barnen fysiskt aktiva på måttlig till hög nivå i 90 minuter dagligen med en spridning från mindre än 30 minuter till mer än 3 timmar dagligen, mätt med accelerometer. FaR är tillämpligt för barn med rörelsenedsättningar och kan bidra till en aktivare livsstil.



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