SILJA PIRILÄ

Children with Functional Motor Limitations

A Three-level Approach

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ACADEMIC DISSERTATION

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ABSTRACT

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The aim of the study was to investigate children with functional motor limitations. An effort was made to integrate the studies into a current structure of International Classification of Functioning, Disability and Health (ICF; WHO, 2001). The studies examined linkages between three levels: (1) the level of most immediate environment of the child: i.e. family strengths, social-economical status, assistance of the child in daily activities; (2) the level of activity limitations: i.e. functional gross and fine motor skills in addition to skills in self-care, mobility and social function; and (3) the level of body functions and structure: i.e. neonatal brain lesion, birth weight, intellectual functions, neuropsychological functions, language and motor speech skills.

Sixty-nine children receiving special health care services in the Department of Pediatric Neurology, Tampere University Hospital, for identified developmental disabilities, such as cerebral palsy (CP), psychomotor or motor delay, chromosomal anomalies, etc., participated in the current study. Most of the children (n=61) and their caretakers were recruited from the Play Project which was an early intervention program for disabled children run in the years 1994 to 1997 (Korpela & Nieminen, 2001). For methodological reasons the sample was enlarged with 8 children. The children were as infants treated in the neonatal intensive care unit and followed up later in the Department of Pediatric Neurology in Tampere. Due to prenatal or perinatal brain insults most of them developed functional motor limitations over time. The extent of disability ranged from mild impairment to severe multiple disabilities.

The results showed that the family strengths were rather high. However, the combination of the severity of the functional motor limitations and cognitive difficulties of a child showed some effects on family functioning. The children showed functional limitations in the domains of self-care, mobility and social function. In the domains of self-care and mobility, the functional limitations triggered the needed assistance by the caregivers. However, in the social function domain the assistance the caregivers offered did not differ from the norm especially when the children had cognitive level in the normal range. The severity of the CP and cognitive level were associated with motor speech and language problems. CP with comorbid cognitive difficulties (IQ < 70) produced language deficits in the expressive and comprehensive domains. Half of the children with the cognitive level at or close to norms showed deficits in their motor speech skills. In children with diplegia spastica, neuropsychological deficits were found within sensorimotor functions, visuospatial processing, attention/executive functions, and speeded naming whereas language and memory/learning functions were at or close to norms. The neonatal brain ultrasound methodology predicted moderately the severity of CP, not the intelligence, neuropsychological functions, language, or motor speech skills.

Children with functional motor limitations, family strengths, functionality in self-care, mobility, and social functions, intellectual and neuropsychological skills, periventricular leukomalacia

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To my parents

LIST OF PUBLICATIONS

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II Pirila, S., van der Meere, J.J., Seppanen, R-L., Korpela, R., & Nieminen, P. (in press). A pilot study on children with limitations in self-care, mobility and social functions: effects on family strengths. *Families in Society. The Journal of Contemporary Social Services*.

III Pirila, S., van der Meere, J.J., Pentikainen, T., Ruusu-Niemi, P., Korpela, R., Kilpinen, J., & Nieminen, P. Language and motor speech skills in children with cerebral palsy. Manuscript submitted for publication.

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LIST OF ABBREVIATIONS

AE	Age Equivalent (language, motor speech domain)
AGE	Age of primary caretaker
ARS	Autti-Rämö's Scale: amount of assistance needed in gross motor function
CP	Cerebral Palsy
DQ	Developmental Quotient
FFSS	Family Functioning Style Scale (family strengths)
ICF	International Classification of Functioning, Disability and Health
ICIDH	International Classification of Impairment, Disabilities and Handicap
IVH	Intraventricular hemorrhage
IQ	Intelligence Quotient
MRI	Magnetic Resonance Imaging
NEPSY	A Developmental Neuropsychological Assessment
PEDI	Pediatric Evaluation of Disability Inventory (functional limitations of a child, amount of caregiver assistance)
PVL	Periventricular Leukomalacia
RDLS	The Reynell Developmental Language Scale
SES	Social-Economical Status
SLI	Specific Language Impairment
US	UltraSound scanning (neonatal, cranial)
WHO	World Health Organization
WPPSI-R	The Wechsler Preschool and Primary Scales of Intelligence – Revised
WISC-R	The Wechsler Intelligence Scale for Children – Revised
WISC-III	The Wechsler Intelligence Scale for Children – Third Edition

CONTENTS

1. INTRODUCTION

Motor impairments in childhood are common neurological problems faced by clinicians in the pediatric and neurological clinics. Impairments vary from motor clumsiness (developmental coordination disorder) to cerebral palsy (Ahonen, 1990; Autti-Rämö, 2004; Herrgård, 1993). In Finland, about 6 percent of the children (in the age range of 4 to 15 years) have neurological long-lasting disorder. In absolute numbers, it means that about 100 000 - 200 000 children need neurological follow-up and care (Sillanpää, 2004), about 2.5 percent of them have severe functional motor impairment conditions, among others, cerebral palsy (Aicardi & Bax, 1992; Autti-Rämö, 2004; Bottos, Granato, Allibrio, Cioachin, & Puato, 1999; Fernandez-Alvarez & Aicardi, 2001; Nordmark, Hagglund & Lagergren, 2001; Sillanpää, 2004).

Motor impairments, like any other disability, can be operationalized in terms of functional limitations (e.g., problems in mobility, communication or self-care), or in terms of impairments (e.g., impairment of brain structure or cognitive abilities related to mobility) (Simeonsson, 2003)**.** In the current World Health Organization (WHO, 2001) envisaged usage, the term 'impairment' means a loss or abnormality in body structure or function. An individual who has a motor impairment may have difficulties in executing activities compared to people without a similar health condition. These difficulties, i.e. activity limitations, can cover a full range of life areas from basic learning or watching to composite areas such as interpersonal interactions and social tasks. A person may also experience problems in involvement in life situations, labelled as participation restrictions. They take into account the actual context, all aspects of the physical, social and attitudinal world, in which people are living. Impairments, activity limitations and participation restrictions belong as key concepts into the International Classification of Functioning, Disability and Health (ICF; WHO, 2001), a recent model of functioning and disability formulated by the World Health Organization.

The ICF model is a revised version of the International Classification of Impairment, Disabilities and Handicap (ICIDH; World Health Organization, 1980). ICF has moved away from a 'consequences of disease' classification to a 'components of health' classification. The 'components of health' classification has two parts, each with two components. Part 1: Functioning (i.e. body functions, body structures and impairment) and Disability (i.e. activities, participation, activity limitations, and participation restrictions). Part 2: Contextual factors (i.e. environmental factors, such as facilitators or barriers; and personal factors). The model presents more fully the impact of disability as both activity limitations at the level of an individual and participation restrictions at the level of society.

It also incorporates social factors in the model as they may influence the impact of a person's impairment or activity limitation (Figure 1).

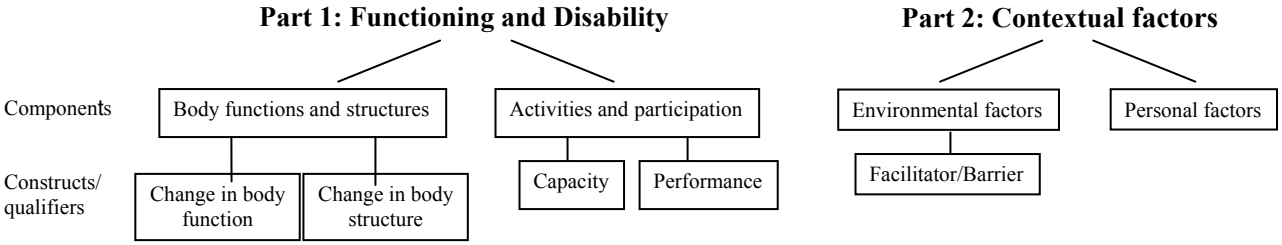

Figure 1: The structure of ICF (WHO, 2001).

The WHO framework tries to depict the complexity of interrelations between functioning and disability and to achieve a synthesis, in order to provide a coherent view of different perspectives of health from a biological, individual and social perspective. It has assisted clinicians and researchers in broadening their understanding of the effects of neurological health conditions on children's function beyond the organ-system level (Simeonsson, 2003). It will be used in this thesis as a guideline in our effort to investigate the multiple interrelations between different levels of functioning in children with disabilities and their families. This thesis tries to examine linkages between three levels: first, the level of most immediate environment, the family child is living in, second, the level of individual activity limitations, and, third, the level of body functions and structure.

2. REVIEW OF THE LITERATURE

The review of the literature proceeds from the more general findings related to the psychosocial adjustment of the families rearing a child with activity limitations to more specific findings concerning motor, cognitive and body function impairments related to cerebral palsy. The overview tends not to be diagnosis-specific and tries to utilize a non-categorical approach underlining the commonalities in psychosocial ramifications in the families rearing a child with motor limitations (Wallander & Varni, 1998).

2.1. Environmental level: parental adjustment, social-economical level and parental age (Studies I, II, III)

Before discussing in depth the concepts used in the Studies I, II and III, i.e., parental adjustment, social-economical status and parental age, it is needed to be emphasize that disability is characterized as an outcome or result of a complex relationship between an individual health condition and personal factors, and of the external factors that represent the circumstances in which the individual lives. Different environments may have a different impact on the same individual with a given physical disorder. An environment with barriers, or without facilitators, will restrict the individual's performance; other environments that are more facilitating may increase that performance (WHO, 2001). However, surprisingly little is known about the multiple interrelations between functioning and disability of children and their environments during the developmental years (Korpela, 1993; Ostensio, Brogen Carlberg, & Vollestad, 2003; Pennington & McConachie, 2001; Simeonsson, 2003).

In the current ICF model (WHO, 2001), environmental factors focus on two different levels, namely individual and societal. Individual level concerns the immediate environment of an individual, including home and school settings. Societal level concerns the formal and informal social structures, services and overarching approaches or systems in the community or society that have an impact on individuals. The environmental factors, investigated in the current thesis, focused on the home environment of the children with activity limitations and, more specifically, on family strengths and amount of caregiver assistance. Other aspects investigated were social-economical status and age of parents.

2.1.1. Parental adjustment

Parents of children with a chronic disorder may function much like others who experience stressful circumstances, some of whom develop maladjustment and others who show resilience in the face of their stress. Given the heterogeneity in chronic disabilities in children, there has been an interest in testing whether differences in disability-related problems matter for parental adjustment. Critical disability-related stress could be identified in the areas of medical issues, nature of onset and course of a disability, expected fatality, and degree of functional limitations (Leskinen & Juvonen, 1994; Wallander & Varni, 1998). Diagnosis, severity within an illness, or whether the disorder is visible or is associated with bowel and bladder, cognitive, or communication impairment, are examples of disease parameters that have been examined. Studies differ with respect to the type of populations they examined (e.g., cerebral palsy, multiple disabilities, pediatric chronic illnesses, cognitive difficulties),

method used (interviews and/or self-reports, questionnaires), and sample size (e.g., n=110 in Leskinen & Juvonen (1994); n=274 in Lin (2000); and n=8 in the study of Taanila and colleagues (2002)). However, the research addresses the mothers' situation almost exclusively, with less attention paid to fathers, siblings, or the family system.

In Finland, the family adjustment has been studied from the viewpoint of the eco-cultural theory (Leskinen, 1994; Leskinen & Juvonen, 1994; Mattus, 2001; Määttä, 1999). The eco-cultural model proposes that a major adaptive task for each family is the construction and maintenance of a daily routine through which families organize and shape their children's activity and development (Weisner & Gallimore, 1994). From this perspective, a child's participation in routine everyday family activities is the pre-eminent experience shaping the child's development. Sustaining a daily routine means adapting to a local ecology of material and social resources (Ferguson & Ferguson, 1994; Weisner & Gallimore, 1994).

Ferguson (2002) has reviewed a large body of literature of stress and coping in the families rearing a child with disabilities. According to his view, professional researchers have largely reversed their underlying assumptions on family reactions to having a child with a disability. In the past, the emphasis was mainly on how poor and probably disabled parents breed poor and inevitably disabled children. Today, assumed causal connections between disabled child and damaged family have changed into adaptive families. In reviewing the recent research on family adaptation, Ferguson formulates three conclusions: First, there is an increasingly dominant body of research that finds aggregate patterns of overall adjustment and well-being to be similar across groups of families with and without children with disabilities; Second, there is an increasing recognition and growing research that the significant number of parents actually report numerous benefits and positive outcomes for their families associated with raising a child with disabilities; Third, this is not to say that having a child with a disability is not a stressful event. The research needs to continue to refine our understanding of why some families are more resilient than others in adapting to this stress.

The re-evaluation from concepts such as suffering parents or powerless parents into adaptive families has led to an increasing number of strengths-based service programs to delivering services to families, sometimes referred to as a family-support perspective (Ferguson & Ferguson, 1994; Deal, Trivette & Dunst, 1988; Dunst, 2002; Dunst & Trivette, 1993; Mattus, 2001; Trivette, Dunst, Deal, Hamer, & Propst, 1990). The Social Systems Model of Family Functioning, as developed by Dunst, Trivette, and Deal (1988) is an aggregation and synthesis of the family strengths literature how to assess, support and strengthen family functioning. Family strengths are competencies and capabilities of both various individual family members and the family unit that are used in response to crises and stress, to meet needs, and to promote, enhance, and strengthen the functioning of the family

system. This philosophy of practice builds on family members' competencies to make decisions themselves and focuses on enhancing parental involvement wherein they are considered to be one of the most important members of the multi-professional medical team (de Geeter, Poppes & Vlaskamp, 2002). That services delivered in a manner consistent with this philosophy are thought to be more effective and empowering to families and lead to better long-term outcomes has recently been reported by Green, McAllister and Tarte (2004), Mattus (2001), and Määttä (1999). Within this perspective, Ferguson (2002) encourages research to learn more about why some families are more able to cope with stressful events than others.

Factors, such as positive familiar appraisal, support from concerned others, mental support, and social actions, seem to be of importance in establishing family adaptation (Lin, 2000; McCubbin & McCubbin, 1996). In general, these factors have been found to be positively associated with adjustment in families with a child with chronic physical disorder (Ferguson & Ferguson, 1994; Leskinen, 1994; Leskinen & Juvonen, 1994; Mattus, 2001).

Unfortunately, most of these studies provide little information about the complex interaction between the severity of activity limitations and cognitive difficulties and family strengths, simply because instruments used involved respondents' self-reports whereas objective measures of the severity of the child's restrictions are lacking (Lin, 2000; Stolk & Kars, 2000; Taanila, Syrjälä, Kokkonen, & Järvelin, 2000; Young, 2000). Severity of the child's restrictions could be a very important factor concerning the daily experiences of the families (Flynt & Wood, 1989) and could affect family's reactions and ability to nurture the child (Mallory, 1986). The cognitive level of the child is of importance since it may inhibit rehabilitation, particularly, when cognitive difficulties limit the utilization of therapy of aids (Nordmark, Hagglund & Lagergren, 2001).

2.1.2. Social-economical level and age of parents

Broader environmental factors such as social-economical status and family resources are important in the families rearing a child with disabilities (Hogan et al., 2000) because they have been shown to be associated with the quality of parenting. Attitudes of the caretakers about the needs of their child and the possibilities to organize or to provide the needed support for the child exert an important influence on the developmental outcome of the high-risk infants as attitudes are considered to affect behaviour and quality of parenting in a stable way (Stolk & Kars, 2000). No association between the social-economical status of the families and the severity of disability was found in a large sample of 205 American children with developmental disabilities, who ranged in

age from 11 to 87 months (Ottenbacher, Msall, Lyon, Duffy, Granger, et al., 1999). However, cross-cultural comparisons are difficult to make. Whether the social-economical factor is relevant for functional outcomes in Finnish families rearing a child with disabilities is still to be seen.

Lin (2000) has investigated family adaptation in a developmental perspective because phases of family development may shape how family members perceive a specific source of potential stress. She showed that families coping with cerebral palsy differed in family life cycles. Specifically, in Lin's study, families with school-aged children appeared to have better family adaptation than families with adolescents and families with young adults. There is, however, a paucity of the studies investigating the family life cycles in Finnish families rearing a child with disabilities. For instance, little is known about the differences in families of young parents against older parents with respect to family strengths.

2.2. Activity level: functional motor limitations and their relation to skills of everyday life (Studies I, II, III, IV)

Functional motor limitations are often related to the presence of comorbid problems such as cognitive difficulties (i.e. mental retardation; UK usage: learning difficulties; 23-27 %); or sensory deficits (e.g., vision and hearing impairment, strabismus; 6-38 %); or epilepsy (25-33 %) (Sillanpää, 2004). It must be underlined that considerable interindividual variability in the functional skills is exhibited by children with the same neurological problems (Rogers, Msall, Guernsey, Brody, Buck, et al., 1994).

The main aim of this chapter is to summarize some of the major comorbid impairments illustrated by children with motor limitations. Nordmark, Hagglund and Lagergren (2001) have investigated the gross motor function and disabilities in 167 children with cerebral palsy at the mean age of 6.8 years in southern Sweden. The purpose of their study was to investigate and analyse clinical features of gross motor function measured with the Gross Motor Function Classification System (GMFCS; Palisano, Rosenbaum, Walter, Russell, Wood, et al., 1997; Russell, Rosenbaum, Gadman, Gowland, & Jarvis, 1989). It was found that the participating children with hemiplegia were able to walk independently at the age of at least 6 years. The children with tetraplegia had very severe clinical features, all of them classified as GMFCS level V (=severely disabled). These children had no means of independent mobility and were transported. Some children achieved self-mobility using a powered wheelchair with extensive modifications. Children with diplegia presented a varied clinical picture of both impairment and disability, that is, only 10 percent were non-walking children, and 60 percent were classified as level I or II (=mild disability). In addition, a significant positive relation has been

found between the severity of motor function classified with the GMFCS and the associated impairments (IQ, epilepsy and visual impairments) (Nordmark et al., 2001). Out of a total population of 167 children with CP, half of the children (n=81, 49 %) had an IQ above 70. Eighty-five percent of the children with normal or borderline IQ (IQ>70) were classified into levels I and II (=mild disability) and were able to learn to walk. Out of 86 mentally retarded children, 38 (44 %) were severely disabled and classified as GMFCS levels IV and V. In addition, 52 percent of the children with epilepsy and 61 percent with visual impairment were severely disabled and classified as GMFCS levels IV and V. In sum, children with motor impairments show various patterns of developmental trajectories, and severe cognitive difficulties are quite common.

In spite of all research effort, there is a lack of standardized instruments tapping the functional status of the child. Moreover, instruments are needed tapping the transactions between the child and the environment. The latter covers the activity level components described in the ICF model (Campbell, 1996; Graveline, Young & Hwang, 2000; Haley, Coster & Faas, 1991; Musikka-Siirtola, 2005; Simeonsson, 2003). Relevant functional domains for children concerning the routines of everyday life include mobility, self-care, toileting, play, learning, and social cognition (Haley, Coster & Faas, 1991; Korpela & Nieminen, 2001; Nieminen, 2004; Pirilä, Nieminen & Korpela, 1997). In addition, some children are dependent on technology, needing support of medical or assistive devices to compensate for impaired body functions. More systematic, comprehensive and causative data on technology dependent children are urgently needed in this field (Korpela, Seppanen & Koivikko, 1992; Ostensio, Brogen, Carlberg, & Vollestad, 2003; Simeonsson, 2003).

2.3. Body function and structure level: neonatal cranial lesions and higher cognitive functions in cerebral palsy (Studies III, IV)

Many infants experience neurological injuries in the neonatal and/or perinatal period. These injuries can be described in detail due to improved neuroimaging techniques, and their consequences for later development can be prospectively observed. Negative consequences of early damage suggest limits to neural plasticity and may be informative as to brain structure-function relations. Positive outcome after a documented early brain injury suggests that the brain has recovered or compensated in some way for early damage (Krägeloh-Mann, 2005).

Cerebral palsy (CP) refers to a heterogeneous group of impairments characterized by a persistent disorder of movement and posture caused by nonprogressive pathological processes in the immature brain (Aicardi & Bax, 1992; Albright, 1996; Autti-Rämö, 2004; Bax, 1964; Bax, Goldstein,

Rosenbaum, Leviton, & Paneth, 2005; Fernandez-Alvarez & Aicardi, 2001). Although the neuropathophysiology of CP is not yet comprehensively understood, its' associated physiological disturbances have been provided by the research in developmental neurobiology and neuroscience. In the great majority of cases, either hemorrhage and/or periventricular leukomalacia (PVL) reflecting necrosis of the periventricular white matter are the primary causes most likely to produce the definite brain cell damage (Filloux, 1996). Overall, CP has been observed to occur in 2 to 2.5 children per 1000 by early school age (Autti-Rämö, 2004). Considering the life-long effects of the disability, children with CP deserve our research attention. Unfortunately, children with major handicapping conditions, including CP, are excluded in longitudinal risk studies most of the time, as reviewed by Ornstein, Ohlsson, Edmonds and Asztalos already in 1991. Since then, little has changed. Whereas follow-up studies focused on CP have limited themselves to the motor part of the disability, ignoring the cognitive development that is considered one of the main factors determining the quality of life the child will enjoy (Nelson, Swaiman, & Russman, 1994). At best, such studies report the IQ/DQ (intelligence or developmental quotient) index (Nordmark, Hagglund, & Lagergren 2001) .

One may have doubts about whether an IQ index is enough to describe the cognitive functioning of motor handicapped children, especially when they reach school age and beyond. By that age, it is assumed that the more subtle long-term morbidities in areas of learning, visuomotor integration and language performance become more detectable because these difficulties usually do not manifest themselves much in infancy. Within this perspective, the study of Olsen, Vainionpää, Pääkkö, Korkman, Pyhtinen and Järvelin (1998) is of importance. In addition to IQ, the researchers used a neuropsychological test (NEPSY) to investigate attention, verbal abilities, perceptual-motor skills and memory in 41 preterm children with a mean age of 8 years. Thirteen children had PVL and out of them, four had developed CP (two suffered from spastic diplegia, the other two suffered from spastic hemiplegia and dystonic tetraplegia). As a group, the children performed poorly in tasks requiring spatial and visuo-perceptual abilities which were associated with the finding of PVL in MRI, especially with posterior ventricular enlargement. Surprisingly, the children with CP performed as good as the clinically healthy preterm children. However, as the authors themselves admitted, the small number of children with CP made it difficult to make any strong conclusions concerning their cognitive functioning.

Witelson (1987) has reviewed experimental outcome on the effects of prenatal damage on the cognitive and neuropsychological development. She underlined that the development of language functions take precedence at the expense of visuospatial functions. Better verbal than nonverbal cognitive functioning connected with motor disorders, especially with CP, has also been reported by others (Carlsson et al., 1994; Fazzi et al., 1994; Fedrizzi et al., 1996). However, many

children with motor impairment are also hindered in their narrative skills and functional communication (Basil, 1992; Jolleff, McConachie, Winyard, Jones, Wisbeach, et al., 1992; Light, Collier & Parnes, 1985; Pennington, 1999; Pennington, Goldbart, & Marshall, 2004; Pennington, Goldbart & Marshall, 2005; Pennington & McConachie, 1999). Children with CP rarely initiate exchanges in conversation with familiar adults, taking a largely respondent role, while adults introduce topics and start most conversations. They also take fewer turns in conversation than do their adult partners and often fail to reply unless obliged to do so. In addition, they produce many yes/no answers and seldom ask questions (Pennington, 1999; Pennington & McConahie, 1999).

Two factors have been shown to be important in communication and speech problems. First, poor speech production may be a direct result of the motor impairment due to a disturbed neuromuscular control of speech mechanism, i.e. dysarthria (Crary, 1995; Pennington, 1999; Pennington & McConachie, 2001; Strand, 1995). The motor speech disorders vary with the neurological subsystems involved and contribute to reductions in speech intelligibility or naturalness (Crary, 1995; Strand, 1995). The second factor involved in language and speech problems is intellectual difficulties (Aicardi & Bax, 1992; Nordmark, Hagglund & Lagergren, 2001). Deficits in intellectual functioning cover beside speech production problems also content aspects of language, such as active and passive vocabulary, use and comprehension of grammatical structures, etc. The two factors are not mutually exclusive.

What do we know about the relationship between neonatal cranial ultrasound (US) abnormalities and domains of higher cognitive functions? Holling and Leviton (1999) have reviewed 15 studies that satisfied the criteria of a sample size of at least 10 infants, details about the size, extent or location of the cerebral white-matter echolucencies seen on cranial ultrasound scans, and a follow-up assessment (i.e. motor, cognitive, behavioral, sensory, and so) related to echolucency characteristics. A total of 373 infants diagnosed with echolucencies were followed, out of which 59 percent developed CP. Cognitive dysfunction, especially an IQ index below 70, was directly proportional to the size and extent of the ultrasonographically-defined white-matter echolucencies. Their pooling, however, did not reveal any specific locations within the brain critical to cognitive impairment which is in keeping with other observations about structure-cognitive function relationships. Also, many children with spastic diplegia had a small number of small echolucencies and were relatively intact, cognitively. In addition, in 9 studies that compared unilateral and bilateral involvement, bilateral lesions occured nearly three times more than unilateral lesions.

With respect to the association between the neonatal cranial US findings and later outcome, the following can be said. First, the association between the neuroanatomical lesions and the clinical features is far from perfect. Cystic leukomalacia usually results in severe motor limitations.

However, cerebral palsy can occur without any detectable lesion in the newborn period, and noncavitated echogenic areas may disappear without sequelae (Olsen, Pääkkö, Vainionpää, Pyhtinen, & Järvelin, 1997; Pinto-Martin, Riolo, Cnaan, Holzman, Susser, et al., 1995; Rogers, Msall, Owens, Guernsey, Brody, et al., 1994). Nevertheless, neonatal cranial serial US abnormalities predict impairments in perceptual-motor (Fujimoto, Yamaguchi, Togari, Wada, & Yokochi, 1994; Mercuri, Guzzetta, Haataja, Cowan, Rutherford, et al., 1999; de Vries, Eken, Groenendaal, van Haastert, & Meiners., 1993; van Wezel-Meijler, van der Knaap, Oosting, Sie, de Groot, et al., 1999), and mental functions (Biagioni, Bartalena, Boldrini, Pieri, & Cioni, 2000; Ringelberg & van der Bor, 1993; de Vries, Rademaker, Groenendaal, Eken, van Haastert, et al., 1998). The majority of these studies investigated children when they were younger than 3 years of age, well before more complex cognitive skills and higher motor functioning could be tested. In the current thesis, an attempt was made to evaluate the degree of association between cranial US abnormalities detected early in life, functional motor limitations, and higher cognitive functions when the children achieved pre-school or elementary school age.

2.4. The aims of the thesis

The present thesis tries to examine linkages between three levels: first, the level of most immediate environment, the family a child is living in: family strengths, social-economical status, age of primary caregiver, amount of caregiver assistance; second, the level of individual activities and activity limitations: functional gross and fine motor skills, functional skills in self-care, mobility and social function; and, third, the level of body functions and structure: neonatal brain lesion, birth weight, age, intellectual functions, neuropsychological functions, language and motor speech skills (Figure 2).

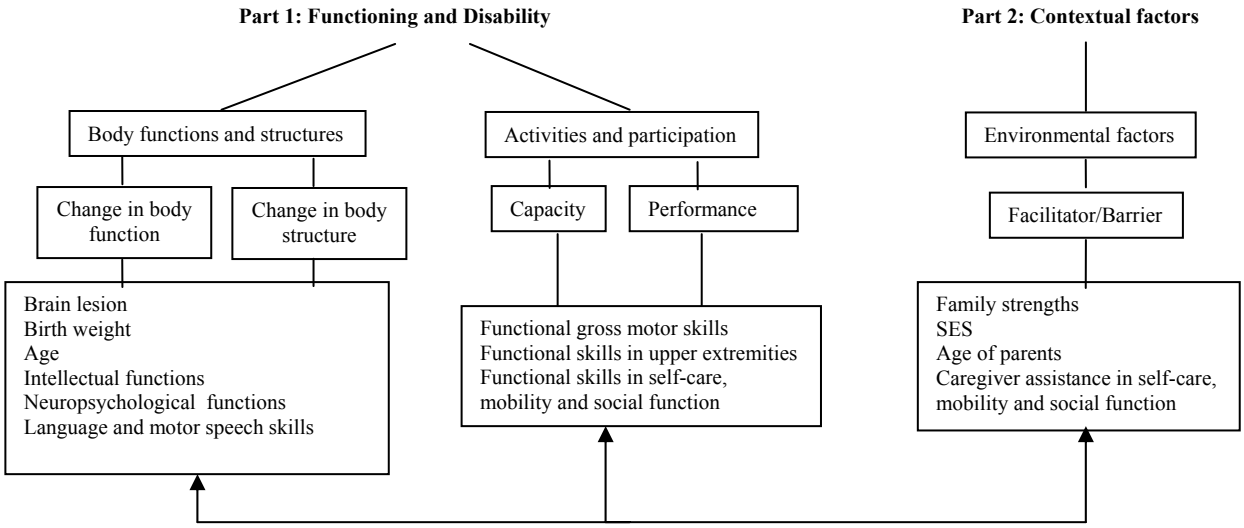

Figure 2: The variables investigated in the current study applied into the structure of ICF.

More specifically, answers were sought to the following questions:

1. Study I focused on the family strengths and their relation to the disability-related characteristics of a child with the following main question: What are the relationships between the family strengths and the severity of activity limitations in children indexed by functional motor limitations and cognitive difficulties, and the family characteristics indexed by social-economical status and age of caretakers?
2. Study II was of an explorative nature (i.e. pilot study). In comparison to study I, the study II investigated in more detail some aspects of activity limitations, namely self-care, mobility and social function, with respect to the family strengths and caregiver assistance. More specifically, what is the relationship between the functional skills of the children, the amount of caregiver assistance and the family strengths? In addition, the amount of assistive devices and environmental modifications in daily use were investigated.
3. Study III focused on the issues of language and speech skills in children with cerebral palsy with the following main questions: What are the associations between the severity of activity limitations indexed by gross and fine motor problems, cognitive difficulties, language, and motor speech problems of such children. In addition, is there an association between the neonatal cranial ultrasound findings and later outcome? It was expected that the severity of the CP is primarily expressed in motor speech skills, whereas CP with comorbid cognitive difficulties will produce additional language deficits in the expressive and comprehensive domains. No specific hypothesis

was introduced between the neonatal US findings and later outcome in language and motor speech skills.

4. Study IV focused on the issues of neuropsychological functions in children with spastic diplegia with the following main questions: What is the neurocognitive profile of children with diplegia spastica? In addition, what is the relation between the neonatal cranial ultrasound and later intelligence and neuropsychological outcome? We expected the most pronounced cognitive deficits in the perceptual-motor domain whereas verbal and memory functions will be relatively intact. Given that diplegia is not generally associated with cognitive difficulties (i.e. mental retardation), no correlation between IQ level and US findings was expected in our sample. However, a high correlation was expected between the cranial US and limitations concerning the gross motor functioning of lower extremities (walking) and fine motor functioning of upper extremities.

3. METHOD

Sixty-nine children receiving special health care services in the Department of Pediatric Neurology, Tampere University Hospital, for identified developmental disabilities participated in the current study. Most of the children (n=61) and their caretakers were recruited from the early intervention program for disabled children, the Play Project (Korpela & Nieminen, 2001). The Play Project was carried out by the Department of Pediatric Neurology, Tampere University Hospital, and the Department of Psychology, University of Tampere, during 1993-1997. The basic aims of the Play Project were to develop and evaluate social competence, play and rehabilitation of children with disabilities. The development of the children was evaluated with traditional methods used by doctors, nurses, psychologists, social workers, rehabilitation counsellor, physiotherapists, occupational therapists and speech therapists. In addition, a transdisciplinary play based assessment and rehabilitation method was developed (Leikkitarkkailu. Käsikirja lapsen toiminnallisen kehityksen arvioimiseksi, 2004).

In the current thesis, majority of the sample, most of the measures and information on family characteristics were based on the compilation of multiprofessional team work in the Play Project. The original sample of the Play Project was randomly selected out of the population of disabled children born in 1989-1994. For methodological reasons the sample was enlarged with eight children later. The children were as infants treated in the neonatal intensive care unit and followed up later in the Department of Pediatric Neurology in Tampere. Due to prenatal or perinatal insults most of them produced functional motor impairment over time. The extent of disability ranged from mild impairment to severe multiple disabilities.

3.1. Participants and procedure

Study I:

Sixty children (36 boys and 24 girls) with functional motor limitations caused by a variety of prenatal or perinatal problems (such as periventricular leukomalacia, hypoxic-ischaemic encephalopathy, brain structural anomaly, meningomyelocele, intracerebral hemorrhage, chromosomal abnormality, etc.) and their caretakers were selected randomly for the present study. For main diagnoses of the sample, see Table 1.

Table 1:

Main diagnosis	n
Cerebral palsy	36
Meningomyelocele	5
Brain structural abnormality	7
Motor/ psychomotor delay	12
	60

The mean age of the children was 3 years and 11 month (SD = 21 months) with an age range from 15 months to 7 years 3 months. The mean ages of the mothers and fathers were, respectively, 34 years (SD = 5 years) with a range from 21 to 42 years, and 36 years (SD = 5 years) with a range from 27 to 48 years. All caregivers were birth parents. Nine mothers (15 %) were non-married (single, widow or divorced). The families were grouped on the basis of the parents' education and occupation into three social-economical categories. The sample consisted of about 72 percent of the total number of families living in the region of Tampere University Hospital rearing a child with activity limitations in this age range and therefore could be seen as representative.

Study II:

The subjects (12 boys and 9 girls) were in the age range from 2 years 10 months to 6 years 2 months (Mean = 4 years 2 months, SD = 12) when they participated in the study. These children had experienced a variety of pre- and perinatal problems (see Table 1). The children formed a randomly selected subgroup of the study I sample. Thirteen children had as a main diagnosis cerebral palsy (n = 8 diplegia and n = 5 quadriplegia). Six children had brain structural anomalies among others

meningomyelocele and / or hydrocephalus. One child had the Williams chromosomal anomaly and one child had a motor developmental delay due to a heart disease. Nine out of 21 children were born premature (gestation age < 37 weeks with a range from 28 to 36 weeks) with a birth weight range from 1280 g to 2870 g. The mean age of the mothers was 35 years with a range from 24 to 45 years. All caregivers were birth parents. The social-economical stratification (SES) was made on the basis of the years and type of professional education of both parents.

Study III:

This sample of convenience consisted of 36 premature born children (18 boys and 18 girls, GA < 37 weeks) in the age range of 1 year 10 months to 9 years 0 months (Mean = 5 years 1 month; SD = 21 months) suffering from CP caused by periventricular leukomalacia (PVL). Majority of them (n = 28 children) were part of the study I sample. Children lived in the environment of Tampere University Hospital, Finland, at the time that this retrospective study was carried out (years 1997-2002). Twenty-two children developed diplegia with spastic paresis most pronounced in the lower extremities, 5 children developed hemiplegia, and 9 children developed quadriplegia with the paresis most pronounced in the upper extremities. Three children with cognitive difficulties were severely hearing impaired. The clinical characteristics of the sample are depicted in the study III, p. 6.

Study IV:

Fifteen children (9 boys and 6 girls) were selected out of a total sample of 27 children suffering from spastic diplegia in the age range of 5 to 12 years living in the region of Tampere University Hospital. The clinical data per subject (gestational age, birth weight, Apgar scores in 1 and 5 min, US classification of PVL, the neonatal and additional problems, the functionality of lower and upper extremities, and IQ scores) is shown in the study IV, pp. 685-686.

3.2. Measures

3.2.1. Environmental factors

Family strengths (Studies I-II)

Family strengths were explored using the Family Functioning Style Scale (FFSS; Deal, Trivette & Dunst, 1988; see Study I, Table 2, p. 285-286). Family strengths are defined in terms of competencies and capabilities of family members and of the family unit as a whole that are used in response to crises and stress, to meet needs, and to promote, enhance, and strengthen the functioning of the family system. The scale items (n=26) are organized into three categories that are believed to represent distinct (but not independent) aspects of family functioning style: Family identity, Information sharing, and Coping/resource mobilization. The Family-identity category measures five aspects of family strengths: (1) commitment toward promoting the well-being and growth of individual family members as well as that of the family unit, (2) appreciation for the small and large things that individual family members do well and encouragement to do better, (3) allocation of time for family members to do things together, (4) sense of purpose that permeates the reasons and basis for going on in both bad and good times, (5) congruence among family members regarding the importance of assigning time and energy to meet needs. The Information-sharing category measures (1) communication among family members in a way that emphasizes positive interactions, (2) rules and values that establish role expectations about acceptable and desired behaviour. The Coping/resource mobilization category measures (1) internal and (2) external coping strategies that promote positive functioning in dealing with both normative and non-normative life events, (3) problem solving employed to meet needs and procure resources, (4) positivism in most aspects of living, including ability to see crises and problems as an opportunity to learn and grow, (5) flexibility and adaptability in the roles necessary to produce resources to meet needs, and (6) balance between the use of family resources for meeting needs. Participants are asked to respond to each item using a 5-point rating scale ranging from 'not at all like my family' to 'almost always like my family'. A mother completed the list. The scale has been translated into Finnish (Mattus, 1994). Trivette and colleagues (Trivette, Dunst, Deal, Hamer, & Propst, 1990) have evaluated the psychometric characteristics of the measure. In the current sample the Cronbach alpha coefficient in the whole list was .92. For the subscales Family identity (10 items), Information-sharing (4 items) and Coping/resource mobilization (12 items), the alpha coefficients were respectively .86, .74 and .75. The collection of the data was conducted by a social worker and a rehabilitation counsellor.

Functional limitations in self-care, mobility and social function and caregiver assistance (Study II)

The Pediatric Evaluation of Disability Inventory (the PEDI; Haley, Coster & Faas, 1991; Haley et al., 1992; Coster & Haley, 1992) is usually given to the primary caregiver and/or to the primary therapist whose perceptions of the child cross all or most of the environments in which the child adapts and functions. The estimated test time required is 45 - 60 minutes. The parent is asked a series of forced-choice questions about the child, and responses are based on typical daily interactions with the child. In the current study, a specially trained therapist who knew the family and the child well carried out the structured interview with the mother. The inventory measures functional performance in the domains of Self-care, Mobility and Social Function. In addition, the inventory measures the level of caregiver assistance needed to accomplish functional activities in the domains of Self-care, Mobility and Social Function. Each caregiver assistance scale is made up of a set of 20 content areas that are scored with a 6-point rating scale. The scoring is the following: 5=Independent; 4= Supervision/Set-up; 3= Minimum assistance; 2= Moderate assistance; 1= Maximal assistance; 0=Total assistance. Moreover, the inventory provides information regarding environmental modifications and equipment used by the child as important adjunct information for understanding the child's functional capability. The Modifications scales are scored with a 4-category scale that reflects the extent of modifications used for an individual item (None; Child-oriented modifications; Rehabilitation modifications; Extensive modifications).

For the Functional skills and Caregiver assistance scales, raw scores are converted to normative standard scores which are based on the chronological age of the child and allow comparison to the normative sample. The mean normative standard score for each age group is 50, with a standard deviation of 10. Thus, the expected range of scores for typically developing children is between 40 and 60. For the Modifications scales, only frequency counts are available. With respect to the content validity of the measure, the reader is referred to Haley, Coster, & Faas (1991), Nichols & Case-Smith (1996) and McCarthy et al. (2002). The collection of the data was conducted by a physiotherapist.

Social-economical status (SES) and age of parents (Studies I, II, III)

Families were grouped on the basis of the parents' education and occupation into three social-economical categories: Category 1: professional education less than 2 years and working class profession (for example cleaner, plumber); category 2: professional education 2 to 4 years and

white-collar profession (for example bank officer, merchant); category 3: professional education more than 4 years and academic profession (for example teacher, architect, economist). Age of caregivers at the time of the assessment was also taken into consideration. The collection of the data was conducted by a social worker.

3.2.2. Activities and participation

Assessment of gross motor limitations (Studies I, III, IV)

The Autti-Rämö's Scale (Autti-Ramö, 1996; ARS) was used to measure the amount of assistance needed in gross motor functioning. The scale classifies the amount of assistance needed in gross motor functioning as mild (I), moderate (II) or severe (III) according to the following criteria:
I) mild disability: the child is able to learn to walk without any assistance, although with spastic gait.
II) moderate disability: the child may learn to walk assisted.
III) severe disability: the child is unable to learn to walk or to use functionally the handicapped part of the body. Instead, he/she may be able to learn to manage a standard electric wheelchair.

It is to be noted that Level I (mild) of the ARS is comparable to the Levels I and II of the Gross Motor Function Classification System (GMFCS) (Nordmark et al., 2001), Level II of the ARS (moderate) equals Level III of the GMFCS, while Level III of the ARS is equivalent with Levels IV and V of the GMFCS.

The scoring of the scale was conducted independently by a pediatric neurologist and a physiotherapist. In the Study I (n=60), a kappa coefficient, indexing the interrater reliability, was 0.73.

Assessment of the functional limitations in self-care, mobility and social function (Study II)

The Pediatric Evaluation of Disability Inventory (the PEDI; Haley, Coster & Faas, 1991; Haley et al., 1992; Coster & Haley, 1992) was used to measure the functional performance of children in the domains of Self-Care (eating, grooming, bathing, dressing, toileting; 73 discrete items: 0 = not able/or limited capability, 1 = capable), Mobility (transfers, indoor locomotion, stairs; 59 discrete items : 0 = not able/or limited capability, 1 = capable) and Social Function (comprehension, expression, problem resolution, play, self and time information, management daily routines; 65 discrete items : 0 = not able/or limited capability, 1 = capable). The number of capable scores is then summed for each content area to give an overall score for that skill. See also chapter 3.2.1. for more throughout information of

the scale. A specially trained therapist who knew the family and the child well carried out the structured interview with the mother. The collection of the data was conducted by a physiotherapist.

Assessment of the functionality of upper extremities (Studies III-IV)

Functionality of the upper extremities (i.e. fine motor limitations) was classified according to a three-grade scale: normal; immature; or deviant concerning muscle tonus, symmetrical use of the hands, and fine motor manipulations. The administration and scoring of the scales was conducted by a pediatric occupational therapist.

3.2.3. Body functions and structure

Assessment of cognitive functions (Studies I-IV)

Studies I-II

Cognitive development was assessed using the Swedish standardized version of the Griffiths Scales of Mental Development (Griffiths, 1970, 1984; Alin-Åkerman & Nordberg, 1991). Test scores were transformed into developmental quotient scores (DQ) using the formula: (developmental age/chronological age) x 100. The administration and scoring of the scale was conducted by a clinical psychologist.

Study III

The Swedish standardized version of the Griffiths Scales of Mental Development (Alin-Åkerman & Nordberg, 1991; Griffiths, 1970, 1984) was used to measure the intelligence level of children younger than 3 years of age. The Wechsler Preschool and Primary Scales of Intelligence – Revised (WPPSI-R; Wechsler, 1989) was used for children in the age range of 3 to 6.5 years and The Wechsler Intelligence Scale for Children – Revised (WISC-R; Wechsler, 1974) was used for children older that 6.5 years. The administration and scoring of the scales was conducted by a clinical psychologist.

Study IV

The cognitive level was assessed with the aid of the Finnish standardized version of the Wechsler Intelligence Scale for Children - Third edition (the WISC-III). Two children were 5 years of age. To estimate their IQ level the WPPSI-R was used. The neurocognitive assessment was made using the NEPSY: A Developmental Neuropsychological Assessment (Korkman et al., 1998). The measure consists of 30 tests that tap various aspects of attention, language, sensorimotor and visuospatial functions, memory and learning. Because of the disabilities of the target group and time constrains, the current study limits itself to 14 tests, presented in the results section. The administration and scoring of the scales was conducted by a clinical psychologist.

Assessment of language and motor speech functions (Study III)

Comprehensive and expressive language skills

The Reynell Developmental Language scale - Revised (RDLS; Reynell, 1977) was used to measure verbal comprehension skills (passive vocabulary and comprehension of grammatical structures). Expressive language skills (active vocabulary and grammatical skills) were measured using the Sproglig Test I by Bo Ege (1985) and the Verbal Language Development Scale by Merlin J. Mecham (1958). The Bo Ege Sproglig test is a screening test used to assess the expressive skills of children in the age range of 0 to 7 years. The Mecham Verbal Language Development Scale is administered by an informant-interview. Items cover ages 0-15. The scale is an extension of Vineland Social Maturity Scale. The raw scores of the comprehension and the expressive tests were transformed into Age Equivalent (AE) scores, and classified as normal (AE within the range expected for child's age); immature (AE delay more than 3 months and less than 1 year expected for child's age); or deviant (AE delay more than 1 year expected for child's age). The administration and scoring of the scales was conducted by a speech therapist.

Assessment of motor speech skills

The assessment of oral motor patterns and structure used in simple and complex orofacial movements and velopharyngeal functions (i.e. swallowing, tongue protrusion, lateral tongue movements, licking off the lips, whistling, blowing, etc.) was rated as normal (age-appropriate oral motor patterns and structure); immature (minor 1 to 2 difficulties); or deviant (difficulties in at least

3 aspects of oral motor functioning or structure). Phonology and articulation skills were classified as either normal (speech is intelligible and appropriate of the age); immature (speech is intelligible but contains 1 to 2 prominent, not age-appropriate features); or deviant (speech is less intelligible or unintelligible without contextual cues or contains 3 or more prominent deviations). The administration and scoring of the scales was conducted by a speech therapist.

Neonatal cranial ultrasound (Studies III, IV)

During the first hospitalization, infants were examined by cranial US at least at the ages of 2 and 3 days, and at 1 to 2 weeks intervals afterwards, using an Aloka SSD-900 mechanical sector scanner with a multifrequency transducer (7.5 MHz crystals). The 7.5 MHz probe was used to ensure the best possible resolution. PVL was classified according to de Vries et al. (1993) as I (mild), II (moderate), and III (severe). Grade I: periventricular areas of increased echogenicity present for seven days or more or mild structural anomaly; Grade II: loss of brain tissue from any cause, including small localized frontal, parietal or occipital cysts or irregular enlargement of the ventricular system; Grade III: periventricular areas of increased echogenicity evolving into extensive periventricular cystic lesions involving occipital and/or fronto-parietal periventricular white matter or generalized atrophy of the brain from any cause. Peri- or intraventricular hemorrhage (PVH, IVH) grading was performed according to Papile et al. (1978). A pediatric neurologist and a neonatologist (study IV, part of data in the study III) were independently performing the US classification in the year 2003. Both were not aware of the neurocognitive findings of the children, thus improving the reliability of the US grading. Inconsistencies between the two scorings regarding the presence or the severity of PVL did not take place.

Table 2 depicts the summary of the methods and participants used in the studies I to IV.

Study I, n=60 children with functional motor limitations

Family variables:
FFSS
SES, age
Child variables:
ARS scale
Griffiths DQ index
Age

Study II, n=21 children with functional motor limitations

Family variables:
FFSS, SES, age
PEDI: caregiver assistance
Child variables:
PEDI: functional limitations
Griffiths DQ index
Age

Study III, n=36 children with cerebral palsy

Family variables:
SES, Age
Child variables:
Griffiths DQ or Wechsler IQ index
ARS scale, US grading, birth weight
Reynell & Mecham Age Equivalent (AE) scores
Motor speech AE scores
Sum score of functionality of upper extremities

Study IV, n=15 children with diplegia spastica

Child variables:
Wechsler IQ index
NEPSY tests
US grading, ARS scale
Scores of functionality of upper extremities

Table 2: Summary of the participants and measures.

3.3. Data analyses

Study I

Correlation analyses (The SPSS, 10.1) were carried out to estimate the relationship between the indices of restrictions, age, social-economical status and family strengths (the FFSS scale). The Bonferroni correction procedure was used in order to avoid that the experimentwise Type I error probability will not exceed alpha. A multiple regression analysis was used to test the following statistical hypothesis: There is no linear relationship between family strengths and the severity of the participation restrictions of the child and the social-economical status of the families. T-tests were used to compare domains of family functioning in families rearing a child with severe or mild limitations. P values <.05 were considered to be statistically significant.

Study II

Nonparametric methods (The SPSS, 10.1) were used whenever possible because of the small sample size and the difficulties in making any assumptions concerning the sample distribution. The Spearman

rank correlation coefficient was used as a measure of association to explore the relationships between the scales. The Bonferroni correction procedure was used in order to avoid that the experimentwise Type I error probability will not exceed alpha. Differences between the subgroups and the normative scores were expressed by one-way ANOVA and t-tests. P values <.01 were considered to be statistically significant.

Study III

Data were analysed with the Spearman rank correlation coefficients (The SPSS, 10.1) to estimate associations between gross and fine motor limitations. The Chi-square test of independence with the Yates' correction for continuity for 2 X 2 tables was used to explore the relation between motor speech and language problems as function of the severity of the CP and the IQ/DQ index. A multiple regression analysis was used to test the following statistical hypothesis: there is no linear relationship between the outcome variables (functional gross and fine motor limitations, intelligence, speech, and language skills) and the predictor variables (neonatal US findings, birth weight and social-economical status). P values less than .05 were considered to be statistically significant.

Study IV

Statistical analyses were made by using nonparametric methods (The SPSS, 10.1) because of the difficulties in making any assumptions concerning the normality of the sample distribution. Spearman rank correlation coefficient was used to explore the linkages between the perinatal predictors and the outcome. Mann-Whitney U test was used to measure subgroup differences concerning the NEPSY. Wilcoxon Signed Ranks test was used with repeated measures. P values less than .05 were considered to be statistically significant.

4. OVERVIEW OF THE ORIGINAL STUDIES

Study I

Pirila, S., van der Meere, J.J., Seppanen, R-L., Ojala, L., Jaakkola, A., Korpela, R., & Nieminen, P. (2005). Children with functional motor limitations: The effects on family strengths. *Child Psychiatry and Human Development, 35 (3), 281-295.*

This paper reports the results of an investigation of the association between family strengths and the severity of functional motor limitations and cognitive difficulties of the children. Subjects of the study were 60 children in the age range 15 months to 7 years 3 months. The family strengths were assessed by the Family Functioning Style Scale (FFSS), the severity of the functional motor limitations by the Autti-Ramo's Scale (ARS) and the cognitive level by the Griffiths Scales of Mental Development (DQ). This study controlled for some important family characteristics, such as social-economical status (SES) and age of primary caretaker (AGE).

Results indicated that the families, even those rearing children with severe participation restrictions, scored above 5 (range from 0 to 8) in most of the subscales, indicating that the strengths were rather high. A score of 5 means that the majority of families identified themselves more to the positive end of the strengths-based statements ('almost always like my family') than to the negative alternative ('not at all like my family'). Thus, they coped relatively well with their family life (Figure 3). However, on the subscales internal coping (promoting positive functioning in the family) and communication (sharing concerns and feelings in productive ways) the families scored below 5. The age of the child and the caretakers and the social-economical status of the families were not related to family strengths.

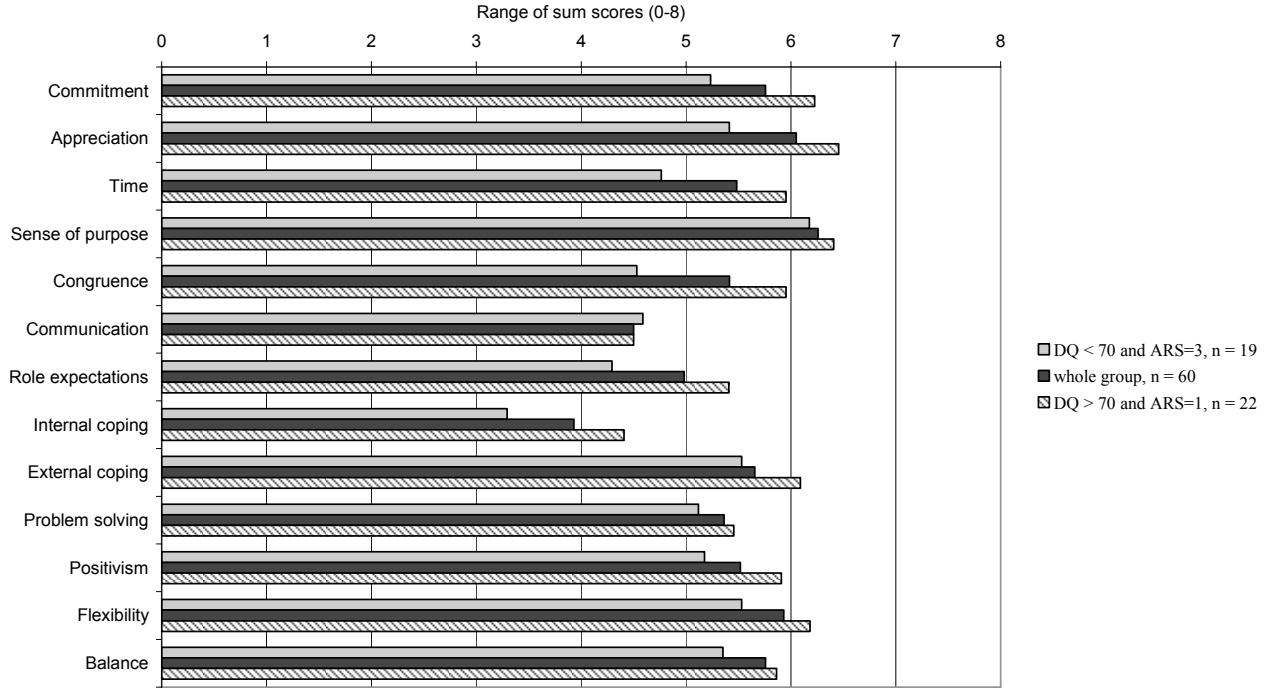

Figure 3 FFSS subscale scores of the three groups.

In order to estimate the contribution of the predictor variables (ARS, DQ, SES, and AGE) to family strengths (FFSS), a multiple regression analysis with Enter Method was carried out (Table 3). The factors accounted for only 12 percent of the variation in the FFSS.

Variables included in the model		Model F (df)	R^2	$^{Adj}R^2$	Beta	SE
Dependent	total sum score of FFSS	1.8 (4, 55)	.12	.05		14.0
Predictors	ARS				-.10	
	DQ				.20	
	SES				.22	
	Age_{mother}				.04	

Table 3 Prediction of family strengths from the variables of disability and family characteristics.

However, the combination of the severity of the functional motor limitations and cognitive difficulties of a child, showed some effects on family functioning (Figure 3). More specifically, differences between families rearing children with severe participation restrictions (i.e.

DQ < 70 and ARS = 3; n = 19) and mild participation restrictions (i.e. DQ > 70 and ARS = 1; n = 22) were most pronounced in the subscales concerning time ($t = -2.54$, $p < .02$), congruence ($t = -3.49$, $p < .00$), and internal coping ($t = -2.00$, $p < .05$). Also the sum scores of the categories Family identity (i.e. sum score of the domains commitment, appreciation, time, sense of purpose, and congruence) and Coping/resource mobilization (i.e. sum score of the domains internal coping, external coping, problem solving, positivism, and flexibility) were higher in the families rearing a child with mild restrictions (t-test, $p < .04$). Note: The two groups did not differ with respect to the main diagnoses of the children, the age of the children, the age of the caregiver, and the distribution of the SES.

The results suggested that identification of the complex interaction between the severity of participation restrictions of a family member and environmental characteristics, such as family strengths, is important in order to provide the necessary support for families. It is proposed that the combination of multiple severe participation restrictions of the children is taxing the coping strategies of the families.

Study II

Pirila, S., van der Meere, J.J., Seppanen, R-L., Korpela, R., & Nieminen, P. (in press). A pilot study on children with limitations in self-care, mobility and social functions: effects on family strengths. *Families in Society. The Journal of Contemporary Social Services*.

This pilot study explored the functional skills in the areas of self-care, mobility and social functions in 21 children (age range 2 years 10 months to 6 years 2 months) with activity limitations caused by a variety of brain complications sustained pre- or perinatal. For this purpose, the Pediatric Evaluation of Disability Inventory (PEDI) was used. Scores were related with the Family Functioning Style Scale (FFSS) in order to study the extent to which caregiver assistance and family strengths were related to each other. In addition, the relation with the cognitive level of the children was investigated.

Results indicated that the children had many functional limitations in the domains of self-care, mobility and social function. These scores were significantly below the normative score of 50 with p values <.00 (one-sample t-test) expressing severe limitations of the children. In these domains the functional limitations triggered the need for assistance by the caregivers expressed by their scores of 29 (SD = 14) and 22 (SD = 14) concerning self-care and mobility (Figure 4). However, an important difference in mean scores between the children's functional skills and

caregivers' level of assistance was found in the domain of social function. With a mean score of 35 (SD = 14), the children expressed their functional limitations (with a p value <.00) compared to the normative mean) concerning their social functioning. In spite of the functional limitations of the children in this domain, the caregivers had a mean score of 47 (SD = 21) indicating that the assistance they offered did not differ from the normative mean (p value > .05).

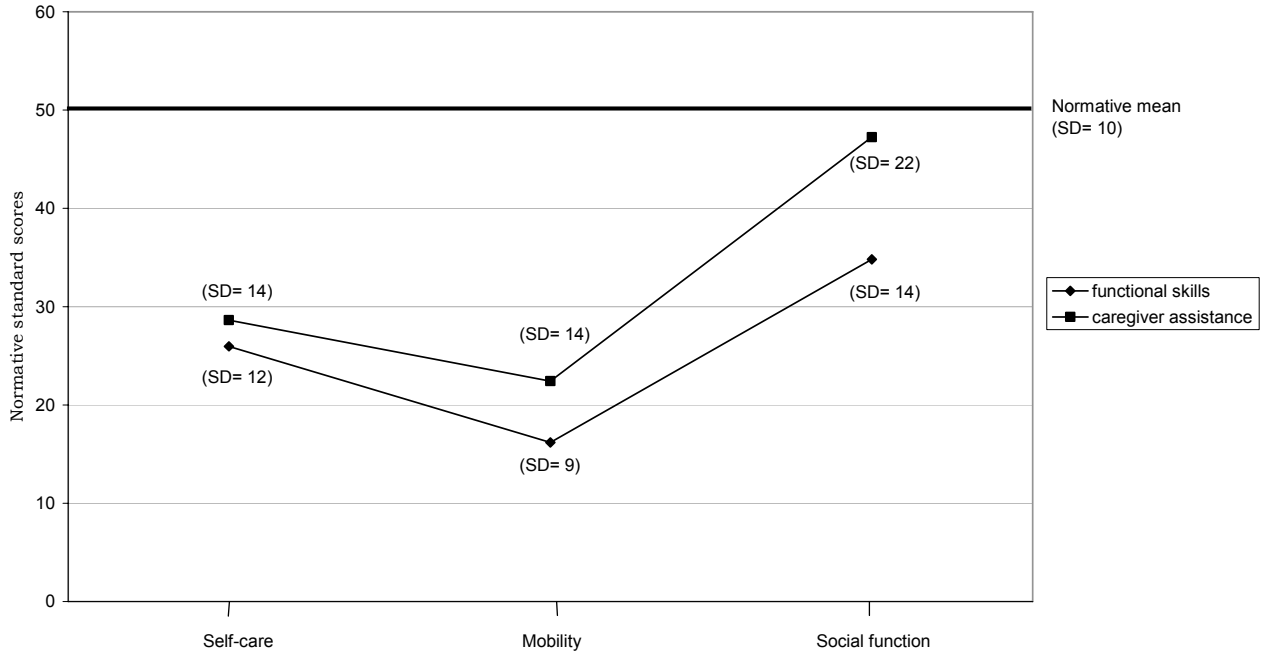

Figure 4 Normative standard scores of functional skills and caregiver assistance of three domains of the PEDI.

This phenomenon is illustrated in more detail in Figure 5 which presents the relation between the normative standard scores of the functional skills in the three domains and its' corresponding amount of caregiver assistance. The Figure 5 shows that in the domain of self-care, 15 children scored in the 3rd quadrant indicating that they had limitations in self-care (normative score < 40) and received the needed amount of caregiver assistance (normative score < 40). Only 3 children scored in the 4th quadrant indicating limitations in self-care (normative score < 40) without receiving the appropriate caregiver assistance (normative score > 40) (Spearman rho between the functional limitations and caregiver assistance = .83, p < .00). The same trend was observed in the mobility domain. Here, 18 children scored in the 3rd quadrant, and 3 scored in the 4th quadrant (Spearman rho = .81, p < .00). In the social function domain, 7 out of 21 children scored in the 4th

quadrant indicating that about one-third of the families had difficulties in recognizing the needs of their child for assistance in this domain (Spearman rho = .80, p < .00).

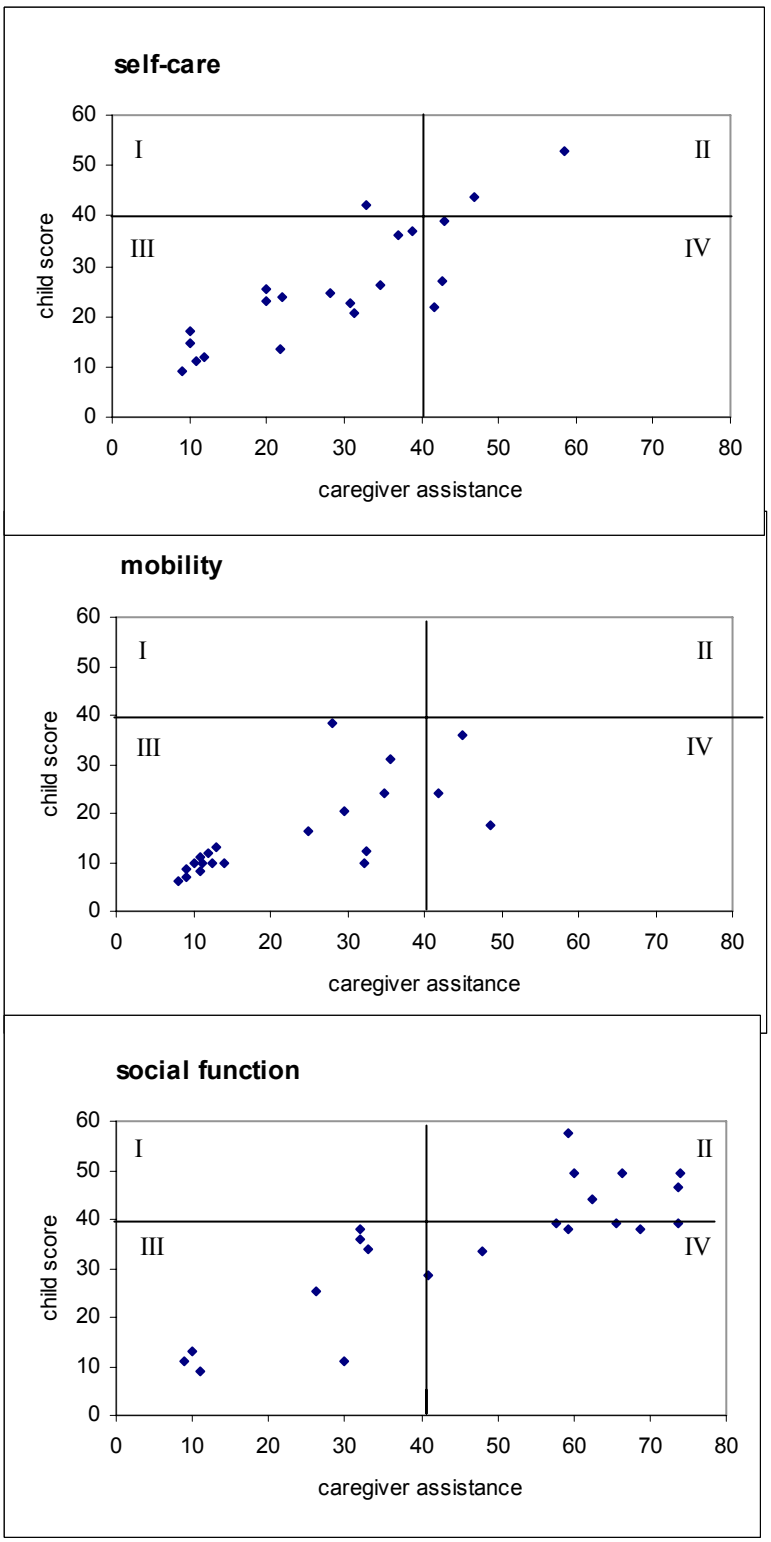

Figure 5 Correlations between the normative standard scores and the caregiver assistance scores of three domains of the PEDI.

When comparing the DQ of the children in different quadrants it was shown that the assistance the caregivers offered did not differ from the normative mean when children had a DQ at or close to norms, which was the case in the 2nd quadrant (independently functioning children, DQ Mean 80 (SD = 12) and the 4th quadrant (discrepant group: functional limitations in the social domain without appropriate assistance, DQ Mean 83 (SD = 11), compared to the children in the 3rd quadrant (DQ Mean 39 (SD = 32). Note: Using one-way ANOVA analysis, the mean ages of the children and the mothers of the 2nd, 3rd and 4th quadrant did not differ from each other, neither was the SES different between the families in the quadrants. The suggestion that the DQ of the children played a mediating role regarding the disparity of the skills of the children and the caregiver assistance was confirmed by the finding that the three children who had discrepant scores in the self-care and mobility domain (see Figure 5) also had a DQ level in the normal range.

When evaluating the environmental modification scale it appeared that 17 children received one or more modified equipment or environment concerning the domains of self-care, 13 children received one or more assistive devices in the mobility domain, however, only 2 children got some modified equipment in the social function domain.

The families scored in the FFSS scale in most of the items above 5 indicating that the strengths were rather high. They had lowest scores on the items of internal coping (promoting positive functioning in the family) and positivism (ability to see crises and problems as opportunity to learn). A final analysis investigated the family strengths in the earlier mentioned 2nd, 3rd and 4th quadrants. No differences in FFSS total score were found between the families in the 2nd and 4th quadrants (independently functioning children with a DQ in the normal range against the discrepant group with a DQ in the normal range). However, the families in the 3rd quadrant (children with additional cognitive difficulties) differed from the families in the 2nd and 4th quadrants (one-way ANOVA, $F(2, 18) = 4.2$, $p < .05$) concerning the subscale measuring 'time allocation' ($t = -2.69$, $df = 12$, $p < .05$; $t = -2.08$, $df = 13$, $p < .05$, respectively). This indicates that multiple restrictions are taxing family strengths, more specifically 'allocation of time'. Finally, it was investigated to what extent families with a child having multiple participation restrictions (3rd quadrant) were spending extra time in therapy, thus having less time for family activities. It was shown by visual inspection that the families in the 2nd and 3rd quadrant did not differ in terms of time spent in therapy, whereas children in the 4th quadrant (discrepant group) received less therapies relative to the others.

In sum, this study has been carried out from the adaptive family perspective. Here, the question is no longer one of listing the unfortunate consequences of an unquestioned tragedy, nor is it a simple counterbalancing quest for purely positive outcomes but the focus concerns factors that may contribute to the successful adaptation of families (Ferguson, 2002). The finding that a DQ in a normal

range can camouflage childrens' needs for assistance is of importance and we believe that this information may lead to some improvements in current clinical practice.

Study III

Pirila, S., van der Meere, J.J., Pentikainen, T., Ruusu-Niemi, P., Korpela, R., Kilpinen, J., & Nieminen, P. Language and motor speech skills in children with cerebral palsy. Manuscript submitted for publication.

The aim of the study was to investigate associations between the severity of cerebral palsy (expressed in terms of functional limitations in gross motor (ARS) and the fine motor domains (i.e. functionality of upper extremities)), cognitive difficulties, language, and motor speech problems in children with cerebral palsy (CP). Also the predictive power of neonatal cranial ultrasound (US) findings on later outcome was investigated. For this purpose, 36 children (age range 1 year 10 months to 9 years 0 months) with CP due to premature birth and white matter damage (periventricular leukomalacia, PVL) participated in this study. It was hypothesized that the severity of the CP in terms of the gross and fine motor limitations is primarily expressed in motor speech skills, whereas CP with comorbid cognitive difficulties will produce additional language deficits in the expressive and comprehensive domains.

Table 4 shows total communicative skills (sum score of expressive and comprehensive language skills), total motor speech skills (sum score of oral motor patterns/structure and phonology/articulation), and use of augmentative and alternative communication (AAC) means and speech therapy as a function of the severity of the CP in combination with the IQ/DQ index.

	IQ/DQ < 70, n = 14			IQ/DQ > 70, n = 22		
Severity of CP	Mild	Moderate	Severe	Mild	Moderate	Severe
Language skills						
Total communicative skills						
Normal	-	1	-	10	6	3
Immature	-	4	4	-	3	-
Deviant	-	1	4	-	-	-
Motor speech skills						
Total motor speech skills						
Normal	-	4	-	6	4	1
Immature	-	1	2	2	5	2
Deviant	-	1	6	2	-	-
Use of AAC means						
No	-	5	1	10	9	3
Yes	-	1	7	-	-	-
Use of speech therapy:						
No	-	5	2	8	8	3
Yes	-	1	6	2	1	-

Table 4 Language and motor speech skills as function of the severity of the CP and the IQ/DQ.

The severity of CP was related to communicative and motor speech problems. The number of cases with normal total communicative skills and normal total motor speech skills were significantly less than the number of cases with immature or deviant communicative or motor speech skills in children with severe motor limitations in contrast to children with mild or moderate motor limitations (Chi-square = 13.32, $p < .01$, Chi-square = 7.01, $p < .05$, respectively). When taking the IQ/DQ index into account, it appeared that the children with an IQ/DQ index above 70 differed in their total communicative skills from the children with an IQ/DQ index below 70 (Chi-square = 20.07, $p < .01$). The two groups did not differ in their total motor speech skills (Chi-square = 0.81, n.s.) indicating, that the motor speech problems were equally distributed. In addition, primarily children with IQ/DQ index below 70 used some AAC means (Chi-square = 13.11, $p < .005$) and received speech therapy more frequently (Chi-square = 3.94, $p < .05$).

A series of multiple regression analyses with Enter method were carried out to estimate the contribution of the predictor variables (US, birth weight and SES) to the severity of CP, IQ/DQ level, language and speech skills. Table 5 shows that in terms of determination of the amount of explained variation (R^2), the factors accounted for 43 % of the variation in the severity of the CP indexed by functional gross and fine motor limitations ($F (3, 32) = 8.2$, $p < .000$). This analysis did not

produce a significant result with respect to IQ/DQ ($F (3, 32) = 1.6$, $p < .21$), language ($F (3, 32) = 1.5$, $p < .24$) and motor speech domains ($F (3, 32) = 0.8$, $p < .51$)

Variables included in the model	Model F (df)	R^2	$^{Adj}R^2$	Beta	SE
Dependent					
Severity of CP	8.2 (3, 32)	.43	.38		1.0
Predictors					
US				.66	
Birth weight				.14	
SES				-.12	

Table 5 Prediction of severity of CP (gross and fine motor limitations) from the variables of neonatal cranial US, birth weight and SES.

As expected, both the severity of the CP and cognitive level were associated with motor speech and language problems. Half of the children with an intelligence level at or close to norms showed impairments primarily in the motor speech skills, whereas CP with comorbid cognitive difficulties produced additional language deficits also in the expressive and comprehensive domains. In addition, US methodology predicted moderately the severity of CP, not the intelligence, language and motor speech domains. It is carefully proposed that US methodology offers little, if any, prognostic information about the development of higher cognitive functions.

Study IV

Pirila, S., van der Meere, J.J., Korhonen, P., Ruusu-Niemi, P., Kyntaja, M., Nieminen, P., & Korpela, R. (2004). A retrospective neurocognitive study in children with spastic diplegia. *Developmental Neuropsychology, Vol 26 (3), 679-691*.

The aim of the study was to investigate the IQ level and the neurocognitive profile of children suffering from spastic diplegia and their relation between brain abnormalities according to neonatal cranial ultrasonography (US). Fifteen children were selected out of a total sample of 27 children suffering from spastic diplegia in the age range of 5 to 12 years living in the region of Tampere University Hospital.

The children's IQ scores were, as a group, at the lower end of the normal distribution (FIQ: M = 81, range = 64 to 107, SD = 12). The children showed relatively intact verbal abilities (VIQ:

M = 97, range = 76 to 125, SD = 14) compared to the performance IQ (PIQ: M = 65, range = 36 to 93, SD = 17). The discrepancy between the verbal and performance IQ was significant (Wilcoxon Signed Ranks Test, Z = -3.41, p < .001). The neuropsychological scores of the NEPSY are depicted in Figure 6.

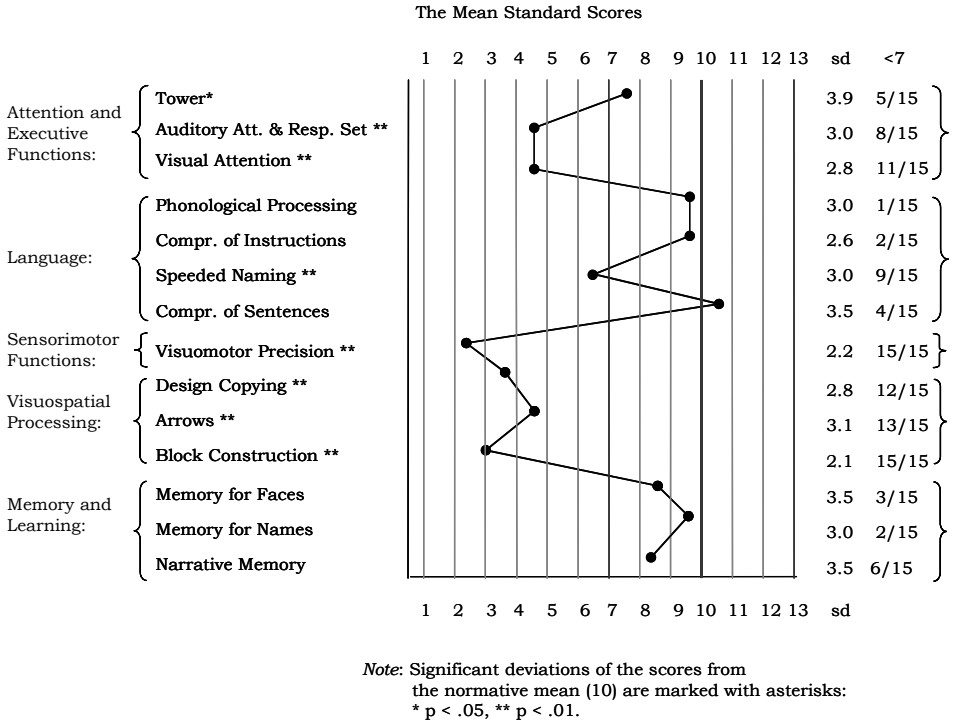

Figure 6 Mean scores of the 14 subtests of the NEPSY in children with spastic diplegia.

Figure 6 shows that only 3 of the 5 neuropsychological domains were affected. As expected, clear deficits were found within the Sensorimotor functions and Visuospatial processing but Language and Memory or Learning functions were at or very close to norms, with the exception of the speeded naming task. In addition, deficits were found in the Attention and Executive Function domain, including tower test and visual attention and auditory attention tasks.

Several statistical analyses were carried out to investigate whether functional impairments of the lower and upper extremities or visual limitations were relevant for the performance on the NEPSY. The ARS scale (tapping the severity of the impairment of the gross motor functions) correlated only with the block construction subtest of the NEPSY (Spearman rho = -.52, p < .05), whereas the functionality of the upper extremities did not correlate significantly with any of the subtest scores of the NEPSY. A series of Mann-Whitney U tests showed that children with visual limitations (n = 10) scored significantly lower than children without such difficulties (n = 5) on the subtests on

visual attention ($p < .01$) and design copying ($p < .05$).

No association was found between the neonatal cranial US findings and the IQ and neurocognitive scores. However, the cranial US findings moderately predicted functional motor limitations (ARS) of the children.

5. DISCUSSION

The purpose of the thesis was to investigate children with functional motor limitations. An effort has been made to integrate the studies into a current structure of International Classification of Functioning, Disability and Health (ICF; WHO, 2001). The interaction of developmental characteristics and disability among children represent special challenges for classification as well as measurement. Viewed within the framework of the ICF, the dimensions covered include variables concerning family, children's activity limitations in daily living and clinical assessment of higher cognitive functions. In addition, an effort was made to get some grip on structure-function relationship.

5.1. Environmental level variables concerning family and their relations to factors of child's activity limitations and cognitive functions

Although family strengths are generally recognized as one set of resources for promoting and enhancing family functioning, it is surprising how little is known about identifying and assessing families' functioning styles. In study I, an attempt has been made to explore family strengths and to link them with characteristics of the impairments of the children. More specifically, the main question of study I was to what extent the degree of functional motor limitations is associated with the strengths of the concerned families. For this purpose, we evaluated in 60 families, the severity of the activity limitations and cognitive abilities in the children in tandem with the family strengths.

It appeared that cognitive abilities and the severity of the functional motor limitations of the children were not, or only marginally, associated with family strengths. At the matter of fact, families demonstrated that their strengths were intact at many levels. However, raising a child with comorbid cognitive difficulties loaded on the strengths, i.e. the more restricted the functionality of the children, (1) the less time was spent to do things together, no matter how formal or informal the activity or event, (2) the less was the sense of congruence among members regarding the importance of assigning time and energy to meet needs, and (3) the less varied was the repertoire of internal coping that encourages positive functioning in dealing with both normative and non-

normative life events. In addition, families with children having the most severe participation restrictions scored lower on the sum scores of the categories Family-identity and Coping/resource mobilization.

In the same study, family strengths were evaluated in a developmental perspective because phases of family development may shape how family members perceive a specific source of potential stress. It appeared that factors such as age of the child and primary caretakers were not related to the family strengths, as was also the case with respect to the social-economical status. This finding is in contrast with the study of Lin (2000) who showed that family coping with CP differed in family life cycle stages. Specifically, in Lin's study, families with school-aged children appeared to have better family adaptation than families with adolescents and families with young adults. The age range of our sample was probably too limited to produce similar results.

One could argue that the families who participated represented a bias, that is to say, families participated in the earlier mentioned Play Project (see method section). Participation could have broadened the scope of the concerned families and may have increased their power and positivism to raise their child. However, data were gathered at the start of the Play Project (before the intervention took place). But of more importance that the current findings are not likely to be explained (at least partly) by a positive effect of the intervention on family strengths is that the current findings are in concert with research outcome of Dunst and Trivette (1993). On the basis of five samples (with a sample size ranging from 51 to 129) consisting out of children with an IQ below 70, physically impaired children and children at-risk for poor developmental outcome, they concluded that worst family functioning was found in households of physically impaired children with cognitive difficulties. Consequently, the Dunst and Trivette study and the present study demonstrate that the identification of strengths and weaknesses in family functioning is of importance in order to provide necessary support for the caregivers to carry out child level interventions.

To the best of our knowledge, the study II is the first that integrates the Social Systems Model of family functioning by Dunst, Trivette and Deal (1988), including the central concept of family strengths, with the PEDI instrument which has closely oriented its content based on the ICIDH (WHO, 1980). Along these lines, functional skills and extent to which assistance was required from caretakers and how it was taxing family strengths were measured in 21 children with activity limitations in the age range 2 years 10 months to 6 years 2 months. However, given the small sample size, the study should be considered a pilot study which at best provides some hints in directing future research with larger samples. For this purpose, correlations between the variables of interest were computed that, by definition, prevent claims about causality. In addition, it must be underlined that

direct measures of family functioning, direct observations of caretakers' interactions, and children's behavior are needed that would enhance our understanding of the complicated transactions between children with participation restrictions and their environment. Consequently, potential insightful information could be yielded from the current data.

Findings indicated that the children obtained low normative standard scores in all three domains of self-care, mobility, and social function of the PEDI and, consequently, needed and received an extensive amount of assistance from the caregivers, except for the social function domain. Here, the sample did not differ from the normative mean in the amount of assistance provided by the caregivers. This was the case in spite of the fact that the majority of the children (15 out of 21) were assessed as having functional limitations in this domain. This discrepancy could be explained neither by the age of the children and the mothers nor the social-economical background of the families. We carefully suggested that parents might have had difficulties in noticing needs for assistance in the social function domain, especially when children had a cognitive level at or close to norms. The families with a child having additional cognitive difficulties provided them the amount of assistance the children needed in the domains of self-care, mobility and social function. These families differed in the amount of 'allocation of time for family members to do things together' from other families investigated.

Again, as was the case in the study I, one could argue that the families who participated represented a biased group of parents because of their participation in the Play Project. In contrast to study I, in study II, data were gathered at the end of the Play Project. Hence, results may partly reflect beneficial effects of this intervention. However, the main finding of study II was that normal DQ level could have camouflaged the needs in the social domain. This finding is difficult to be explained in terms of a bias of the parents.

Although the study II was not mainly focused on the issue of assistive devices in daily use, it was a remarkable finding that assistive devices were frequently used for mobility and self-care, but seldom to support social function. This finding has been confirmed by others investigating larger samples of children with activity limitations (e.g. Korpela, Seppanen & Koivikko, 1992; Ostensio, Brogen Carlberg & Vollestad, 2003). It is well recognized that the prescription of technical aids for children is complex because age, physical, psychosocial and environmental needs must be taken into account (Korpela, Seppanen & Koivikko, 1992). It is obvious that more systematic and comprehensive research on this topic is urgently needed.

5.2. Activity limitations and their relations to higher cognitive functions and brain impairment

As expected in the study III, the severity of CP, expressed in terms of functional gross and fine motor limitations, together with the intelligence factor, were related with language and motor speech problems. More specifically, children with an intelligence level at or close to norms showed impairments primarily in the motor speech domain, whereas children with additional cognitive difficulties (DQ/IQ score below 70) showed impairments both in language and motor speech skills. This latter group of children had additional limitations in particular in their communicative skills. It is possible that communicative difficulties have their effects on coping abilities of caregivers as reported by us in the study I and II. The age range of the group of children was between 1 year 10 months and 9 years. To get more grip on the development of the language and speech skills it is obvious that the present findings need a replication using a longitudinal design to pinpoint more precisely the development of communication and pragmatic skills.

One question that emerges is to what extent CP is associated with specific language impairment (SLI). Criteria for SLI are (1) a language age equivalent delay more than one year expected for child's chronological age, and (2) a discrepancy of more than 15 to 20 points between verbal and performance IQ (Haynes & Naidoo, 1991; Stark & Tallal, 1981). The study III demonstrated that none of the children with an IQ/DQ above 70 showed such a language delay, and that in the whole sample only one child showed a significant discrepancy between verbal and performance IQ in favour of the performance skills. Nevertheless, individual children had delays in skill attainment that warranted language intervention. Within this perspective, it was an interesting finding, as reported in study III, that children with an intelligence level at or near to norms but with motor speech problems received less often speech therapeutic intervention or used augmentative and alternative communication means in order to enhance their communication and pragmatic skills than children with additional cognitive difficulties. This finding may mirror the tendency of parents and, also, professionals to overlook social needs of children with motor restrictions without learning difficulties as reported by us in the study II.

The aim of the study IV was to investigate the IQ level and neurocognitive profile of children suffering from spastic diplegia and their relationship with brain abnormality according to neonatal cranial ultrasonography. The children formed a relatively homogeneous group with respect to cranial US: all had periventricular findings and posterior cortical abnormality, some unilateral and some bilateral. The findings were as follows:

First, children scored at the lower end of the normal distribution of intellectual functioning with relatively intact verbal abilities. Better verbal than nonverbal cognitive functioning in

CP has also been reported by others (Carlsson et al, 1994; Fazzi et al., 1994). As expected, children had low scores on the performance-based IQ scores. This finding was confirmed by the neuropsychological investigation (NEPSY) where clear deficits were noticed in the areas of visuomotor and visuospatial processing, together with relatively intact language and memory/learning functions. These findings fit well with the earlier discussed study of Olsen et al. (1998). The current study and the study by Olsen and colleagues showed cognitive correlates with posterior cortical damage. However, our sample of prematurely born children was more affected than theirs with cystic forms of PVL in parietal, parietal-occipital or frontal-parietal-occipital areas in 9 out of the 15 children. As a result, our sample demonstrated severe problems in the domains of visuomotor and visuospatial processing whereas their sample of premature born children with CP performed poorly in the same domains but in the normal range.

Given the severe motor limitations of the children, one may question to what extent the performance on NEPSY could be explained by impairments of the lower and upper extremities or visual limitations. It appeared that the ARS scale tapping the severity of the impairment of the lower extremities correlated only with the block construction subtest of the NEPSY, whereas the functionality of the upper extremities did not correlate significantly with any of the subtest scores of the NEPSY. In addition, children with visual limitations scored significantly lower than the children without such difficulties on the subtests of visual attention and design copying.

We acknowledge that the findings in the study IV are based on a small sample of children and the results might best be seen as a preliminary and descriptive study with clues about what to look for in new research. As far as our knowledge goes, correlates to posterior cortical damage have so far not been looked for and even less found, with the exception of the study by Olsen et al. (1998), suggesting that posterior cortical findings are associated with deficits on visuospatial and visuomotor tasks in addition to deficits in visual and auditory attention tasks and speeded naming. These latter tests load also on the speed factor of motor processing.

5.3. Structure-function relationship: prognostic value of the neonatal cranial ultrasound concerning the later outcome in functional motor skills and higher cognitive functions

The studies III and IV showed that the neonatal cranial ultrasound grading (mild, moderate, severe) was associated with the severity of the CP expressed by the ARS score and the functionality of upper extremities (study III), and with the functional motor limitations expressed by the ARS score (study IV). The findings confirmed other studies investigating the predictability of neonatal cranial US findings on cerebral palsy (e.g., Rogers et al., 1994; Fujimoto et al., 1994). However, no

significant correlations were found between the ultrasound grading and the IQ scores, neuropsychological functions, language or motor speech skills. These results are in contrast with, for instance, Ringelberg and van der Bor (1993), de Vries, Rademaker, Groenendaal, Eken, van Haastert, et al. (1998), and Biagioni, Bartalena, Boldrini, Pieri, and Cioni (2000) who investigated children when they were younger than 3 years of age, well before more complex cognitive skills could be tested. In the study III, only 6 out of 36 children were younger than 3 years and, in the study IV, children were 5 years or more. Our findings confirm the findings of Costello, Hamilton, Baudin, Townsend, Bradford, et al. (1988) investigating very preterm children at four years of age and are in accord with the remark of Levene (1990) that it is unreasonable to expect early neonatal US findings to predict minor impairments which are only detectable after the age of 5 years or more. To what extent the age factor plays a role in the predictability of the US is an issue that needs more research. However, the topic US methodology and its predictive value are more complicated than only the age factor. Comparisons with other studies are difficult to make because groups under study differ in type of impairment (e.g., premature born children in Costello et al. (1988) and Rademaker, Uiterwaal, Beek, van Haastert, Lieftink, et al. (2005), concepts, and classifications used (i.e. composite scores of motor and cognitive abilities, see, e.g., Costello et al., (1988), Ringelberg et al. (1993) and Fazzi et al, (1994)).

In study III, 26 out of 36 children had bilateral PVL lesions preventing a systematic evaluation of the laterality factor on verbal deficits. The amount of bilateral white-matter damage in our sample was 72 percent and is in congruence with the review of Holling and Leviton (1999) in which they found that approximately 85 percent of infants with echolucencies followed for motor development had bilateral white-matter damage.

5.4. Clinical implications

The ability to recognize family strengths and weaknesses as part of intervention practice is fundamentally important in work with families (Dunst, 2002). It is well recognized that every successful intervention with a family rests as much on the resources of the family as on those of the interventionist. The usefulness of the Family Functioning Style Scale has been demonstrated in showing that families rearing children with severe participation restrictions have less coping strategies available. Consequently, such families deserve a holistic intervention approach.

Parents of children with activity limitations are increasingly considered as experts in the field of care because they have developed a great deal of practical knowledge from their special bond with the child and their long-term experience. Their expertise can deliver an important contribution

towards planning their child's care and education. As a result, there is a shift in services from a professionally dominated medical model towards a model of family-centered services (Dunst, Trivette & Deal, 1988; Korpela & Nieminen, 2001; Leskinen & Juvonen, 1994; Mattus, 2001; Määttä, 1999). It is also reflected in the paradigm shift of disability from one based on a medical framework (i.e. ICIDH) toward one reflecting social dimensions of disability (ICF) in the WHO framework (WHO, 1980, 2001).

As indicated by the FFSS scores, the family strengths were rather high, and, as indicated by the PEDI scores, the functional limitations of the children were linked with the amount of assistance provided by the caregivers. However, the current study indicated that a cognitive performance in a normal range may camouflage children's needs for assistance in the social function domain. In addition, it was of interest that, overall, children received therapies focused mainly on gross motor, fine motor, and oral motor functions. It is important that this information may lead to some improvements in current clinical practice concerning the social domain, play and communication. An example of such improvements was the intervention program for disabled children in the Department of Pediatric Neurology, Tampere University Hospital (Leikkitarkkailu. Käsikirja lapsen toiminnallisen kehityksen arvioimiseksi, 2004; Korpela & Nieminen, 2001; Pirilä, Korpela & Nieminen, 1997). The project aimed at developing rehabilitation services for families and assessing relevant functional domains for children concerning the routines of everyday life, such as mobility, self-care, play, and social cognition (Nieminen, 2004).

The children with cognitive performance at or very close to norms have their own special needs in utilizating therapeutic devices and health care professionals should be more precise in motivating parents to provide assistance and assistive devices in order to diminish children's participation restrictions. Research has shown (Pennington, 1999; Pennington, Goldbart & Marshall, 2004) that children with motor and speech impairments are potentially able to produce more functions of communication (e.g., asking questions, providing clarification when misunderstood) than they are generally given the opportunity to with familiar adult partners. It is suggested that, if needed, social training through means of video-taping, counselling, etc. might help parents to recognize their children's signals and to provide a wider range of opportunities for communication, thus enabling the child to develop his/her pragmatic skills. This suggestion fits well with strengths-based practices and with the ICF framework of a person-environment interaction. The framework is likely to influence and strengthen the extent to which parents actively engage in intervention planning to the degree that parents feel respected, valued and treated as if they are knowledgeable and capable to work towards their goals (Dunst, 1995, 2002; Ferguson, 2002; Korpela & Nieminen, 2001).

The ICF model was used in this thesis as a framework in our effort to depict the complexity of interrelations between functioning and disability. However, as noticed in the Chapter 2.2., there is a paucity of pediatric standardized instruments for the measurement of functional status and the transactions between the child and the environment. Only the PEDI instrument, used in the study II, addresses the domains of Activity level and Participation component proposed in the ICF model. Other pediatric measures, used in the studies I-IV, are designed for clinical identification of the nature and extent of children's developmental needs and belong to the Body functions and structures component of the ICF. The necessity to elaborate assessment measures adapted to everyday functioning of families and children with disabilities is well recognized by professionals working with families in clinical settings. (Korpela & Nieminen, 2001; Nieminen, 2004; Pirilä, Nieminen & Korpela, 1997; Musikka-Siirtola, 2005). An important application of such measures could be that they can contribute to identifying children on the basis of functional profiles, such as play and social abilities (Nieminen, 2004). This approach, underlined by Korpela and Nieminen (2001), Musikka-Siirtola (2005) and Simeonsson (2003), is especially important in learning and rehabilitation, where categorical identification of children could become barriers to intervention. It also calls for multiprofessional assessment and planning of rehabilitation together with families (Korpela & Nieminen, 2001).

The current study indicated that verbal comprehension and expression skills were more sophisticated than motor speech skills in many children with an IQ/DQ index above 70, whereas language and speech skills were at a similar (low) level in many children with an IQ below 70. Intervention planning should take this into consideration so that communication could be matched to the child's level in each area and provided in meaningful contexts.

Whether CP is associated with SLI has still to be seen. Although a considerable body of knowledge about SLI has been derived from clinical studies, the growth of precise knowledge has been hampered by the absence of any agreed criteria for defining language impairment (Haynes & Naidoo, 1991). Usually SLI is defined in terms of exclusion, for instance, language impairment not secondary to emotional disturbance, hearing impairment, or mental subnormality (Stark & Tallal, 1981). About 60 percent of our sample of convenience in the study III consisted out of children with an IQ/DQ index at or close to norms and none of them had severe hearing impairment. None of them fulfilled the criteria of SLI. However, children with additional cognitive difficulties showed language and speech disorders. A direct comparison with, for instance, children with Down syndrome or non-verbal learning disorder is needed to understand more clearly the essence of motor, language and speech problems in children with CP plus additional cognitive difficulties.

Incidence studies using cranial US scanning constitute an important source of information for parents on what they can expect with regard to their child's future outcome. The current studies III and IV showed that the cranial US findings were moderately predictive with regard to functional motor limitations. One may argue that the small number of children who participated in the studies III and IV (n=36 and n = 15, respectively) may have prevented a significant correlation between US abnormalities and cognitive outcome. This argument seems unlikely. The sample sizes were large enough to demonstrate a clear correlation between the US findings and the gross motor impairment. A possible explanation is that cranial US scans are simply not sensitive enough to reveal subtle injuries and that the use of structural magnetic resonance imaging, which provides superior soft tissue contrast and spatial resolution compared to ultrasonography, might be a more appropriate methodology to predict neuropsychological disabilities (Krägeloh-Mann, 2005; Olsen, Vainionpää, Pääkkö, Korkman, Pyhtinen et al., 1998). Thus, it is carefully proposed that US methodology offers little, if any, prognostic information about the development of higher cognitive functions in children with CP.

Finally, clinical assessment methods were used to estimate higher cognitive functions including language and speech abilities. However, such methods often require behavioural repertoires that exceed the capabilities of many children with moderate to severe impairment (e.g., oral answers and object manipulations). Consequently, the outcome of the study III warrants in depth neurophysiologic measures, e.g., Evoked Response Potentials (N400) (Byrne, Dywan & Connolly, 1995), to specify more precisely the anatomy of motor speech and language problems connected with CP.

5.5. Limitations and future research

In the thesis, an effort was made to incorporate developmental perspective into the concept of family adaptation by including age factor into analyses. On some occasions, correlations between child's age and dependent variables were made. However, it must be admitted that age factor does not cover the totality of family life span. A more throughout developmental perspective needs to be fully incorporated into the conceptualization of the child's needs and family functioning in future research since family responsibilities and tasks shift in response to the child's ever-changing needs.

It is acknowledged that the current studies were based on small samples of children and the results might best be seen as preliminary and descriptive studies with clues about what to look for in future research. However, in spite of this shortage, it was possible to demonstrate the linkages between the three levels: the families, the individual activity limitations and the body functions. In

addition, a hospital-based study, like the current one, has the advantage that all children are treated and assessed by the same team of professionals. Consequences of multiprofessional assessment of clinically relevant samples of children are often that sample sizes tend to be small, work is time-consuming - but the evaluations are, at best, less routine and variable.

Little is still known about how a person-environment interactions frame a developmental process of the children with disabilities and their acquisition of increasingly complex skills involving actions and reactions to the physical and social environment. Longitudinal studies are urgently needed in order to investigate more detail the developmental trajectories of activities and participation, such as play, learning and functioning at home, in school and neighbourhood.

Finally, there is a paucity of research concerning the development of neuropsychological functions in children with CP (Kemp, Kirk & Korkman, 2001; Temple, 1997). In this thesis, an effort was made to demonstrate neurocognitive correlates with posterior cortical damage. In addition to the Sensorimotor and Visuospatial functions, the deficits were also found, among others, in the Attention and Executive Function domain. The question emerges what is the essence of the deficits in executive functions in children with CP? Direct comparisons are needed with number of syndromes within which disorders of executive function have been identified (Temple, 1997), such as, children with phenylketonuria, autism, ADHD, Turner's syndrome, or Gilles de la Tourette syndrome. In addition, the neuropsychological findings were presented groupwise. It is obvious that imaging-based brain - behaviour studies and neurophysiologic measures are needed in order to investigate more throughout the interlinking of different deficits and to incorporate the traditions of cognitive neuropsychology and neuroscience into the research field concerning children with cerebral palsy.

Just about finishing the thesis, a new definition of cerebral palsy was proposed by the Executive Committee for the Definition of Cerebral Palsy (Bax, et al., 2005). As a final exercise, the proposed definition will be discussed from what I have learned from my research. For the first, the proposed definition underlines that motor disorders of cerebral palsy are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behavioural, and/or by a seizure disorder. The nucleus of the thesis was the investigation of the comorbid deficits in the cognitive and communicational sphere found in children with motor impairments. The second issue of the proposed definition of the Committee was the emphasis on the functional consequences of different health states delineated in the ICF framework. In accord with this viewpoint, it was noticed in the thesis that research is urgently needed in the sphere of activity limitations of the motor, hand and arm functions, and speech and oromotor difficulties. Third, the Committee underlines that correlations between radiographic findings and the clinical presentation in CP were

weak. In addition, information is insufficient to recommend any specific classification scheme for neuroimaging findings. The correlations, as reported between the ultrasound methodology and the motor and cognitive outcome variables are, at least, partially in line with the claim of the Committee. However, to get more grip on radiographic findings and clinical presentations, neuropsychologists have their role to bridge the gap between these two entities.

TIIVISTELMÄ

Pirilä, Silja (2006). Children with functional motor limitations
A three-level approach

Väitöskirjatutkimukseni aiheena olivat liikuntavammaiset lapset. Tutkimuksen aihetta tarkasteltiin WHO:n vuonna 2001 julkaiseman Toimintakyvyn, toimintarajoitteiden ja terveyden kansainvälisen luokituksen (ICF; International Classification of Functioning, Disability and Health) viitekehyksessä kolmella tasolla: (1) ympäristötekijöiden taso, johon sisällytettiin perheeseen liittyviä muuttujia, kuten perheen voimavarat, sosioekonominen asema, huoltajan ikä ja huoltajan suorittaman avustamisen määrä lapsen päivittäisissä toiminnoissa; (2) suoritusten ja osallistumisen taso, johon sisällytettiin lapsen liikkumiskyky, yläraajojen toiminnallisuus ja päivittäinen toimintakyky itsestä huolehtimisessa, liikkumisessa ja sosiaalisissa toiminnoissa; ja (3) ruumiin/kehon toimintojen ja rakenteiden taso, johon sisällytettiin kehon ja mielen toimintoja kuvaavina muuttujina vastasyntyneisyyskauden aivovaurion vaikeusaste, syntymäpaino, ikä, älyllinen toimintakyky, neuropsykologiset, kielelliset ja puhemotoriset taidot.

Tutkimusaineisto koostui 69 liikuntavammaisesta lapsesta, jotka olivat seurannassa Tampereen yliopistollisen sairaalan lastenneurologisessa yksikössä. Valtaosa aineistosta, 61 lasta, osallistui vuosina 1994 - 1997 vammaisten lasten varhaiskuntoutuksen kehittämistutkimukseen, Leikkiprojektiin (Korpela & Nieminen, 2001). Lapset valittiin varhaiskuntoutusprojektiin satunnaistetulla otannalla kaikista tutkimusvalintakriteerit täyttävistä 0–6 –vuotiaista lapsista, jotka olivat neurologisen vammaisuuden kannalta riskilapsia, ja joille kehittyi liikuntavamma. Tutkimusaineistoa täydennettiin myöhemmin kahdeksalla CP-vammaisella lapsella. Aineiston lisäksi myös mittausmenetelmät ja moniammatillisen työryhmän toteuttama aineistonkeruu olivat pääasiallisesti Leikkiprojektista peräisin.

Tulokset osoittivat, että liikuntavammaisten lasten perheiden voimavarat olivat yleisesti ottaen aika hyvät. Voimavarat olivat kuitenkin keskimääräistä heikommat niissä perheissä, joissa lapsi oli vaikea-asteisesti monivammainen, eli lapsella oli vaikean liikuntavamman lisäksi kehitysvamma. Lapsilla oli monenlaisia toimintakyvyn rajoituksia omatoimisuustaidoissa, liikkumisessa ja sosiaalisissa toiminnoissa. Vanhemmat avustivat heitä erityisesti omatoimisuustaitojen ja liikkumisen rajoituksissa. Näihin vaikeuksiinsa heillä oli myös käytettävissä apuvälineitä. Sen sijaan sosiaalisissa toiminnoissa, joissa puutteet ilmenivät kommunikaation, vuorovaikutuksen, leikin, ja ikätoverisuhteiden alueilla, lapsia avustettiin silloin, kun heillä oli liikuntavamman lisäksi laaja-alaisia kognitiivisia vaikeuksia (ÄO < 70). Niillä liikuntavammaisilla lapsilla, joilla ei ollut kognitiivisen kehityksen laaja-alaisia puutteita, avustaminen oli keskimäärin samansuuruista kuin vastaavanikäisillä vammattomilla lapsilla. CP-vamman vaikeusaste ja älyllinen toimintakyky olivat yhteydessä kielellisiin, puhemotorisiin, puheen tuoton ja ymmärtämisen ongelmiin. Puolella tutkituista CP-vammaisista lapsista, joiden älyllinen kehitys oli normaalirajoissa, oli puhemotoriikan, artikulaation tai fonologian ongelmia. Mikäli liikuntavammaan yhdistyi kehitysvamma, lapsilla oli lisäksi ongelmia puheen ymmärtämisessä ja tuotossa. Alaraajapainotteisesta liikuntavammasta (diplegia spastica) kärsivillä lapsilla oli todettavissa ongelmia sensomotorisissa ja visuospatiaalisissa toiminnoissa, visuaalisessa ja auditiivisessa tarkkaavuudessa sekä nopeassa nimeämisessä. Muuten kielelliset, muistiin ja oppimiskykyyn liittyvät neuropsykologiset toiminnot olivat kehittyneet normaalirajoissa. Vastasyntyneisyyskaudella aivojen ultraäänitutkimuksissa todettu valkean aivoaineen vaurio (periventrikulaarinen leukomalasia) ennusti liikuntavamman vaikeusastetta, mutta se ei ennustanut myöhempää älyllistä toimintakykyä, neuropsykologisia, kielellisiä ymmärtämisen ja tuottamisen tai puhemotoriikan ongelmia.

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