

**To get things done,
the challenge in everyday life for children
with spina bifida**

Quality of performance, autonomy and participation

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To get things done, the challenge in everyday life for children with spina bifida - Quality of performance, autonomy and participation

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Abstract

The overall **aim** of this thesis was to increase knowledge of the quality of performance of everyday activities, autonomy and participation in children with spina bifida (SB) and to explore how they relate to each other.

Methods: In *Study I*, the quality of performance of everyday activities in 50 children with SB (of the 65 in a population-based cohort) aged 6 to 14 years was assessed with the Assessment of Motor and Process Skills (AMPS). Their ability measures were compared with international age norms and with the ability measures from a control group of typically developed Nordic children. In *study II*, the cross-cultural differences in the ADL motor and process ability measured with the AMPS between children from the Nordic countries, (n=2374), and from North America (n= 2239), aged 3-15 years, without known disabilities were analysed using a two-way ANOVA. In *Study III* the autonomy levels of the 50 children with SB were rated both by the children themselves and by their parents. The agreement between the children's and the parents' ratings was analysed, and the relationship between the autonomy levels and the child's age, motor and process ability measures from the AMPS assessment in study I was analysed with binary logistic regression. *Study IV:* The frequency of participation in school-related activities in the 50 children with SB was rated both by the children themselves and by their teachers (in 48/50 cases). The teachers also rated the children's level of active participation using the School Function Assessment (SFA). The relationship between the children's level of active participation and their motor and process ability measures was analysed with binary logistic regression.

Results. The majority of the children had difficulties performing well-known everyday activities in an effortless, efficient, safe and independent way, demonstrated by low ADL motor- and process ability measures. This deficient quality of task performance, in particular the process skills, was strongly related to both their level of autonomy in daily life and their level of active participation in school. The children with SB had low autonomy levels in goal-directed situations that needed personal initiation. The agreement between parents' and children's ratings of the children's autonomy level was low. The frequency of participation among the children was high in school activities, although their teachers rated their active participation as restricted. The results also showed that the age norm in the AMPS is valid for use in a Nordic context.

Conclusions: This thesis demonstrates that children with SB have difficulties getting things done due to deficient quality of task performance. It is therefore crucial for Occupational Therapists to assess, understand and support the development of the performance skills in children with SB, in order to enhance their autonomy and active participation in everyday life, school and society.

Keywords: spina bifida, myelomeningocele, lipo-myelomeningocele, autonomy, participation, performance skills, Assessment of Motor and Process Skills (AMPS), School Function Assessment (SFA)

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- I. Marie Peny-Dahlstrand, Anne-Christine Åhlander, Lena Krumlinde-Sundholm, Gunilla Gosman-Hedström
Quality of performance of everyday activities in children with spina bifida: a population-based study.
Acta Paediatr 2009;98:1674-1679
- II. Marie Peny-Dahlstrand, Gunilla Gosman-Hedström, Lena Krumlinde-Sundholm,
Are there cross-cultural differences of ADL ability in children measured with the Assessment of Motor and Process Skills (AMPS)?
Scand J Occup Ther 2011; Feb 25 [epub ahead of print]
- III. Marie Peny-Dahlstrand, Lena Krumlinde-Sundholm, Gunilla Gosman-Hedström
Is autonomy related to the quality of performance of everyday activities in children with spina bifida?
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- IV. Marie Peny-Dahlstrand, Lena Krumlinde-Sundholm, Gunilla Gosman-Hedström
Patterns of participation in school-related activities and settings in children with spina bifida; a population based study.
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ABBREVIATIONS

ADL	Activities of Daily Living
AMPS	Assessment of Motor and Process Skills
HC+	Shunt-treated hydrocephalus
HC-	Non hydrocephalus
ICF	International Classification of Functioning and Health
ICF- CY	International Classification of Functioning and Health. Children and Youth
IMOD	Interactional Model of Occupational Development
Lipo-MMC	Lipo-myelomeningocele
MMC	Myelomeningocele
OT	Occupational Therapist
PA	Percentage Agreement
PEDI	Pediatric Evaluation of Disability Inventory
PEO	Person-Environment-Occupational Model
PTO	Process Transforming Occupation
SFA	School Function Assessment
SB	Spina bifida

PREFACE

As an occupational therapist, I have worked in paediatric rehabilitation, (in Sweden called “habilitation”) for over three decades. The primary goal for an occupational therapist in habilitation is to enable the child to do, i.e. to perform, and to be engaged in the occupations in life he/she must, wants to, and needs to do. Even though, or maybe due to the fact that I have worked with children with different kinds of disabling conditions all these years, the group of children born with spina bifida (SB) have puzzled and challenged me professionally for a long time. How could it be explained that parents of children born with SB so often expressed phrases like – *“He can do it, it is just never done”* when I asked them about their child’s level of independence in daily life? And why didn’t the children or adolescents really benefit from all the training in self-care they have had? When compared with other groups of children with physical disabilities, children with SB seem to have relatively good prerequisites, they are often very verbal, they are mostly in mainstream schools, they handle their wheelchairs so competently yet, when growing up, they often seem to be participating less and less in society. This was the “clinical irritation” that led me to this research, facing the gaps in knowledge concerning the children born with SB.

INTRODUCTION

The primary focus of this thesis is the doing in everyday life and participation of children with spina bifida. The thesis has three starting points (not in hierarchical order) representing three perspectives that I will try to merge.

The first perspective is the importance of occupation and the effect it has on health and development [1-3]. Occupation has been conceptualised as a “*synthesis of doing, being and becoming*” [4 p 2]. Doing is the child’s means of connecting with the social world [4, 5], and through doing he/she gains not only skills but also self identity [5]. Doing leads to being, which is seen as the inner life, discovery of oneself, enjoyment and existence [4]. To become what one has the potential and willingness to become depends on both doing and being [4].

The second perspective is that neurological disabilities with early onset should be viewed as developmental disabilities; or, as stated by Peter Rosenbaum, “*neurodevelopmental disabilities are conditions involving the developing nervous system that have or are likely to have trajectories of that child’s development*” [6 p. 251].

The third perspective is that it is through an interactive and dynamic relationship between the person, the occupation and contextual factors that participation in occupations is enabled. In occupational therapy this is described in many models [7-11]. A similar integrative/dynamic perspective forms the basis for the International Classification of Functioning and Health (ICF) published by WHO [12]. In the ICF, health, body functions, activity and participation and contextual factors are described in different life situations, and the interaction between them is dynamic and works in any direction [12]. One aim of the ICF is to be a common language for all health professions [13], and the classification is widely used as a conceptual framework in habilitation in Sweden [14].

CURRENT KNOWLEDGE OF SPINA BIFIDA

Spina bifida is an umbrella term for congenital defects of the spinal cord due to incomplete closure of the neural tube [15]. There are different forms of spina bifida, spina bifida occulta, which is a bony defect covered with skin, meningocele (herniation covered

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by normal skin) and myelomeningocele (MMC), the most common and severe type of spina bifida, is a herniation of spinal cord and nerves, usually an open defect [15]. A fourth form is the Lipo-myelomeningocele (Lipo-MMC) that is a form in which subcutaneous fatty mass infiltrates the spinal canal. In this thesis, the term spina bifida is used for MMC and Lipo-MMMC.

The prevalence of SB in Sweden has decreased during the last 20 years, and is since 1999 down to approximately 2 children in 10 000 births [16]. Spina bifida is a complex condition and its genesis is still not totally explained. Folic acid has been identified as an important contributing factor, but to date it is not clear to what extent [17]. One recent discovery is that it is probably a combination of folic acid, genetic factors and environmental factors that interfere with each other, but no particulate gene has been identified yet [17]. The fact that folic acid has been recommended to women of fertile age and in some countries even added to food has not made spina bifida disappear. A combination of in vitro diagnostics and counselling is probably a more important factor in decreasing the number of children born with SB [17].

It is not until now, in the beginning of the 21st century, that the first large generation of children with SB has reached adulthood, as few of the children born with SB before the 1960s survived until adulthood [18]. In the 1960s shunt treatment for the hydrocephalus became more advanced [19], and new treatment to solve problems with bladder incontinence was introduced, clean intermittent catheterisation, which decreased mortality rates due to renal dysfunction considerably [19-21]. Spina bifida has earlier primarily been described as a condition that leads to different degrees of motor and sensory impairments [22], hydrocephalus and neurogenic bladder and bowel dysfunctions [23], all problems that can be related to lesion level. Contemporary research has now also demonstrated that the developing brain is often more affected in children with SB than previously understood [24]. The neuropathology of the brain has proved to be so complex that hydrocephalus cannot be seen as the only explanation of the cognitive profile [25], and that other brain malformations as in the cortex, corpus callosum, cerebellum and the brainstem are very common [24, 26]. The Chiari type II malformation, which is a herniation of the hindbrain, is now understood to be present in all cases of MMC [26].

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Neuropsychologists have strongly argued that, as a result of this commonly seen reorganisation of the brain, children/individuals with SB show a behavioural pattern that can be regarded as a “cognitive phenotype” [27, 28]. This cognitive phenotype has been described as being process-specific but not domain-specific. This is to say that children with SB should not be categorised as having “perceptual deficits”, “executive deficits”, or even “motor deficits” in the sense of having general/global problems in those domains [27, 29]. On the contrary, while they often have deficits in each domain of functioning, they also have assets. Difficulties are seen with assembled processing, i.e. with all situations that involve something to be “*on-line iterative cycles of activations, disengagement, and integration*” [27 p.5], which means having deficiencies in the ability to think, feed forward and initiate new performance on their own. But they are often relatively intact in everything that is guided, and that can be learned by association and categorisation [28]. This is seen in all domains according to Dennis and Barnes [27]. In the domain of executive functions, for example, they have problems in planning and organization [29] and in disengaging attention but assets in sustained attention [27, 30]. Whereas in the domain of motor function, where problems are well documented [22, 31], children with SB seem to do well in motor learning that can be based on error-learning but have problems with predictive movements [32].

On the activity and participation level, it has been reported that the timetable for when children and adolescents with SB achieve milestones of independence (for example, when they are independent in planning activities with peers, or when they saves money) is delayed compared to age peers without disability [33]. Teenagers with SB participate less than their peers in activities that are common for adolescents and in such activities that could prepare them for adult life [34]. The dependency in self-care and restricted participation have generally been treated as secondary problems in spina bifida [35, 36]. They have been explained as secondary due to motor functional problems [37], or to environmental factors as low accessibility [36], or as consequence of parental styling (e.g. overprotective parents) [38-41]. In many studies, the severity of the spina bifida in itself (lesion level and hydrocephalus) has been given as the explanation for the low participation level [35]. Adolescents with SB have themselves reported that low

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motivation is the most important factor that prevents them from participating in sport and leisure activities [42, 43].

Long-time surveys of cohorts of children/adolescents with SB into adulthood have shown that individuals born with SB have low rates of independence even in their lives as adults [18, 44-46]. At the age of 30-35 years, two thirds of the adults were still not independent in daily living [18], were in addition often unemployed [47], and their participation in social life and leisure activities was restricted [48].

It has been stressed that there is an urgent need for research aimed at finding ways to enhance functional independence for individuals born with SB [49]. To do that and to understand the impact of cognitive skills on activity and participation, new methods need to be developed [29]. As described above, with reference to the ICF framework, there exists a large body of knowledge about the functional outcome of spina bifida on the body functions and structure level, and how they are connected. On the activity and participation level, there is evidence and extensive knowledge about what the child does or does not do, and at what age [33, 50]. However, no study has been found that describes **how** children with SB actually “do”, i.e. perform those activities. Participation on the society level has also been found to be restricted in children as in adults with SB [48], but on this level the traces from the cognitive phenotype are not easily detected. Only very recently has this been studied in relation to independence and quality of life in adult years [51, 52]. Thus, there has been a lack of knowledge about **how** activities are performed, and no study appeared to exist about how the ability “to do” influences participation. The question that started the rationale for this thesis was; as we now know that children with SB have difficulties in all processes and feed-forward-dependent parts of cognitive functions, how come this is so scarcely studied and discussed on activity and participation levels?

CONCEPTUAL CONSIDERATIONS

In order to be able to define a research perspective and to understand how to operationalise, measure and explore the ability to participate in society in children with SB, it was necessary to look into the concepts of doing/occupation, participation and development more deeply.

Doing

Doing from an occupational perspective

In occupational therapy, occupation is the core concept [53] in the sense of doing and being engaged in something that is meaningful, important and needed for the individual concerned [10, 11]. The word occupation comes from the Latin word “occupare” and has been defined as “*to occupy time and space*” [8 p. 2]. Occupation contains both the performance aspect of doing and the contemplative aspect of experience as in being. Through both doing and being, the person becomes what he wants and has the potential to become [4].

The doing part of occupation is often referred to as the performance [54]. A person can either do something in the physical/direct sense or influence the doing by taking the initiative, thereby directing someone else so it will be done for him/her. But in both cases the performer has to take some form of action. In every specific occupation, performance is influenced by the interaction between the person doing something, the environment in which it takes place and what will be performed [7].

How is occupation performed? Most models of occupation end up with a hierarchical description of how different levels of doing: occupation, activities, tasks, and actions, are linked to each other. Both Fischer [55] and Kielhofer [11] explain this hierarchy when defining three related concepts: occupations, occupational performance and performance skills. Within the Occupational Science tradition, Polatajko and colleagues [53], has been working on a taxonomy that could unify occupational therapy language; The Taxonomic Code for Occupational Performance (TCOP). This taxonomy makes clear distinctions between the occupation as a whole in which a person engages and the subset of the occupation in an hierarchical way starting with defining occupations as a “*a set of activities that is performed with some consistency and regulation that brings structure and is given values and meaning by individuals and a culture*” [53 p.19]. Activities in turn are the outcome of a set of tasks with a defend endpoint and a task is accomplished by a series of actions. According to Fischer [55-56] and Kielhofner [11], the skills to carry out those actions can be termed *performance skills*. On the other hand, not many models of occupation describe the actual process of doing something in a general sense, one exception being Fisher [57] who describes the overall task

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performance as the actual carrying out of a task in the environment, via initiation of all different steps that the task consists of, to the endpoint, i.e. getting the task done.

According to Fischer [57], there are three types of performance skills; motor skills, process skills and social interactive skills. The performance skills depend on the person's motor, cognitive and communicative functions, but they are not these functions *per se*. Performance skills are the way that the body functions are used and organised into actions in task performance [57].

Doing or execution from a neuropsychological perspective

In psychology doing is equivalent to behaviour or adapted behaviour [58]. When we do something, there is one part that is observable and physical, but how and when we do it is directed by the executive functions. Goldberg describes [59] how an individual, for every goal-directed behaviour, has to *execute* a chain of steps. The first step is to identify a goal and a purpose, and the behaviour has to be initiated. The next step is to create or identify a plan of how each part is going to be done and in a proper timely order. The third step is the actual observable performance of the plan: the enacting. To succeed, all actions have to be carried out in the right order and without any hesitation between them. If the result is found to be not as one wanted, new plans/strategies have to be identified, and the process starts all over again [59].

Doing in the cultural historical perspective (action theory)

Lentontjev, being a Russian neuropsychologist and working in the 1950s in the group of psychologists with Luria and Vygotsky, has presented a model of doing: "The action theory." This model is hierarchical and examines human processes from the perspective of different levels of analysis [60, 61]. The first level is the level of the intention, the motives, goals and needs that drive the activity. The second is the level of actions and their associated goals, which have to be performed in correct order to reach the main goal/motives at the first level. Level three is the level of operations. The operations are actions that have become so learned that they are performed automatically and, as such, serve as means of achieving the higher-order goals. Finally, the fourth level is the one of body functions needed to learn an operation or action [60, 62].

Definition of doing in this thesis

In this thesis, doing is defined as one part of occupational engagement. The individual engages in occupations and performs the activities and tasks that are imbedded therein by carrying out the actions (i.e. using their performance skills) needed to complete the tasks. Doing something can be understood both in an abstract, general sense and in a concrete, specific sense.

The general sense is the process of undertaking a whole task (i.e any task). This process starts with an idea or motive that might be invisible to others. To perform it, the individual has to plan, initiate, enact and adjust every step of the task in a timely order until the task is done. The capacity to carry out this process varies from individual to individual.

The outcome of all specific activities can and will vary for different reasons and can be explained in many different ways according to the Person-Environment-Occupational Model (PEO) [7] or other models of occupation [10, 11]. In this thesis, the doing of each specific activity is seen as a complex dynamic interaction between the person, the environment and the task performed. All the parts influence each other and the final outcome in a specific task, and one is not possible without the other. This implies that the capacity to do something in a general sense described above is one of the personal factors in the interaction of a specific activity.

Participation

Since WHO introduced the concept of participation in the first revision of the International Classification of Impairment Disability and Handicap (ICIDH Beta -2) in the 1990s and with the publication of the International Classification of Functioning and Health (ICF) [12] in 2001 and a complementary children and youth version (ICF-CY) [63], this concept has had a tremendous impact on the language and thinking in the habilitation services for children [14, 64]. In the habilitation context, participation should be considered not only a goal but also the process of reaching a goal [64]. To participate in activities together with others is believed to be the key to development [65] and is closely related to health [12] and quality of life [66, 67]. But how is the concept of participation to be understood? Participation is a complex and broad concept, and it is

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therefore defined in many different manners [64, 68-70]. There is not only one interpretation of participation in the literature. The word originates from the Latin word "participare", which has two meanings; *take part in* and *share* [71, 72]. But from there to define the concept so it can be operationalised is the subject of ongoing debate [70, 71, 73, 74].

In the ICF classification [12], participation is introduced as a positive term instead of "handicap", which was used in the earlier versions (ICIDH 1980) [13, 75]. Participation is now defined as involvement in life situations and connected with activity, which in turn is defined as the execution of a task or an action by an individual. The ICF describes environmental and personal factors as ones that enable, or hinder, participation. This definition of participation has been criticised by many authors [68-70, 76]. They argue that a clearer fit is needed between a component of acting, the personal will and motivation (personal factors) and sharing in togetherness and the sense of belonging of the individual [66, 68, 69]. Questions have also been raised whether it is all about a personal perspective or if a person from the outside can judge participation in others [68, 77-79]. The ICF has also been criticised for not making a clear distinction between activity and participation [70]. Nor does it explain how the variation in complexity of the life domains it describes is to be understood [71]. Coster and Kheteni [71] point out that it is clear that some domains are to be seen as prerequisite for being able to accomplish the more complex ones (for example the life domains of *mobility* or *general tasks and demands* are needed in a complex one such as *domestic life*).

In most models of the concept of participation, both **internal and external** factors have been described as influencing participation [63, 80-83]. Internal factors are described as the abilities or capacities the person possesses [12, 76] and his/her motivation and personal will [76, 83-85]. It has been argued that the diagnosis is not a determining factor in participation [80], but that functional skills and level of autonomy are [80, 82, 86, 87]. External conditions that are often described as influencing participation are opportunities [76], access [88], coaching, adequate support and legislations [63] and social attitudes [63]. Participation in school is also influenced by educational models and teaching styles in the school context [89]. To summarise the different models described above, participation is enabled not only by having access and

opportunities but also by the capacity to involve oneself and to grasp opportunities. The concept of participation is very complex and unfolds like a Russian doll [90] into more related concepts.

Unfolding participation; related concepts

Involvement

To participate is to be *involved* in something [12, 70, 77]. This *something* is mostly described as an activity of some sort [12]. According to this, involvement might be the key to the whole concept of participation as it is the actual interface between the person and the environment [63]. The word involvement also has Latin roots: “involvere”, to *roll up or take in, and include oneself* [91]. The involvement does not need to be physical involvement [77] but still some action needs to be taken to involve oneself and contribute or decide something even in a psychological sense. This way of seeing involvement makes it clear that involvement demands some sort of action from the person, which in turn is dependent on autonomy [92].

Autonomy

Autonomy has been described as the most important prerequisite or personal factor for participation [82, 92-94]. Autonomy is not a single skill, but a state of behaviour or personal characteristic involving: to act from one’s own will and a personal endorsement of the actions taken [95, 96]. The concept of autonomy can thus be explained as being your own person. Autonomy develops gradually in the family context during childhood [97, 98]. Being autonomous puts high demands on executive functions. Ylvisaker [96] believes that autonomy **is** executive skills, or one could say that autonomy is the outcome of executive functions [96]. Cardol [93] has stated that there are two dimensions of autonomy: decision-making autonomy and executive autonomy. So autonomy as a prerequisite for participation can be explained as the capacity to involve oneself [93, 94].

Definition of participation and autonomy in this thesis

In this thesis, participation is defined as being involved in a life situation on the societal level with others in the activities occurring in the settings of different life-domains.

Autonomy, on the other hand, is defined as carrying out something by oneself on one's own initiative, **or** making decisions about what should happen.

Development

Theories of typical child development

The child is a “moving target” in the sense that the child develops rapidly in all aspects from birth to adulthood. The definition of what development is and how it is accomplished has been debated since ancient times [99]. Discussions about whether the development of the child is a result of nature or nurture can be traced back to Plato and Aristotle [9, 99]. Plato believed that the child was born with inner knowledge, but Aristotle thought that all knowledge was derived from experience and learning. Over the centuries ideas about what child development is have followed the philosophical and historical ideas of their time, but the debate has often been kept between the two poles of inner or trained knowledge [99], even though they have had various expressions in different epochs.

At the beginning of the 21st century, most developmental theories accept the idea that development is due to the interaction between genes/nature and environment/nurture [9, 100]. The question in focus now is how this interaction functions, i.e. how the child goes “*from here to there*” [100 p.26]. The active child has become a new theme [101] in the sense that the child is to be seen as an active agent in creating its own environment [102]. Scarr [103] argues that the child creates its own environment, and that a supportive parent is needed, but in the case of “normal” or good enough parenthood, the parents do not cause problems in their children's cognitive outcome. Another change in recent time is that learning and development are seen, once again, to act together [100]. According to Vygotsky [104], children normally learn to perform by taking part in activities that are slightly too difficult for them together with a more experienced person as an adult or a peer. The span between what they can do themselves and what they do with others is called “zone of development” [104]. This idea

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is also supported by many others and has been developed further by Rogoff [65], who introduced a socio-cultural developmental approach that highlights that children typically learn about what it is desirable to do from their proximate society. According to her, children do not have to perform but learn also by listening and observing the cultural behavior [105].

It has also been underlined that the child does not have to train every new activity from scratch, but “methods” (strategies and skills) are transferred from one activity to another [106]. Siegel concludes [100] that theories now approach each other, and that according to most theories, a wide range of constraints: anatomical, physiological, cognitive, and environmental guide the form of learning. Most contemporary developmentalists think that action from the child him/herself is to some extent necessary for development [100, 101, 107].

Occupational development

In the early years of the 1970s Mary Reilly [108] presented a model of occupational changes that could be seen as an occupational developmental model [11]. After her, until Coster [109] emphasised in 1998 the occupational perspective in assessment of children, most discussion of development in occupational therapy has stressed performance components such as motor, sensory and cognitive development, which were thought to give performance readiness [110]. In the beginning of the 21st century, two major models of occupational development were presented: the Interactive Model of Occupational Development (IMOD) [9] and the Process Transforming Occupation (PTO) [110, 111]. The IMOD [9] is based on an interactional perspective, and the interaction should be understood as bidirectional, meaning that the child is not only influenced by the environment but the environment is also influenced by the child. The PTO is derived from the socio-cultural developmental approach [65], emphasising that the child develops through social transaction, construction of occupational opportunities in socio-cultural groups and self-organization, i.e. learning by doing [105, 112-114]. Both models highlight the concept of the active child and how activity in itself is the engine for development [9, 112, 115].

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The IMOD is based on the premise that interaction is a key mechanism for occupational development [115]. The IMOD points out how *multiple determinacy* influences each child's development, meaning that no single fact is more important than the other in forming development, but as Davis and Polatajko states "*Rather, occupation results from an intentional and particular behaviour by a particular person in a particular environment*" [9 p.144]. Four groups of determinants are described but not in hierarchical order. The first determinant is the *person* (heredity/genes, learning/plasticity, and active participation/motivation). The second determinant is the *environment*: physical/social environment and the historical/cultural. The third is the *occupational* determinant: exposure and expectations. The fourth is the *interaction* determinant, i.e. the fit or how well the other can interact. The interaction is more than all these determinants put together. The determinants are not just additive, but one can diminish the other and vice versa. If the opportunities don't match the person, it can be a great hindrance [9].

Definition of development in this thesis

In this thesis development is seen as an interactional process of multiple determinacy where all factors are equally important, and it is the fit between the factors that is important to enable development. To maximise the fit, all parts have to be understood. The whole is different for each individual as all the integral parts create the whole [9].

RATIONALE FOR THIS THESIS

The current knowledge and research on spina bifida showed evident gaps when it came to describing how the cognitive phenotype in children with SB [27, 28] influenced everyday life such as task performance, autonomy and participation. This knowledge gap led to the research question for this thesis; if a child has deficiencies in their executive functions, seen as a low inner drive, to what extent does that impact on his/her possibility "of taking over" and performing everyday activities in a competent manner?

The hypothesis of this thesis was that children with SB have deficits in their ability **to do**, and that these difficulties influence both their autonomy and their participation in everyday life and society. This aim of this thesis was to fill the current knowledge gap by studying how children with SB "do things" and to investigate how this

ability to do is related to their participation in everyday life. Exploring possible relationships between doing, autonomy and participation may help identify new patterns for understanding the complex map of functioning in everyday life for children with SB. Hopefully, the results of this thesis will be able to guide interventions and methods in order to promote autonomy and participation in children born with SB.

Research perspectives

The diagnosis of spina bifida, the concepts of occupation/doing, participation and development are all described as highly complex. According to complexity theories, being complex means that the whole is more than the integral parts simply added to each other. To understand the landscape of complexity, not only the whole has to be studied but also the **patterns of interaction** between the integral parts [116]. Using complexity theory is to understand patterns, not only “*read the map but also assume an active role in cartography itself*” [116 p.592]. Developmental disability is such a complex phenomenon that there must be collaboration between disciplines [117]. This thesis makes an attempt to present an occupational therapy perspective of the functioning of children with SB and has the ambition to add new knowledge by describing the pattern of relations between the ability to do and autonomy /participation. This pattern of relations has not been described earlier.

How is research on such a complex construct as the one above possible? If a construct is complex and pluralistic, the most suitable philosophical choice is to take a pragmatic position that can allow the researcher to use mixed models and/or methods to find the best solution to answering the research question [118]. In a pragmatic research perspective, the research questions direct the methods and not the other way round as is often the case in perspectives that are called either qualitative and quantitative [118]. The data collection in three of the studies included in this thesis can be regarded as mixed models according to the conceptualization described by Johnson and Onwuegbuzie [118], that is, the way to collect data in itself was both qualitative and quantitative. The methods for analysing the data in this thesis were, on the other hand, all done with quantitative methods.

AIMS

The overall aim of this thesis was to increase knowledge of the quality of performance of everyday activities, autonomy and participation in children with spina bifida and to explore how they relate to each other.

The specific aims were:

- To evaluate the quality of the performance of everyday activities in children with spina bifida compared with children without known disabilities (*Study I*).
- to investigate if there were systematic differences in performance ability measures, measured with the Assessment of Motor and Process Skills (AMPS), between children from the Nordic countries and children from North America, and to investigate if the internationally based age-normative values in the AMPS are applicable to children from both of these two regions (*Study II*).
- to investigate the relationship between the level of autonomy and the quality of performance of everyday activities, expressed as motor and process skills, in children with spina bifida, and to study the agreement between the children's and the parents' ratings of autonomy (*Study III*).
- to evaluate the patterns of participation in school-related activities and settings in children with SB as perceived both by the children and their teachers, and to explore how the child's motor and process skills in task performance were related to his/her level of participation at school (*Study IV*).

METHODS

This thesis contains three studies concerning children with SB and their everyday life and one study concerning the validity of the age-mean values for the main instrument the AMPS used in the three other studies.

The thesis addresses performance skills, autonomy and participation. Two dimensions of participation are addressed as both the quantitative dimensions, i.e. frequency of participation in different types of school-related activities and the dimension of the quality of the participation, i.e. the level of active participation in different school settings, were rated.

PROCEDURES AND PARTICIPANTS

Studies I, III and IV were prospective studies of a population-based cohort of children all born with spina bifida in the period 1993-1999. All children (n = 65) born in this period with the diagnosis myelomeningocele (MMC) or lipo-myelomeningocele (lipo-MMC), living in the Swedish region of Västra Götaland, including the counties of Halland and Värmland on 31.12.2006 were identified. Inclusion criteria for the studies were: undergoing annual routine check-ups at the Regional Rehabilitation Centre, Queen Silvia Children's Hospital in Gothenburg (n=64). Exclusion criteria were: (a) motor dysfunction of upper extremities of such severity that the child was unable to drive a manual wheelchair independently thus making it impossible to participate in a performance assessment (n=2), or, (b) had autism as an additional diagnosis (n=1) thus making it difficult to verbally answer questions about his/her level of autonomy and participation (figure 1).

Sixty-one children/families were subsequently invited to participate in the study, data collection taking place during the child's annual routine check-ups in 2006-2007. Fifty of these 61 families accepted to participate (n=50), giving a consent rate of 82% (figure 1). Those 50 children constituted the study group of children with SB in studies I, III, IV. At the time of the data collection, the children were between 6 and 14 years of age, mean age 10.5 years (SD 2y). Forty-five of them were diagnosed with MMC and five with lipo-MMC. The majority (78 %) of the children had shunt-treated

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hydrocephalus (HC+). The group of children whose families declined to participate did not vary significantly from the study group with regard to age, gender, type of SB or whether the child had shunt-treated hydrocephalus or not (table 1).

Study I, the participants were the study group of children with SB (n=50) and their performance ability measures were compared to an equally large control group of children without any known disabilities living in the Nordic countries matched for age and gender. The children in the control group were randomly selected from the AMPS international database [119] (figure 1).

Study II was a retrospective study of data available from the AMPS international database on March 4th 2010, which consisted of the motor and process logits from all children from the Nordic countries (n=2 374) and from North America (n= 2 239), aged 3-15 years, that were without known disabilities (total n = 4 613).

In *Study III* the study group of children from study I with SB (n=50) were together with their parents (n= 50) the participants (figure 1). The children and their parents participated individually as respondents to the ratings of the child's autonomy. The parents were the ones that accompanied the child during the visit to the Regional Rehabilitation Centre in Gothenburg when the assessment and interview took place.

In *Study IV* the participants were the 50 children in the study group of children with SB described above and the teachers of 48 of those children, both responding to the ratings of the child's frequency of participation in school activities (figure 1). The teachers were contacted by phone by the doctoral student (MP-D) and asked for their informed consent to participate (teachers of two of the children declined to participate).

DATA COLLECTION

Participant's characteristics

The characteristics of the study group in studies I, II and III were provided by the parents of the children except for the diagnoses that were collected from the medical records available at the Regional Rehabilitation Centre, Queen Silvia's Children's Hospital in Gothenburg. The ambulation level of the children was classified according to the Hoffer

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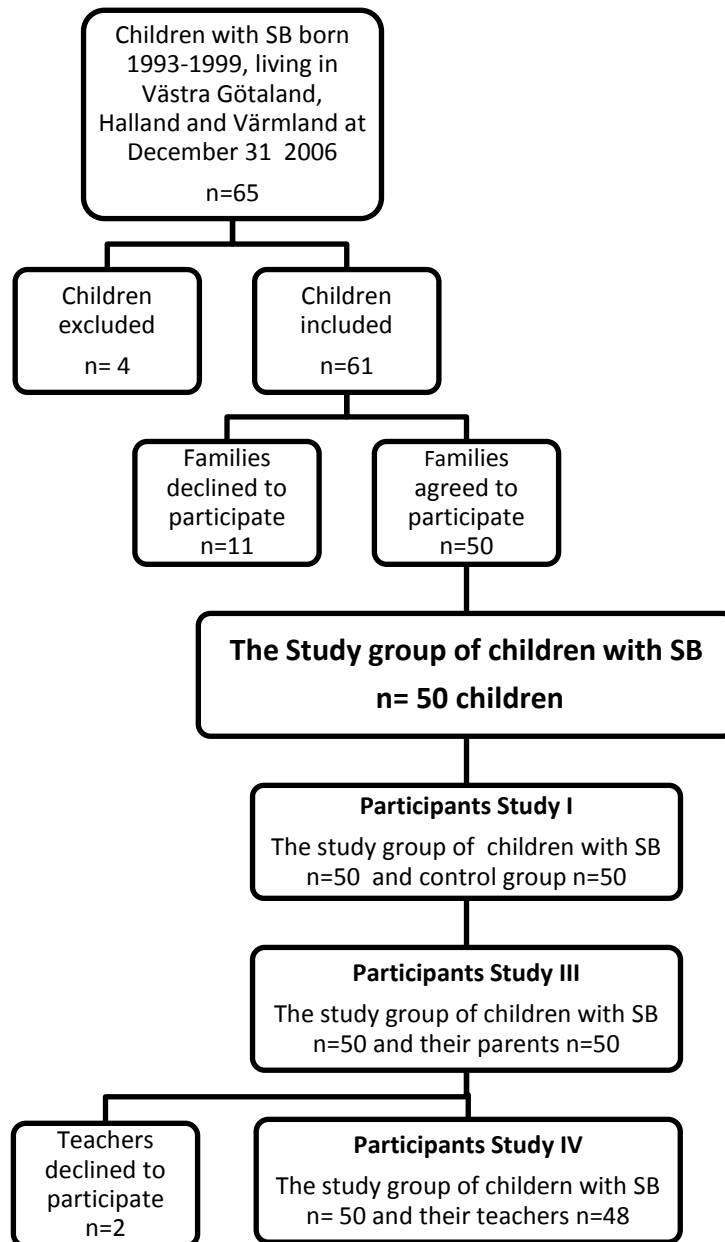


Figure 1. The study group of children with SB (n=50), and participants in study I, III, IV.

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scale [120] after the parent's description. The Hoffer Scale describes the ambulatory status of an individual with SB on a 4-point Likert scale: 1: community ambulator, 2: household ambulator, 3: non-functional ambulator (only for training) and 4: non-ambulatory children.

Table 1. Characteristics of the study group of children with spina bifida (n=50) and the non- participants (n=11) ns = non significance

	Study Group n =50(%)	Non Participants n =11(%)	p-value
<i>Gender</i>			
Male	26 (52)	7 (63)	ns
<i>Age</i>			
Mean, years: months	10:5	10:8	ns
Range, years	6-14	6-13	
<i>Hydrocephalus</i>			
Shunted (HC+)	39 (78)	7 (64)	ns
Not shunted (HC-)	11 (22)	4 (36)	ns
<i>Type of SB</i>			
MMC	45 (90)	8 (73)	ns
Lipo-MMC	5 (10)	3 (27)	ns
<i>Ambulation (Hoffer scale)</i>			
1- in community	23 (46)		
2- in household	7 (14)		
3 - for training, non-functional	8 (16)		
4 - non-ambulator	12 (24)		
<i>School form</i>			
Mainstream school	41 (82)		
Special education	5 (10)		
Preschool	4 (8)		
<i>Additory diagnosis</i>			
Epilepsy	3 (6)		
Cerebral palsy	1 (2)		
Attention Deficit Hyperactive Disorder (ADHD)	1 (2)		

Quality of performance of everyday activities

The quality of performance of everyday tasks was evaluated with the AMPS, a criterion and norm-based standardised observational assessment in which the child's performance

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skills are assessed while performing well-known, self-chosen and meaningful tasks [57]. The AMPS evaluates how the individual can apply skills in task performance, i.e. the quality of the motor and process skills used [57]. The person is observed during the actual doing of a task in a natural environment and rated by a trained and AMPS-certified occupational therapist (OT) [57]. The AMPS indicates whether a person has sufficient performance skills to successfully complete well-known everyday tasks in a competent manner i.e. in an effortless, efficient, safe and independent way [57]. The performance skills (16 motor skills and 20 process skills) evaluated are regarded as universal goal-directed actions that are comprised in and support all activities of daily living (ADL). Motor skill items are the observable actions taken to move oneself and the objects during the task with posture, coordination, mobility, strength and effort sufficient to perform the task. Process skill items are actions taken to proceed with the task from start to end, which covers actions to initiate, to use knowledge, to organise and logically sequence the actions of the task performance over time, and to solve problems occurring, select proper tools and keep on heading towards the goal [57].

The AMPS differs from the other assessments of ability in self-care and ADL as it evaluates **how** a task is performed instead of evaluating **what** task the child performs [121]. The AMPS has been recognised as an ecologically valid instrument for assessing consequences of executive dysfunctions [122] even though is not designed to measure executive function specifically [121]. The 36 motor and process skills scored in an AMPS assessment are measured in the context of a task performance in an ecological setting, and not as specific component skills that can be assessed in parts, and this is what captures the essence of executive skills, so hard to measure in “laborative settings”[57, 121]. The AMPS is an example of a mixed model instrument [118]. An AMPS assessment is carried out by an observational method, which means that the OT observes and judges the quality of the performance skills of a person. But to be able to predict and to measure changes in the performance skills, the instrument is based on a standardised procedure to rate the observed quality according to an ordinal scale. The scale is finally transformed via logistic transformation according to Rasch analysis to an interval scale, thus making quantitative and parametric analysis possible [57, 123].

Methods

The first step in an AMPS assessment is to interview the child in order to make him/her decide what task to perform. The child chooses two or three ADL- tasks out of 83 presented in the AMPS manual that he/she is motivated to do and normally does. After the observations, the occupational therapist rates the quality of the child's performance of the motor skills and the process skills needed to complete the task on a four-point Likert scale; 4: adequate skill, 3: questionable skill, 2: ineffective skill and 1: markedly deficient skill. The raw ordinal scores obtained are then converted into interval data with the use of a many-faceted Rasch analysis in the AMPS software program [57, 124]. Through the Rasch analysis, the person's ability measures are adjusted for item difficulty, task challenge and the rater severity [57]. The result is presented as two ability measures denoted logits, one for ADL motor ability and one for ADL process ability. High motor and/or process ability measures indicate that the child is more able and low motor and process skills that he/she is less able. The AMPS software program provides both cut-off values, under which the child's performance was at risk of not being safe and efficient enough to be performed independently, as well as internationally based age normative values. The AMPS has been found to have excellent intra- and inter-rater reliability [125] and validity for use from the age of three years to adults [126], for different diagnoses, including developmental disabilities [127].

Autonomy level

The "Autonomy Scale" from "Children's participation in school", described by Eriksson and Granlund [128], was used to rate both the children's and the parents' perceptions of the child's autonomy level. The questionnaire used, for children aged 7-12 years [128, 129], is a short version of the autonomy scale in the Arc's Self-Determination Scale, which was originally constructed by Wehmeyer [130], translated into Swedish and adapted to Swedish conditions. This scale has been used in several studies [79, 82, 128, 131, 132]. The questionnaire consists of 23 items/activities separated into five sections that represent different situations in daily life, such as routines in the family, interacting with the environment and community involvement, leisure and in how to express oneself. Both executive autonomy and decision-making autonomy are represented in the items that are presented as statements [130, 132] such as; *I make my own sandwich, I decide*

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how I use my pocket money, or My friend and I decide together what to do. Each item is scored on a four-point Likert scale designed with the aim to rate the child's perception of his/her autonomous behaviour when the environmental conditions do not interfere; 1: I never do that even when I have the chance; 2: I do that sometimes if I have the chance, 3: I do that most of the time if I have the chance, 4: I always do if I have the chance [130]. Internal consistency is reported as Cronbach alpha; α 0.82 for children 7-12 years and α 0.93 for the age group 13-17 years [79].

Frequencies of participation in school activities

In order to rate the child's frequency of participation in school-related activities, both children and teachers were interviewed using a Swedish version [79, 131] of the "Availability and Participation in School scale", an instrument originally developed by Simeonsson and collaborators in 2001 [67]. This instrument was constructed to rate the frequencies of the child's participation in an available school-related activity [67]. The scale for participation is based on frequency measures from 0-3; never, seldom, often or always participate [67]. The Swedish version of the instrument was adapted to the Swedish school context by Almqvist /Eriksson [79, 131] and originally consisted of 25 activities. When used in study IV of this thesis, three activities from the Swedish version (*Play at schoolyard, Outdoor play and Recess/pause activities*) were merged into one activity under the label of *Recess activities*. This was done for two reasons; firstly, the older children said that they never "played", and secondly for a cultural reason, as Swedish children of young age usually spend their recess outdoors in the schoolyard. The Swedish version of the instrument has been used in several studies [79, 131] and tested for internal consistency and found to have a Cronbach Alpha of: α 0.71 for children and α 0.88 for adults [131].

Level of active participation in school

The level of participation in school was rated only by the teachers using a Swedish version of the School Function Assessment (SFA) [109], part one (Participation). SFA is divided into three parts and designed to measure a pupil's participation in different school settings, functional performance of school-related tasks and activities, and assistance

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needed [109]. SFA is a criterion reference instrument and this used part of the instrument is based on the teachers' or school assistants' judgment of the child's active involvement/participation in six school settings considered to be the major settings of a school day i.e. *Classroom, Playground/recess, Transportation (to and from school), Bathroom /toileting, Transfers (in school) and Mealtime/snack time* [109]. SFA examines the teachers' perception of the student's level of participation in a qualitative manner, defined as the level of active involvement on a 6-point Likert scale; 1: participation extremely limited, 2: participation in a few activities, 3: participation in all aspects with constant supervision, 4: participation in all aspects with occasional assistance 5: modified full participation, 6: full participation [109]. The sum of raw scores in SFA can be transformed to a criterion score [133]. The criterion score ranges from 0-100. A score of 100 represents a criterion of full grade appropriate participation. Scores below 100 represent some reduction in participation level [133]. The validity and reliability for the SFA to be used for children with disabilities have been reported in several studies [134, 135]. The preliminary Swedish version was translated by a paediatric occupational therapist and back-translated by an authorized translator for use in the present study. The reliability of this Swedish version was also tested before the use in this study in a pilot study on test-retest with 13 teachers of 6-12 year old children in 10 mainstream schools. The result of the pilot study showed that the Kappa coefficient (k) was between 0.51 and 0.77, and the percentage agreement (PA) ranged from 63-91% (mean 78%), which is, according to Dekker and colleagues [136], a moderate to good test-retest variability sufficient for group comparisons.

Table 2: Summary of instruments, data analysis and data analysis methods.

Study	Instruments	Data analysis and methods
I	Assessment of Motor and Process Skills (AMPS)	Descriptive and comparative, <i>Fisher's exact test and Mann Whitney U-test</i>
II	Assessment of Motor and Process Skills (AMPS)	Descriptive and comparative, <i>Two -way ANOVA</i>
III	The Autonomy Scale Assessment of Motor and Process Skills (AMPS)	Descriptive, comparative and explorative, <i>Percentage agreement, Weighted Kappa, Sign-test, Binary logistic regression analysis</i>
IV	The Availability and Participation Scale School Function Assessment (SFA) Assessment of Motor and Process Skills (AMPS)	Descriptive and explorative, <i>Binary logistic regression analysis.</i>

DATA ANALYSIS

Study I:

The results (the two ADL ability measures, one for motor and one for process skills) of the AMPS assessment were compared with age norm values for children without known disability presented in the AMPS software program [124]. The results from the study group were also compared, using a non-parametric method; the Mann-Whitney U-test, with data from a control group of children without known disabilities from the Nordic countries from the international database, matched for age and sex. Non-parametric methods; Fisher's exact test and the Kruska Wallis test, were used to compare the different subgroups of children in the study group: (i) with shunt treated hydrocephalus (HC+) and non-ambulators, (ii) with HC+ and ambulators and (iii) without shunt treated hydrocephalus (HC-) and ambulators.

Study II:

The cross-cultural differences in the age norm values in the AMPS were analysed using a parametric method (two-way ANOVA) comparing the two largest groups in the standardization sample. The actual differences in logits were calculated.

Study III:

The agreement between the children's and the parents' ratings was analysed using methods suitable for ordered categorical data: percentage agreements, weighted Kappa analysis, and Sign test. The relationship between the autonomy levels and the child's age, motor skills and process skills from the AMPS assessment was analysed with binary logistic regression.

Study IV:

The frequencies of participation were presented as medians for each activity, the children's and teacher's ratings separately.

The results of the teachers' ratings with the SFA were presented as medians for each setting and as means for the criterion scores. Binary logistic regression analysis was used

to explore the relationship between the teacher's rating on the SFA and the child's motor and process skills measured with AMPS (Table 2).

ETHICS

The Regional Ethical Review Board in Gothenburg, Sweden, was consulted prior to the studies I, II and IV (dnr 574-05). Their response was that formal ethical committee approval was deemed unnecessary but they supplied guidelines for the information in the consent letters that were sent to the children and the parents. For Study II the Research Ethics Committee, Faculty of Medicine, Umeå University, Sweden had approved the use of the data from the database in cross-cultural studies (dnr; 03-509).

Ethical considerations were identified mainly on two issues, the first concerning the children's consent to participation in the study, the second concerning the information on the results to the parents. An information and consent letter was sent to families before their yearly visit to the Urotherapeutic Unit at the Regional Rehabilitation Centre at Queen Silvias Children's Hospital. Included in this was both a letter to the parents with information on the studies and information of their right not to participate and to end the participation at any point in the procedure, and a letter to the children (that was adapted for two different age groups; 6-9 and 10-14). The parents were asked to sign the consent agreement that was to be sent back to the doctoral student (M P-D) before the visit to the centre. The children (as some of them were very young) did not themselves sign any consent agreement; the discussion on the child's willingness to participate was handled within the families. Some of the families that declined participation reported that it was due to the child him/herself not being willing to participate.

The parents were informed that they had the possibility to receive the results concerning their child (except for the children's self-ratings), and most of the parents asked for and were informed of the results of the AMPS assessments.

RESULTS

Summary of results

The hypothesis of this thesis that children with SB have deficits in their ability to do, and that these difficulties influence both their autonomy and their participation in everyday life and society was confirmed by the results of the studies in the thesis. The children with SB in these studies had difficulty performing well-known everyday activities in an effortless, efficient, safe and independent way demonstrated by low ADL motor and process ability measures. This deficient quality of task performance was strongly related to both their level of autonomy in daily life and their level of active participation in school. In particular the process skills had in many items a significant relation to how the child's autonomy in everyday life was rated, especially in the parental rating. For the level of active participation in school, the two performance skills both predicted active participation, but in the classroom motor skills seemed to have the strongest influence.

The thesis showed that the children with SB had lower autonomy levels in activities and situations that were goal-directed and needed personal initiation or decision-making than in leisure activities. The results also showed that parents and children did not agree on the child's autonomy level but that the children, when asked in a concrete manner, were aware of their own doing in everyday life. In school, children with SB had a high frequency of participation, higher according to themselves than according to their teachers. However, the teacher rated the children's **active** participation as restricted. This thesis demonstrates the need for OTs working with children born with SB to specifically evaluate the child's performance skills. The results of this thesis also showed that the AMPS is a valid instrument to use for this purpose in a Nordic context.

Study I

The first study showed that children in the cohort with SB had low levels of both motor and process skills compared to children without known disability. The results from the AMPS assessment of the children with SB compared to age norm values revealed that 60% of the children with SB in the present study group had motor ability measures (logits) that were 2SD below their age means, and that 48% had process ability measures

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(logits) that were 2SD below their age mean. Children from all three subgroups were found among those that fell below 2SD both on motor and on process ability measures, but there were significant differences between the groups ($p = .009$) indicating that a larger proportion of the children with low levels of performance skills had shunt-treated hydrocephalus (HC+) and were non-ambulators (table 3).

Compared with the control group of Nordic children (matched for age and gender), the children with SB had significantly lower medians of both motor ($p < .001$) and process ability measures ($p < .001$), which confirmed the results from the comparison with the international age normative values.

Table 3. Motor and process ability measures of children with spina bifida, divided in three subgroups, compared with the internationally based age norm from the AMPS data base. (HC+ = shunted hydrocephalus, HC- non shunted hydrocephalus)

		HC+ non ambulator n = 20	HC+ ambulator n = 19	HC- ambulator n = 11	Total n= 50 (%)
Motor ability measures	<i>Over age norm (+2SD)</i>	0	0	0	0 (0)
	<i>High (+1SD)</i>	0	0	0	0 (0)
	<i>Within mean range</i>	1	2	5	8 (16)
	<i>Low (-1SD)</i>	0	8	4	12 (24)
	<i>Under age norm (-2SD)</i>	19	9	2	30 (60)
Process ability measures	<i>Over age norm (+2SD)</i>	0	0	0	0 (0)
	<i>High (+1SD)</i>	1	1	1	3 (6)
	<i>Within mean range</i>	0	6	4	10 (20)
	<i>Low (-1SD)</i>	4	5	4	13 (26)
	<i>Under age norm (-2SD)</i>	15	7	2	24 (48)

The profile of raw scores from the AMPS assessment revealed that the motor skills most often rated as marked deficiency were: *positions* (severe deficits in positioning the arm or body appropriately in relation to task objects), *bends* (failure to bend or twist the body appropriately to the task), *reaches* (failure to secure task objects when reaching), *calibrates* (severely deficient regulation of force or speed of task related actions) and *paces* (severely deficient rate of task performance). The five process skills most often rated as markedly deficient were *accommodates* (i.e., severe deficit to modify actions to overcome problems), *initiates* (failure to initiate actions or steps of tasks),

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notices-responds (failure to respond to task-relevant cues from the environment), *inquires* (asks many questions related to information that had already been discussed/clarified prior to beginning the task) and *adjusts* (failure to change workplaces or adjust switches and dials to overcome problems).

Study II

This study showed that there were no systematic differences of clinical relevance in the AMPS between children from the Nordic countries and children from North America. The age mean values should therefore be considered valid for use in research concerning children who have grown up in those two regions.

The result of the two-way ANOVA test of variance for the ADL motor ability measures showed no significant age by region interaction effect; $F = 1.455$ (df12) $p = 0.133$, but a significant difference between the regions $F = 30.80$ (df1) $p < 0.001$ was found, with the Nordic children having the higher values (figure 1). The differences were not large enough to be considered clinically relevant as no age group showed a difference in logits for ADL motor ability measures larger than 1.96 SEM (± 0.49 logits) The largest actual difference in motor logits (0.18 logits) was found for the 13-year-olds.

The ANOVA comparison of the variance for the ADL process ability measures between the two regions showed neither a significant age by region interaction effect; $F = 1.086$ (df12) $p = 0.367$ nor a significant difference between the regions $F = 1.88$ (df1), $p = 0.170$. In no age group was the difference larger than 1.96 SEM ($< \pm 0.39$ logits) (table 2), and the largest actual difference in logits for process ability was found for the 4-year-olds (0.12 logits).

The ADL ability measures of the Nordic children and the North American children respectively did not differ from the current age-norm values presented in the AMPS manual by more than ± 1.96 SEM.

Study III

The study showed that the children with SB had low levels of autonomy in self-care activities and in decision-making. Autonomy levels were higher in leisure and relationship-based situations. Process skills seem to have a strong relation to autonomy in

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children with spina bifida. It was noteworthy that the children and parents did not agree about the child's level of autonomy, but that children with SB seemed to be aware of their own lack of autonomy in everyday life.

Low levels (medians of 2 or below) of autonomy were most frequent in the section of the Autonomy Scale concerning; "routine, personal care and family oriented functions" both according to the children's and the parents' rating. The lowest median was found in this section in the item *Packing my things for physical education* (median of 1). In sections "interacting with the environment" and "personal expression", there were low ratings in two of four possible items from the children and in one of four possible from the parents. In no other section were there low ratings in half or more of the items.

In this study it was the process skills that had the most striking relation to the autonomy level of the child with SB, both according to the children and the parents. Motor skills also had a significant, and for some items, a strong relation to the autonomy level. Age was found to have a more moderate relation (table 4).

There was little agreement between children and parents concerning the child's level of autonomy; the PA ranged from 26-82% with a median of 46%. Only one item had a high PA (>80%) between the parents and their children. The results showed that for most items there were no significant one-sided statistical differences between the raters. In the two items where there were differences, the children rated themselves as being less autonomous than their parents did.

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Table 4. Odds Ratio (OR) for the child being autonomous in children's and parents' rating when having process ability values within their age norm ($\pm 2SD$). The logistic regression models also included age, which is not presented in this table but explained in the text. * $p < 0.05$ ** $p < 0.01$ *** $p < 0.001$, n.v. = non valid

Autonomy Scale Sections	Items	AMPS Motor Ability < 2SD Child	AMPS Motor Ability < 2SD Parent	AMPS Process Ability < 2SD Child	AMPS Process Ability < 2SD Parent
Routine, personal care and family oriented functions	I make my own sandwich	3.971	2.659	1.562	2.887
	I do my chores	0.618	2.688	0.772	5.618**
	I know where my own things are	0.777	1.454	1.371	1.421
	I decide what clothes to wear	1.813	4.428*	6.802**	6.191**
	I pack my things for physical education	3.271	4.398	6.882*	14.468*
	I know what day we have physical education	3.779	2.682	4.944	10.373*
Interacting with the environment	I make friends with kids my own age	0.530	9.013	0.542	2.021*
	I can be in time for a meeting	3.794*	8.903**	3.590*	7.558**
	I talk to people I don't know on my own	0.701	0.342	0.674	0.398
	I participate in setting up my individual plan	3.960	3.281	12.411**	4.785
Recreational and leisure choices	What I do during leisure time is my choice	8.790	1.567	8.877	4.839
	I participate in the decision about what my family do on the weekend	1.006	1.509	1.711	2.190
	I meet my schoolmates in my free time	1.962	3.461*	0.872	2.066
	My friends and I decide together what to do	4.174	1.884	2.583	1.880
	I write letters/ e-mails and make phone calls	0.846	2.188	0.732	1.292
	I listen to music that I like	0.880	2.061	1.523	0.606
Community involvement and interaction	I do leisure activities based on my interests	n.v.	0.620	9.154	1.411
	I join activity groups for children in the same situation as I am.	0.671	1.328	1.150	2.035
Personal expression	I choose my clothes and the personal items I use everyday	2.015	3.795*	3.690*	13.279***
	I choose what gifts to give	0.961	7.710*	1.834	6.099*
	I have chosen how to furnish my room	10.155**	2.494	6.518**	2.112
	I decide how I use my pocket money	1.928	4.116	3.047	3.838

Study IV

Study IV showed that children with SB participated frequently in most school activities. However, the teachers did not rate the actual level of the child's active participation as being full in all school settings. The lowest level of active participation was found during

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recess. Motor skills had the strongest relation to active participation in the classroom and in mealtime settings, while in recess and bathroom settings the relation to process skills was equally strong (table 5).

The results of frequencies of participation presented in this study were all based on the ratings of the children in the activities that were available in their school. This study showed that, both according to the children and their teachers, children with SB had high frequencies of participation in most school activities, although the teachers rated lower frequencies in *Recess activities, Organised games, Helping peers out, and Gym/sports*. In two activities, *Pupils council* and *Meal council*, both the children and their teachers rated very low (medians of 0 and 1 i.e. never or seldom participating).

On SFA 89.6% of the teachers rated the children's level of active participation as restricted. The mean SFA criterion score for participation from the teachers' ratings was 62.8 (SD 16.3), range 27-93. The medians of the raw score for each setting ranged from 5 (modified full participation) in *Transportation, Transition* and *Mealtime/snack time* to the lowest median of 3 (participation in all aspects with constant supervision) found for the setting of *Playground/recess. Classroom* setting had a median of 4.5.

Table 5. Odds Ratio (OR) for the child being actively participating, resulting from logistic regression made in two equal models of age and motor skill (model 1) and age and process skills (model 2). OR >1 is indicating that the child was more likely to be actively participating when older or when having performance skills within age norm. * p<0.05 ** p<0.01

Settings	Model 1		Model 2	
	OR age	OR motor	OR age	OR process
Classroom	1.17	9.38*	1.03	5.84**
Playground /recess	9.16	6.45**	0.84	5.48*
Transportation	1.21	5.88*	1.10	4.64*
Hygiene/toileting	1.31	4.18*	1.22	4.11*
Transitions	1.46*	4.29 ¹	1.34	2.51
Mealtime/Snack time	1.75	10.71*	1.52*	5.31*

¹ p= 0.052

DISCUSSION

METHODOLOGICAL CONSIDERATIONS

The studies in this thesis were cross-sectional, and as such they do not describe the long-term development of the performance skills and autonomy and social participation in children with SB. However, according to long-term outcome studies [18, 44, 137] of individuals with SB, it can be presumed that the problems will follow the individual into adulthood, as the results from those studies actually show that their independence rate as adults is still low.

In all studies in this thesis, quantitative statistical methods were used for data analysis. If these studies had been supplemented with a qualitative or mixed-method study, this could have generated different and perhaps deeper information concerning the children's subjective experiences of their situation. However as the aim of this thesis was to explore (i) how the ability to perform everyday tasks is related to autonomy and participation in children with SB, (ii) the agreement between the children and the parents opinions of the child's autonomy level, objective and comparable data were needed.

The selection of participants was one of the assets in this thesis. This was the case both in the studies concerning children with SB (studies I, III and IV), which were based on a population-based cohort, and in the methodological study, which had a very large sample (study II). At the same time, the participant selection in study III had a limitation since the parents were not separated into mothers or fathers but all seen as parents. This might have influenced the result in this study [138].

The study group of children in studies I, II and IV had a relatively large age span (6-14 years). This created difficulties, both in how the rating scales were used and in the use of the actual items in the Availability and Participation in School Scale. The first issue was solved by the procedure used that all the questions/statements in the rating scales were read aloud to all participants, children as well as parents, and even teachers as they were interviewed by phone. In this way, the interviewer (M P-D) was able to make sure that the questions/statements were understood. Concerning the second issue about the items in the participation scale, this was solved by the merging of all recess activities

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into one and by omitting the few activities that were not applicable for more than 80% of the children.

The data collection instruments in themselves also had their strengths and limitations. The age norms presented in the AMPS was proved in study II to be suitable for use in children living in the Nordic countries. The AMPS is described as an ecologically valid instrument to evaluate the consequences of executive dysfunctions in everyday life [122], and as such it showed how large these consequences were in this group. Executive functions are hard to assess, and it is best done in an actual life situation [139, 140] as in the AMPS. Still, the AMPS has one limitation, as the procedure of the assessment compensates for the child's possible inability of getting the idea of doing something (as the OT proposes the initial idea of choosing the tasks) [57]. Due to this limitation children with SB might have even larger problems with getting something done in real life than the ones demonstrated in study I. The largest asset of the Autonomy Scale [130] was the possibility of asking very concretely; What do you do? (addressing the child) and What does your child do? (addressing the parent). It is important to differentiate what the child with SB actually does on his/her own initiative as opposed to what he/she can do together with someone. The Availability and Participation in School Scale [67] does capture if the reason for not participating is lack of access and availability, but it does not capture why the child does not participate in an available activity. Neither does it capture how actively the child participates. In this respect, using it in combination with SFA [133] was very fruitful. It was a limitation though, that a similar scale was not available for use for the children.

GENERAL DISCUSSION

This thesis has investigated and revealed a pattern of relations between the ability to do/perform and autonomy in everyday life and participation in school not earlier explored in children with spina bifida. This new pattern will be discussed in the light of the complex situation of the child with SB in relation to the three perspectives described in the introduction.

Doing and its impact on autonomy and participation

This thesis has shown that children with SB commonly had problems “getting through” the process of doing on their own, even in familiar and self-chosen activities (study I). Their most used strategy was **to ask and to ask again**, to get guided. The result is in accordance with contemporary findings with regard to neuropsychological function in individuals with SB [27, 28, 51]. The results throw light on the everyday consequences of the cognitive phenotype in individuals with SB. This phenotype (or modal profile) is described [27, 28] to be processing-specific, implying that individuals with SB often are successful in everything that is guided and led by someone. However, they often run into difficulties when they have to construct, predict and initiate something and act on his/her own initiative. Thus it is possible that the difficulty doing things and being autonomous should be considered to be regarded as an activity/participation-related phenotype, expressed as: **Children with SB commonly have difficulty getting things done on their own, and this influences their autonomy and restricts their active participation.** It might even be so that the difficulty getting things done is a more hindering factor in everyday life than the difficulties connected with ambulation. According to the AMPS assessment and the teachers’ rating of active participation (studies I and IV), getting around in a wheelchair was rarely a problem. Ambulation difficulties can and were often compensated, but the skills in initiating and problem-solving are not so easily compensated, and this affects the efficiency in doing most things in life. Getting things done is necessary to be able to cope with everyday routines, but for individuals with SB, of which the majority have bowel and bladder dysfunction and have to carry out clean intermittent catheterisation every 3 hours, 5-6 times a day, the consequences of “not doing” might even be fatal [137].

SB is an extremely complex condition and, according to complexity theories, different patterns of interaction between the ingoing parts have to be studied [116]. The results of this thesis present a new pattern of relation between two outcome variables as it describes the patterns of interaction between the quality of performance on the one hand and autonomy and participation on the other. Many previous studies looked at the outcome in social participation in relation to body structural factors or medical factors as lesion levels and hydrocephalus [141, 142], others have studied independence

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in relation to movement-related functions [37]. Study I in this thesis showed that the ability “to do” was linked to body structural and functional factors, as the highest proportion of severe problems with motor and process skills was found in the group of children with shunt-treated hydrocephalus who were non-ambulators (per se often a result of higher lesion level and form of SB). This result is congruent with studies that show that children with hydrocephalus have been found to have more severe neuropsychological and especially executive problems [30, 141-143]. However, and this is an important result of this study; some of the children from the group with shunt-treated hydrocephalus and non-ambulators did well, while some of the children from the other two sub-groups (ambulators with hydrocephalus and ambulators without hydrocephalus) performed poorly (below 2SD from their age mean in motor and process skills) (table 3). This indicates that it is of highest importance for the OTs engaged in clinical work to assess the performance skills in children with SB, as high motor and process skills seem to predict higher levels autonomy and active participation in school settings (studies III and IV). Study II showed that the AMPS is a reliable instrument for such an assessment also in a Nordic context. In study II, the age-norms of the AMPS were evaluated from a cross-cultural perspective. The result demonstrated that the quality of performance (the “how” something is done) presented in the instrument was not culturally-dependent (study II). This study pointed out that OTs need to understand both what (specifically) the child does in daily life and how he/she is able to perform activities on a general level (i.e. go through the process of doing) and thereby the need of looking at doing or performance in both a general and a specific way.

The teachers of children with SB rated the children’s level of **active** participation as low, especially in recess activities. To be regarded as actively participating in recess and playground settings, the child needs to involve him/herself in an unstructured situation and without guidance [144]. In the actual setting high levels of active participation were strongly related to both high process and high motor skills. It is important to emphasise the children’s decreased level of active participation in school (especially recess) as school is probably the most important arena for learning social skills and friendships [67, 145, 146]. As both motor and process skills had such strong relations to the level of active participation, being less active can not be explained solely

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as question of choice or motivation. It is probably largely due to difficulty initiating involvement and not being able to easily solve problems that occur in the changing scene of different settings in school. Therefore it is very important not to be content with the children being passive, particularly in peer-related activities. Earlier studies have shown that children and adolescents with SB have few friends [147-149] and become increasingly isolated in adolescence [48]. Adults in school (as teachers, recreation instructors and assistants) need to understand the impact that performance skills, and particularly process skills, has on the child's possibilities of participating in order to better tailor the environment and the support that they should give to the child.

The thesis has shown that children with SB do have the ability to express if they usually do or not do things in daily life in a concrete way, which is very important, especially as their opinions sometimes differed from those of their parents. However, we must be aware that, just because they can express what they do not do, this might not mean that they can just pull themselves together and do [150]. To remediate one's difficulties anticipatory awareness is also needed [150], which, according to Dennis and Barnes [27], individuals with SB often lack.

Wilcock [4] argues that it is through both doing and being that a person becomes what he has the potential to become. As children with SB become less autonomous and more passive because they have poor performance skills (i.e. have difficulty doing), it is extremely important to maximise the interaction between the child, his/her environment and the occupations he/she needs or wants to perform to enhance their ability "to do".

Development and children with spina bifida

In a family setting, it is not easy to detect if a child has difficulty initiating and carrying out tasks due to low process skills. Parents and children usually do things together, and modern developmental theories point out that children, typically developed, participate or observe activities together with more competent persons and little by little they "take over" the performance on their own initiative [105]. Skills learned in one activity are described to carry over to another one [105, 106, 112]. This thesis shows that children with SB had problems with carrying out even familiar activities and seemed to depend on

acting together with someone else even in well known everyday activities (studies I and III). The pattern of relations between the performance skills, autonomy and active participation in children with SB that this thesis has shown could contribute to a greater understanding of the high rates of dependence seen in all age groups of persons with SB [18, 33]. The results show that children with SB might commonly not be able to take over and initiate the doing, either literally or symbolically. Therefore low levels of independence in children and adolescents with SB should perhaps not be labelled delayed development [33] but alternative development. It is important to remember that for a child with a neurodevelopmental disability, typical patterns of developmental theories are not always applicable [6, 151].

Interaction between the child born with spina bifida, the environment and the occupation

According to the arguments above, studies concerning the role of parental styling in children with SB can be viewed in a new light. Maybe the difficulties initiating and carrying out everyday tasks commonly seen in children with SB **actually cause a typical reaction** from their parents [6, 103]. Scarr and McCartney [102] suggest that the child partly creates its own environment. According to the bidirectional interaction of factors in occupational development [9], the person is influenced by the environment, but the environment is also influenced by the person. In view of this interaction, it is obvious that the child is a part of the parents' environment and will therefore most probably influence the parents' rearing style. Zuckerman [51] argues that the achieving of adulthood milestones in young adults with SB depends on both their executive functions and their parents' rearing style. Still it is of highest importance to discuss the patterns of cause and effects. In their theory of genotype vs. environment effects Scarr and McCartney [102] argue that: *"It is more likely that persons with certain genotypes will receive certain kinds of parenting"* [102 p.428]. Thus over-protectiveness or parental intrusiveness [39, 41, 152] is perhaps not the right expression to use when discussing the cause of low levels of autonomy in children and adolescents with SB. These expressions put a heavy load of guilt on parents and even on the child him/herself as being "lazy" and considering the result of this thesis, one can argue, that the parents are probably not the source of the

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problems. However, still according to Scarr and McCartney [102], the parental role as mediators is very important for the outcome. To be able to deflect the disability by creating a better fit between the child's ability, the environment and the task, the parents have to understand the child's capacity and needs. If the determinants for the child's development is understood, it will be possible to coach the child in a more tailored and successful way [153]. Having the right information about one's child often leads to adequate expectation in parents, teachers and assistants [154], and can create hope for the future. This has been found to be important for the well-being of parents of children born with SB [155]. So it is important to point out that the type of environment (i.e parental styling) might be a solution – not the cause.

CONCLUSIONS

In conclusion, this thesis has demonstrated that children with SB commonly had extended problems with **getting things done** on their own, and that their level of autonomy and active participation in school was closely related to their ability to do. This finding emphasizes the importance of not waiting for the child with SB to “catch up” but to tailor the support from an early age, so that the child learns not only to know how to do something but also how to get it done. Much emphasis must therefore be put on how to increase the child’s ability to get things done, and especially to support and compensate for the low quality of process skills in children with SB.

The first implication of the results is that performance skills have to be specifically assessed by the OT working in the clinic, as without this assessment it is impossible to create effective interventions. Through an advocative /educative intervention model [156, 157] the child him/herself, the parents and the school personnel will get to know and understand something about the impact that low process and motor skills has on autonomy and participation. This knowledge should make it possible to maximise the fit between the child’s capacity, the environment and occupation at stake. Maximising the fit can be done by using adaptive intervention models [156, 157] aiming to compensate for lack of initiation and problem-solving abilities. To tailor such an adaptive model is not an easy target though, as Fletcher [28] argues that children with SB often have difficulty handling situations that give a freedom of choice, such as completing a planning list on their own.

There is no evidence to be found for any restorative/acquisitional intervention model in occupational therapy aiming to enhance the ability to get things done in everyday life for children with SB. In the last decade, the model of Cognitive Orientation to Occupational Performance (CO-OP) [158] has established some evidence for such a model in children with developmental coordination disorder [159]. Likewise, different intervention models exist for adults with brain injuries e.g. the Multicontextual approach [160] which aim to help the individual to learn and generalise strategy use. With the knowledge gained from this thesis that process skills are strongly related to

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autonomy and school participation in children with SB, strategy-learning and generalisation appear to be an interesting and important intervention model.

Children with SB in our studies were able to express their own opinions of their own doing/not doing in everyday life, often in disagreement with their parents. This leads to another important implication not to forget the child's own view in planning interventions. Still we must be aware that a child's concern is the "here and now", and that adults in their environment are obliged to lead the child to do things they do not wish for at the moment, such as algebra or being active in pupils' councils.

A third important implication is that in occupational therapy both the **what** and the **how** in occupational performance should be considered. The process of doing can and should be seen and analysed both in a general sense and in a specific sense. This should result in a new and more occupation centred role for the OTs working in the habilitation field.

FUTURE RESEARCH

As this thesis has revealed a new pattern of relations between the quality of the performance skills (the ability to do) and participation and autonomy in children with spina bifida, it paves the way for a large range of research questions that need to be further studied. Follow-up longitudinal studies of this pattern of relations should be conducted, in order to understand if the difficulties seen in the cross-sectional studies follow the child born with SB through adolescence and into adulthood.

There is also an urgent need for intervention studies to find intervention methods for children with SB that will enhance the quality of their motor and process skills and enable them to be more autonomous and participate more actively. Several questions remain to be answered, such as: How can children with SB learn to generalise strategy use? Do they need to learn each and every activity until it is done automatically? How can difficulties with initiation and low inner drive be compensated?

Qualitative studies need to be conducted with children born with spina bifida. Their special cognitive and activity/participation phenotype probably provides them with experiences of everyday life that are exclusive to this group.

Similar research to that presented in this thesis is consequently also needed for other groups of children with developmental disabilities to understand the pattern of relations between doing and autonomy/participation in those groups. Problems with doing in a general sense, i.e. with carrying out a whole process of doing, might be hidden in the shadow of physical disabilities in the same way as social-behavioural problem sometimes seems to be [146]. If this is the case it will open up a large, important and ground-breaking research field for occupational therapists.

Summary in Swedish

"Han kan nog men det blir liksom inte av" är en vanlig beskrivning av förmågan att utföra vardagsaktiviteter hos barn födda med ryggmärgsbråck. Ryggmärgsbråck är samlingsterm för en medfödd defekt i ryggradens kotor och dess taggutsnitt. Nyare forskningsresultat har visat att det är vanligt att barn med ryggmärgsbråck, förutom motoriska funktionsnedsättningar och neurogen blåsa och tarm också har kognitiva funktionsnedsättningar. Dessa har beskrivits som svårigheter med allt som individen själv skall förutse, konstruera och initiera. Studier har också visat att ungdomar med ryggmärgsbråck är mindre delaktiga i de aktiviteter som är viktiga för att mogna till autonoma vuxna och att unga vuxna med ryggmärgsbråck ofta hamnar utanför arbetsmarknaden. Som vuxna är de oftare än andra jämnåriga boende hos föräldrarna och en så låg andel som ca 35 % anses klara sitt dagliga liv helt självständigt. Orsakerna till delaktighetsinskränkningar har tidigare framförallt oftast sökts i miljön, såsom överbeskydd, socioekonomiska förutsättningar och tillgänglighet, eller i hälsotillstånd såsom svårighetsgrad grad av själva ryggmärgsbråcket och rörelseinskränkningar, eller inom personella faktorer såsom motivation. Trots nya forskningsresultat om svårigheter med att planera och initiera har inte samband mellan dessa förmågor och grad av autonomi och delaktighet tidigare belysts i gruppen barn med ryggmärgsbråck.

Hypotesen för denna avhandling var att en bidragande orsak till att barn med ryggmärgsbråck får delaktighetsinskränkningar och är mindre autonoma är att de har svårigheter att självständigt starta och genomföra relevanta vardagliga aktiviteter och inte "tar över" utförandet själva.

Syftet var att undersöka färdigheten att utföra vardagliga aktiviteter och att undersöka barnets och föräldrarnas uppfattning om barnets autonomi och den skattade delaktighet i skolaktiviteter hos barn med ryggmärgsbråck, samt att söka eventuella samband mellan dessa. Ett ytterligare syfte var att undersöka om instrumentet Assessment of Motor and Process Skills (AMPS) är lämpligt att använda för att bedöma barns färdighet i att utföra aktiviteter i en nordisk kontext.

Deltagare i studie I, III och IV var 50 av de 65 barn födda med ryggmärgsbråck 1993-1999 som bodde i Västra Götaland, Halland och Värmland den 31

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december 2016 och deras föräldrar (artikel III) och lärare (artikel IV). I artikel II bestod undersökningsdata av de motor- och processfärdighetsvärden mätta med AMPS som var tillgängliga i en internationell databas från barn boende i Norden (n=2374) och Nordamerika (n= 2239) utan kända funktionshinder.

Avhandlingen visade att majoriteten av de barn med ryggmärgsbråck som ingick i studierna hade svårt att initiera och på ett självständigt sätt "få gjort" vardagsaktiviteter och detta påverkade både deras autonominivå och deras möjlighet till aktivt deltagande i olika skolsituationer. Bedömningsinstrumentet AMPS visade sig vara användbart för att mäta färdigheter att utföra vardagsaktiviteter hos nordiska barn, då resultatet av studie II visade att de befintliga åldersnormerade värden i bedömningsinstrumentet var valida för barn som lever i de nordiska länderna. Avhandlingen visade också att barnen med ryggmärgsbråck i studien hade låg autonomi i vardagssituationer som var målinriktade och krävde egen initiering. Både barnen och deras lärare skattade frekvensen av delaktighet som hög i de flesta skolaktiviteter, dvs. de var med i de flesta skolrelaterade aktiviteter. Men detta står i kontrast till att lärarna skattade graden av **aktiv** delaktighet hos barnen med ryggmärgsbråck som lägre än hos klassen som helhet, speciellt på raster/skolgård.

Det är därför av yttersta vikt att arbetsterapeuter inom barnhabilitering specifikt bedömer och beskriver utvecklingen av utförandefärdigheter hos barn med ryggmärgsbråck för att kunna öka deras möjligheter till autonomi och delaktighet. Speciellt viktiga för detta förefaller barnets processfärdigheter att vara. Avhandlingen visar att arbetsterapeuter bör bedöma, inte bara vad barnet med ryggmärgsbråck kan göra, utan också hur "görandet" är och om de **får något gjort** på egen hand.

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