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DEVELOPMENT OF VISUAL-MOTOR COORDINATION IN CHILDREN WITH NEUROLOGICAL DYSFUNCTIONS

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To my parents Selma and Nedim

'Anyone who has never made a mistake has never tried anything new'
Albert Einstein

ABSTRACT

Graphomotor skills are complex perceptual-motor skills that are dependent upon the maturation and integration of a number of cognitive, perceptual and motor skills. They gradually develop through practice and instruction, and require good communication between the visual and the motor systems. During typical development, children at 3 years of age start to use graphics to record personally or culturally significant images in their environments. They build up rich graphical representations in preschool ages, and during school they learn to write the alphabet, numbers, and symbols to communicate. Problems in graphomotor skills are commonly seen in children with neurological dysfunctions.

The overall aim in this thesis was to further investigate the development of visual-motor coordination required for graphomotor skills in different paediatric populations with neurological dysfunction in relation to sensory-motor, cognitive and behavioural parameters.

In Study I, development of human figure drawings was assessed in 5 ½ year old preterm and term children. Preterm children, especially boys, showed delayed drawing skill. In whole group, stages in human figure drawing were predicted by visual perception, and proportion and circularity of human figures. In addition, developmental stage of human figure drawing explained a substantial proportion of performance IQ and overall motor ability. In Study II and III, handwriting skill was investigated in the non-involved hand of children with unilateral cerebral palsy (CP). They showed problems in size, alignment, shape, and spacing along with the readability of handwriting when compared to typically developing peers. Poor handwriting quality was related to decreased sensory-motor and cognitive functions. At follow-up after 16-months, they showed improvement in handwriting skill and in different visual-motor coordination tasks. Age and IQ explained the improvement in handwriting quality. All children with unilateral CP kept up their academic skills and passed to the next grade. In Study IV, children with Childhood Apraxia of Speech (CAS), who have motor deficit in speech, was investigated. The question was if they have a risk for global motor deficit, affecting more than the speech and non-speech oral motor function. Results confirmed that children with CAS had decreased performance in fine and overall motor functions and adaptive behaviour. Some relationship was found between motor control of drawing and speech/non-speech oral movements. Individual data however showed a very heterogeneous profile of problems in motor and behavioural domains.

To conclude, delayed development of graphomotor skills should be accommodated in educational settings with preterm children and children with unilateral CP. Intervention strategies should focus on facilitation of visual-motor coordination tasks. Children with CAS can benefit from a broader motor examination and follow-up for adaptive behavior in the clinic.

LIST OF PUBLICATIONS

I.   Tükel Ş, Eliasson AC, Böhm B, Smedler AC. What Can Children's Drawings of a Human Figure Tell Us About Cognitive and Fine Motor Development? (Manuscript)

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IV.   Tükel Ş, Björelius Hort H, Henningsson G, McAllister A, Eliasson AC. Motor Functions and Adaptive Behaviour in Children with Suspected Childhood Apraxia of Speech (Submitted).

CONTENTS

ABBREVIATIONS

ASHA	American Speech and Hearing Association
CAS	Childhood Apraxia of Speech
CP	Cerebral Palsy
DCD	Developmental Coordination Disorder
DSM-IV	Diagnostic and Statistical Manual of Mental Disorders- Fourth Edition
FSIQ	Full-Scale Intelligence Quotient
ICF-CY	International Classification of Functioning- Children and Youth version
mo	Month
MVPT	Motor-Free Visual Perception Test
PIQ	Performance Intelligence Quotient
SNP	Stockholm Neonatal Project
VIQ	Verbal Intelligence Quotient
VMI test	Visual-Motor Integration test
y	Year

1. INTRODUCTION

1.1. GENERAL INTRODUCTION

The motor behaviour we perform in everyday life arises from the interaction between many systems. The motor system controls the precision, direction, strength, and velocity of movements according to information provided by sensory systems regarding internal (body) and external (environment) states. Apart from sensory systems, cognitive processes including attention, motivation and planning are needed to be integrated with the motor system for the intentional movements.

The motor execution of graphics, such as letters, numbers, symbols, geometrical or representational figures, is called graphomotor skills. They enable a visual form of communication to share knowledge, ideas, and emotions and help the thinking and creative processes. Acquisition of graphomotor skills namely, drawing and handwriting, depends on effective interplay between perceptual-motor and cognitive systems. Visual-motor coordination is a foundational skill required for the accuracy and quality of graphomotor production during childhood. The coordination of such precise and continuous fine motor movements is highly demanding although in typical development, they seem to appear without any effort at expected ages.

Development and acquisition of graphomotor skills are highly challenging for children with neurological dysfunctions due to the problem in visual-motor coordination. Thus, the overall aim of this thesis is to investigate the development of visual-motor coordination required for graphomotor skills in relation to sensory-motor, cognitive and behavioural parameters in children with different neurological dysfunctions. This will be investigated in three paediatric populations: children born preterm, children with unilateral cerebral palsy (CP) and children with childhood apraxia of speech (CAS).

Following sections present visual-motor coordination and graphomotor skills first in typical development and then in mentioned paediatric populations.

1.2. DEVELOPMENT OF VISUAL-MOTOR COORDINATION IN FINE MOTOR SKILLS

The visual-motor coordination is the ability to match motor output with visual input. It refers to a complex process of integrating information from the visual and the motor systems in order to reach the optimum movement pattern that is visually accurate and cost efficient in terms of energy and time (Shumway-Cook and Woollacott 2001).

Visual-motor coordination in fine motor skills includes (a) motor processes, including the eye, head, and hand movements; (b) sensory processes, including the

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visual, vestibular, and somatosensory systems; (c) internal representations of sensory perception and action; and (d) higher-level processes for adaptive and anticipatory aspects of fine motor functions (Shumway-Cook and Woollacott 2001).

Studies of the development of visual-motor tasks showed an early coupling between visual and motor systems in infant's reaching behaviour. Infants reach towards objects more accurately when they locate object's position with vision, showing a clear effect of vision on movement control. Similarly, when accidentally touching an object, infants turned the eyes toward it, showing the importance of somatosensory information of the hand for the direction of the eye movements (Shumway-Cook and Woollacott 2001). The interplay between these systems changes during skill acquisition. Age, novelty and complexity of task, and amount of practice are important factors that change the role of visual system in a motor task (Newman, Atkinson et al. 2001; Shumway-Cook and Woollacott 2001; Von Hofsten and Rosander 2007). The following sections will describe the development of visual and motor systems in regard to graphomotor skills.

1.2.1. Development of visual perception

Visual perception enables us to perceive a wide array of visual qualities such as movement, depth, spatial relations, facial expressions, and the identity of objects. The development depends on physical maturation of the visual system, experience from actions, and an ability to process the information. The physical maturation of the visual system has been reported to be related to many factors, i.e., genetic coding, endogenous brain activity, external visual stimulation, and protected sleep cycles (Graven and Browne 2008).

Visual perception involves 'a number of related abilities that tend to be interdependent upon one another' (Kurtz, 2006, p. 33). Consequently it is difficult to explicitly distinguish one perceptual ability from another during development (Kurtz 2006). These abilities of visual perception can be categorized theoretically into six types: 1) spatial relationships that enable perception of the relative position of objects; 2) visual discrimination that enables the discrimination between the features of different objects, such as position, colour and shape; 3) figure ground that distinguishes an object from surrounding or background objects; 4) visual closure that identifies a whole figure when only fragments of the figure are presented; 5) visual memory that recognizes a stimulus item following a brief interval; 6) form constancy that constitutes the ability to recognize the dominant features of objects when they appear in different sizes, shadings, and/or textures (Frostig, Lefever et al. 1961; Colarusso and Hammill 1996; Reynolds, Pearson et al. 2002).

The infants typically can receive continuous stimuli from lines, patterns, movement, and different light intensities. This enables perception of edges and contours parsing the visual world into components. Infants start to perceive the colour when the

colour pathway begins to operate at 2 to 3 months of age. Spatial relationships have been shown as early as 3–4 months of age. Infants show sensitivity to above and below and left and right as long as the object is the same. By 9–10 months the spatial relation of 'between' takes place. Later with age, spatial relations are generalized to all objects, and more complex relations are perceived. Perception of depth or three-dimensional features depends on binocular information that is likely to appear with the development of stereopsis, from 4 months of age. Moreover, infants are shown to have size and shape constancy (Arterberry 2008). All these aspects of visual perception help object recognition and form representation. Such visual qualities are processed and connected in the brain by 'ventral visual stream' (Atkinson and Braddick 2011; Kravitz, Saleem et al. 2011).

Perception of motion -both in humans and in objects- has been examined in infants. Studies show some evidence that infants can recognize humans out of their motion characteristics (Bertenthal, Proffitt et al. 1984; Morita, Slaughter et al. 2012). When moving an object, the infant's attention has been shown to shift from the edges to the interior of the object. Thus, infants trace the motion trajectory to be able to interact with moving objects and humans. The motion perception in children has been shown to reach adult level around the age of 8 years (Boot, Pel et al. 2012). Visual perception of motion along with kinaesthetic perception play important role in mental representation of motion, which have been linked to motor planning and the development of motor skills in general (Caeyenberghs, Tsoupas et al. 2009). These links have been described with a brain network between occipital-parietal-frontal regions, so called 'dorsal visual stream' (Atkinson and Braddick 2011; Kravitz, Saleem et al. 2011).

To sum up, the visual qualities start to be perceived early in infancy, and this is the basis for further development of refined visual perception that is required for the identification of graphics. Appreciation of lines and angles, appreciation of size and relative size, and representation of relationships of parts to the whole are important visual perceptual factors considered relevant prerequisites for graphomotor skills (Miller 1986). By school age, children are expected to identify graphical symbols and comprehend their meanings.

1.2.2. Development of motor control

The motor control refers to coordinated movements and skilful actions that are planned and commanded by the central nervous system. The motor control of a movement depends on the interaction between the individual, the task, and the environment, and it is a result of perception, cognition, and action systems (Shumway-Cook and Woollacott 2001). The motor control system has exceptional abilities to find solutions to problems inherent in sensorimotor control, i.e., redundancy, noise, and delays. The brain uses different computational mechanisms to limit the effects of these problems and to perform skilful action (Franklin and Wolpert 2011).

Computational mechanisms are not mature in childhood, development depends on practice and experience in different contexts (Wolpert 2007; Franklin and Wolpert 2011). During the early phases of motor learning, movements are unskilled and highly feedback dependent, and they require strong demands on attention (Atkeson 1989). In this phase, the movement patterns vary substantially thereby this phase is called 'motor babbling' (Bullock, Grossberg et al. 1993; Guigon and Baraduc 2002). Feedback from sensory modalities reaches the motor system after the movement is executed, causing the 'motor babbling' in childhood. Delays depend on the particular sensory modality (e.g., longer for vision than proprioception) and perceptual complexity (e.g., longer for face recognition than motion perception) (Franklin and Wolpert 2011). It might be understood like: it is not until the line is drawn, it is possible for the child to perceive whether or not the line accurately corresponds to the intended drawing. It means that visual feedback is not yet used to continuously guide the movements. These temporal aspects of motor behaviour show great variability up to late teenage periods (Largo, Caflisch et al. 2001). Through practice, the children get more skilled, also accuracy and velocity of actions increase, whereas feedback processing becomes less important. Finally, the movement pattern becomes automatic in a skilful manner (Halsband and Lange 2006).

Drawing and handwriting are particular skills that require high-frequency, low-amplitude serial movements in fingers, hand, and arm. During development, after infant gains head control, hand movements are first controlled by the proximal joints (arm, shoulder) thereby first drawing movements have big range of motion with a very primitive grasp of the writing tool (Eliasson and Burtner 2008). When children gain control over isolated finger movements and in-hand manipulation, they are able to accomplish drawing and writing movements with a mature grasp of the writing tool. Movement trajectory during handwriting consists of a continuous flow of letter strokes up and down. Coordination of muscles in the hand and fingers is dependent on how the writing tool is grasped. In childhood, pencil grip postures develop from immature grips, that is, the whole hand grip using the palm and all fingers, to mature ones, that is, the dynamic tripod grip. The dynamic tripod grip is a precision grip that involves the tips of three fingers and the webbed space between the thumb and the index finger (Schneck and Henderson 1990). This grip posture enables the fingertip movements during a graphomotor task. Force control on the writing tool is also an important motor aspect that affects movement acceleration and velocity during drawing (Lange-Kuttner 1998). Also, different graphical forms have been related to different movement parameters in preschool age children; for example, pencil lifts are more common in vertical line drawing, pressure (force control) is higher in angular forms than in circular forms, and open forms take more time to draw than closed forms (Lange-Kuttner 1998). During childhood, mature grip positions can be seen in half of the children at 4 years of age and almost all the children at 7 years of age (Schneck and Henderson 1990). However, fine-tunings of

the mature grip pattern continue until the age of 10.5 years (Rueckriegel, Blankenburg et al. 2008).

1.2.3. Relationship between visual-motor coordination and cognitive functions

The ability to construct or reproduce spatial relationships in two or three dimensions -for example, drawing or block construction tasks- is named in psychology as visuo-spatial skill. Visuo-spatial skills have historically been chosen as an essential component of psychoeducational evaluations in children. Numerous visuo-spatial ability tests measure the visual perception and also the accuracy in the construction of visual models (Harris 1963; Frostig 1966); intelligence tests also contain subtests for the same purpose (Palisano and Dichter 1989). Visual-motor coordination skills have been found to be correlated with school readiness, and social-emotional functioning and academic skills.

Learning basic academic skills, such as writing, reading, spelling, and math, is commonly related to visuo-spatial skills. The ability to copy geometric/abstract patterns, for instance; is associated with learning handwriting, math, reading, and spelling skills in the first years of primary education (Weil and Amundson 1994; Daly, Kelley et al. 2003; Barnhardt, Borsting et al. 2005), even after controlling for intelligence (Taylor Kulp 1999). Visual ability in children with average intelligence is shown to relate to reading ability (Kulp and Schmidt 1996). Visual perception, motor skills, and visual-motor integration have been found to explain a substantial proportion of variance in mathematical skills (Pieters, Desoete et al. 2012). These results indicate the importance of visual perception and visual-motor coordination in hand as a foundation of development of academic skills.

1.3. GRAPHOMOTOR SKILLS

Graphics are visual presentations on some surface, such as a wall, canvas, screen, paper, or stone that are produced to inform, illustrate, or entertain. Gender (Brown 1990; Williams, Zolten et al. 1993; Coulbeau, Royer et al. 2008), culture (Cox 1993; Martlew and Connolly 1996), and formal school education (Martlew and Connolly 1996) have been shown to affect the development of graphomotor skills. Steps in graphomotor production include: 1) the ability to accurately interpret or give meaning to what is seen; 2) the ability to both store in memory and retrieve from memory graphical patterns; 3) the ability to ideate, plan, and execute the movement; 4) kinesthetic feedback; and 5) visual-motor coordination. In the present thesis, graphomotor skills in letter production and human figure drawing will be presented.

Handwriting and drawing skills are recognized as developmental functional skills in the International Classification of Functioning, Children and Youth version (ICF-CY) (2007). They are both included in the activity and participation section. Learning to write is included under the 'learning and applying knowledge' title and

is defined as 'developing the competence to produce symbols that represent sounds, words or phrases in order to convey meaning, such as spelling effectively and using correct grammar'. Drawing is included under the 'communication' title and is defined as 'conveying meaning by drawing, painting, sketching, and making diagrams, pictures or photographs, such as drawing a map to give someone directions to a location'.

1.3.1. Drawing skill

Drawing is an emerging childhood skill that serves not only as a representational communicative task but also supports literacy acquisition. It is also shown to be important in childhood for creativity, innovation, evaluation, and problem solving (Ainsworth, Prain et al. 2011; Robbins, Jane et al. 2011). Children use drawing or mark making to record personally or culturally significant images in their environment. They build up rich graphical representations in preschool ages, and during school they learn literacy and numeracy. Parents and teachers play important roles by giving feedback to children's graphical representations towards socially accepted patterns of graphical representations (Cox 1993).

Drawing development has roughly three stages: scribbles, pre-schematic stage (representational forms), and schematic stage (realistic drawings). Children start to scribble at around 2 years of age. It is initially a physical activity rather than an attempt at picture making. Around age 3, children start to notice recognizable shapes in their scribbles and relate them to the self, ideas, events, and people. For example, they notice the resemblance between their scribbles and noodles so that they label the picture as a 'noodle'. They create forms consciously for the first time in this stage, and these forms provide a tangible record of the children's thinking processes (Berk 2005). To be able to build the resemblance between drawings and real objects children must have symbolic thought -that pictures can serve as symbols for objects or humans or spoken language-. At about 18–24 months, according to Piaget, children begin to conceive of objects and events that are not immediately present. This ability to represent objects in one's mind is a crucial step on the path towards abstract symbolic thought. Two-year-olds have learned how to represent the world mentally but have not yet learned how to interrelate these representations in a coherent way (Piaget and Inhelder 1948; Cox 1993; Berk 2005).

A major milestone in drawing occurs when children begin to draw lines to represent the boundaries of objects. Usually the first recognizable objects are human figures, indicating the child's interest in people. A tadpole is a very common form of a first human figure attempt at 3–4 years of age. Four-years-olds add features into tadpole, such as eyes, nose, mouth, hair, fingers, and feet (Cox 1993).

Graphic communication begins in the pre-schematic stage when children can draw things important to them bigger or replace or colour them in unusual ways. Human figures are looking at the viewer and usually smiling. A conventional human figure

is built gradually by adding the body and other parts. Therefore, transitional figures are present at this stage. Between 7 and 9 years of age, the schematic stage takes place by advancements in perceptual, motor, and cognitive functions. Children represent more visual and spatial realism in their drawings. For example, people stand on a baseline and birds fly above, and colouring is correct. Also the schematic stage shows great individuality among children (Cox 1993). Three-dimensional drawing is a very advanced level that requires perspective and light. Perspective can be seen in drawing geometrical shapes, such as a cube or cylinder, most commonly after age 13 years, although children starting from the age of 7 draw differentiated figures when they are asked to draw a cube (Toomela 1999). Perspective and light in human figure drawings can develop later in life for those children who have an interest in drawing skills (Berk 2005).

1.3.2. Handwriting skill

Handwriting is the process of forming letters, figures, numbers, or other significant symbols purposefully onto a surface by using tools like a pen or pencil. Both legibility (readability) and speed are important parameters of handwriting, especially in a school setting. Factors such as shape, alignment, size, and the spacing between letters and words affect the legibility of handwriting. These factors as well as the speed of handwriting depend on visual-motor coordination.

Literacy skills, learning to read and write, are the major focus of the first three years of mainstream education. Handwriting has been shown to relate to other language-related abilities such as reading, spelling, and the composition of text (Abbott 1993; Berninger, Nielsen et al. 2008) Studies showed problems in handwriting could create a barrier to accomplish spelling and composition of well-structured text (Berninger 1991; Graham 2000; Case-Smith 2002; Feder and Majnemer 2007) because both motor execution and the generation of ideas during writing require attention at one time (Sweller J. 1994), and this competition between two different systems can result in making errors or low performance. Therefore, it is important to learn handwriting to be able to use it as a tool for higher-order cognitive functions.

Children with drawing experience, especially of geometrical shapes, have been known to learn handwriting more easily. In preschool ages, drawing geometrical shapes develops from simple to complex ones: vertical strokes (age 2 y), horizontal strokes (age 2 y 6 mo), circles (age 3 y), cross (age 4 y), square (age 5 y), and triangle (age 5 y 6 mo) (Beery and Buktenica 1989). Therefore, the Visual Motor Integration test (VMI) that consists of copying geometrical shapes has been widely used as an important indicator for learning to write. Apart from visual-motor coordination, motor planning, cognitive and perceptual skills, and accurate processing of tactile and kinaesthetic information have been indicated as prerequisites for handwriting skill acquisition (Van Hoorn, Maathuis et al. 2010).

Handwriting in typically developing children was found to develop quickly during grade one (age 6-7 y) and reach a plateau by grade two (age 7-8 y), and it becomes automatic, organized, and available as a tool to compose a text with further development in grade three (age 8-9 y) (Feder and Majnemer 2007). From grades 3 to 6, writing performance becomes gradually faster, more fluent, and more efficient. In the following years, a handwriting pattern develops in a personalized way. The maturation of movement parameters in handwriting strongly correlates with age and depends on the complexity of task (Rueckriegel, Blankenburg et al. 2008). It has been shown that straight writing movements are replaced by slightly more curved movements, probably corresponding with the natural curvature of stationary movements of the hand and fingers (Meulenbroek and Van Galen 1988). The graphic motor patterns in handwriting are visually guided in the early stages of learning (Jones D. 1999; Adi-Japha E. 2001), then they become more dependent on kinaesthetic feedback when visual and kinaesthetic senses are integrated. Finally, it becomes automatic (effortless and fast retrieval and production of legible letters without the need for attention) at early adolescence (Adi-Japha E. 2001).

The speed of handwriting, on the other hand, has been shown to have a more or less linear progression during primary school (Ziviani 1984; Graham and Weintraub 1996; Karlsdottir and Stefansson 2002). Studies in which the correlation between the legibility and speed of handwriting was examined have found either only weak positive correlation (Ziviani 1984; Karlsdottir and Stefansson 2002) or no significant relation (Graham and Weintraub 1996; Graham, Berninger et al. 1998). Weintraub and Graham, however, reported that an instruction to write neatly decreased the speed of handwriting (Weintraub and Graham 1998).

1.4. ATYPICAL VISUAL-MOTOR COORDINATION AND GRAPHOMOTOR SKILLS IN RELATION TO NEUROLOGICAL DYSFUNCTIONS

In the face of any neurological deficit, visual-motor coordination is likely to be affected, and the problem is commonly seen in graphomotor skills. The neurological symptoms affecting visual-motor coordination can be in motor control, the visual system, attention mechanisms, or memory. In developmental disorders, visual-motor problems appear in preschool and school ages as a dysfunctional drawing/handwriting that can accompany motor disorders and learning difficulties.

A deficit in handwriting skill is considered to be characteristic of certain developmental and also adulthood neurological disorders, such as Developmental Coordination Disorder (DCD) (2000), Parkinson's disease (Van Gemmert, Teulings et al. 1999), and Alzheimer's disease (Silveri, Corda et al. 2007). Poor writing or dysgraphia defines poor legibility and/or slow writing speed. In the Diagnostic and Statistical Manual of Mental Disorders- Fourth Edition (DSM-IV) (2000), dysgraphia is characterized as a learning disability in the category of written

expression, when one's writing skills are below those expected given a person's age, measured through intelligence and age-appropriate education.

Similarly, drawing skill is highly sensitive to neurodevelopmental delay and/or deficit, explaining its use in test batteries for identifying problems in children's general development and readiness for school or learning ability. There are a considerable number of tests that assess or use drawing ability in childhood, both in clinics and in research: Goodenough-Harris Draw-a-Person Test, Rey-Osterreith Complex Figure Test, the Visuo-Motor Integration Test, the Bender Gestalt Test, the Clock Drawing Test, and the Bicycle Drawing Test (Berk 2005). Among all the tests, the 'human figure drawing' test has been shown to have moderate correlation with Wechsler's Intelligence Scales for Children (WISC-R and WISC-III) (Abell, Wood et al. 2001). It has been used to detect learning difficulties (Plubrukarn and Theeramanoparp 2003), visual-motor problems (Schepers, Dekovic et al. 2012), and socio-emotional problems (Lim and Slaughter 2008).

Following sections present the characteristics and reported visual-motor coordination difficulties in three paediatric populations that those are of interest in this thesis.

1.4.1. Children born preterm

Preterm birth (defined as < 37 weeks gestation) is an important risk factor for major and minor neurodevelopmental disorders in addition to neonatal deaths. The prevalence of preterm birth in high-income countries like Sweden is about 4–5%. The cause of preterm birth is still unknown, but several risk factors have been presented -for instance; maternal age, smoking, infections, inflammation, stress, and multiple foetuses (Copper, Goldenberg et al. 1996; Andrews, Hauth et al. 2000; Blondel, Macfarlane et al. 2006; Hoffman, Jeffers et al. 2007).

Technological advances and improvements in perinatal protocols have increased the survival rates for infants born as early as 23 weeks. According to a study in the United States, each week in utero after week 23 raises survival chances by 6–9%, up to 95% by 33 weeks (Ward and Beachy 2003). Preterm births at 32–36 weeks' gestation are five times more common than births before 32 weeks' gestation. Severity of neurodevelopmental outcome is related to gestational age and birth weight. Immaturity of organs, particularly the brain, lungs, and cardiovascular system, leads to major disability within survivals (Saigal and Doyle 2008). Central nervous system haemorrhage and/or ischaemia, sensory impairments, and retinopathy of prematurity (ROP) are the neonatal complications that have a significant impact on long-term development and outcome (Ward and Beachy 2003). When we consider the overall preterm population, the rate of major disabilities, such as CP, mental retardation, and visual and auditory deficits, is fairly low and is more common in the most immature children (< 28 weeks gestation) (Saigal and Doyle 2008). However, mild to moderate developmental deficits have

been reported more frequently, including neurocognitive deficits, neuromotor dysfunction, and lower academic achievement (Goyen, Lui et al. 1998; Hadders-Algra 2002; Van Baar, Van Wassenaer et al. 2005; Marlow, Hennessy et al. 2007; Aarnoudse-Moens, Weisglas-Kuperus et al. 2009).

1.4.1.1. Visual-motor coordination children born preterm
Visual motor coordination has been one of the most commonly reported problems, and preterm cohorts in many different countries have been tested for this function. A recent meta-analysis has revealed that very preterm children, particularly boys, have deficits in visuo-spatial abilities and visual-motor integration (Geldof, van Wassenaer et al. 2012).

The visual system is affected in the face of prematurity. Apart from ROP, which can lead to eye diseases from myopia to severe situations like blindness, problems can be seen in visual acuity and the visual field because of the changes in the optic disc caused by white matter damage (Dutton and Jacobson 2001). Oculomotor control -smooth pursuit eye movements (Strand-Brodd, Ewald et al. 2011) and strabismus (Holmstrom, el Azazi et al. 1999) - has been shown to be affected in very preterm children. Studies about visual perception have reported somewhat different results according to which measurement tool has been used. Specific aspects, such as perception of line orientation, seem to be affected rather than broad indices of visual perception (Geldof, Van Wassenaer et al. 2012).

Extremely preterm children even without CP and with normal intelligence can present with gross and fine motor difficulties, including graphical skills like handwriting (Feder, Majnemer et al. 2005; Davis, Ford et al. 2007). In a series of works, Atkinson and Braddick have shown that preterm children have impaired visuo-spatial manipulation, that is, in block construction, and visual-motor control (Atkinson and Braddick 2007). They have explained that these impairments are result of problems in the brain network, the so-called dorsal stream, which is responsible for spatial function, selective attention, and executive control. White matter abnormalities, which are estimated to be present in 50–70% of very preterm and very low birth weight children (Volpe 2009) affect neural connectivity. Moreover, deficits in connectivity may hinder optimal signal conduction within the network for visual motor processes (Bourne 2010).

Intelligence measures of very preterm children are within normal range but still 0.5 to 1.0 SD lower than term peers (Bhutta, Cleves et al. 2002). Performance IQ (PIQ) is usually lower than verbal IQ (VIQ), and children do not have problems in expressive language skills. On the other hand, they are prone to have learning difficulties that affect their academic achievement. Difficulties have been frequently reported in writing output, arithmetic, and reading and have been associated with visuo-spatial and visual-motor ability (Grunau, Whitfield et al. 2002).

In this thesis, the drawing skill of preterm children enrolled in the Stockholm Neonatal Project (SNP) was reported. At 10 months of age, the SNP preterm cohort without major neurological complications performed significantly lower than term infants in visual-motor coordination (Gerner, Katz-Salamon et al. 1997). Problems in visual-motor coordination were found to persist at 5½ years of age. And visual-motor skill was found to be related to PIQ, hyperactive behaviour, and inattention (Böhm, Lundequist et al. 2010). Human figure drawing is a projective visual-motor task that has been shown to correlate with cognitive functions in childhood. It is sensitive to developmental delays but has not been investigated in preterm children. Moreover, how visual-motor control and visual perception are involved in the human figure drawing task is not known.

1.4.2. Children with unilateral Cerebral Palsy

Cerebral palsy is one of the most common diseases causing physical disability in childhood. By definition, 'cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeletal problems' (Rosenbaum, Paneth et al. 2007). The largest epidemiological risk factors for CP are preterm birth, intrauterine growth restriction, perinatal infection, and multiple births (O'Callaghan, MacLennan et al. 2011). Gestational age is inversely related to the incidence rate of CP (Himmelmann, Hagberg et al. 2010). Neuroimaging and neuropathology indicate the importance of white matter disorders and of ischaemic stroke in CP; birth asphyxia, congenital malformations, placental pathology, and genetic variants also contribute to CP risk (Nelson and Chang 2008).

The prevalence of CP in Turkey is higher (4.4 per 1,000 live births) (Serdaroglu, Cansu et al. 2006) than that in developed countries; for instance, it is two times less in Sweden (2.2 per 1,000 live births) (Himmelmann, Hagberg et al. 2010). An increased level of obstetric and neonatal problems and other factors such as genetic malformations result in higher prevalence (Serdaroglu, Cansu et al. 2006). The Surveillance of Cerebral Palsy in Europe (SCPE) (2000) classifies CP according to type of motor symptoms (spastic, ataxic, and dyskinetic) and the location of major impairment (unilateral or bilateral). Spastic CP is the most common type and characterized by abnormal patterns of posture and/or movement, increased muscle tone, and pathological reflexes. In Turkey, 28% of CP cases have unilateral or hemiplegic spastic CP, where symptoms are mainly seen in one side of the body (Serdaroglu, Cansu et al. 2006).

1.4.2.1. Visual-motor coordination in children with unilateral CP
Children with unilateral CP use their non-involved hand in many daily activities. The non-involved hand is, in most cases, the dominant side (independent of right or

left) that initiates and directs the action in daily life. Children with unilateral CP have by definition no or subtle motor impairment in the non-involved hand. Fingertip force coordination in the non-involved hand (Gordon, Charles et al. 1999) and movement planning (Steenbergen and Gordon 2006) have been found to be lower than in typically developing children.

Children with unilateral CP have problems in visual perception and visuo-spatial skills (Barca, Cappelli et al. 2010). Findings also have indicated the poor integration of visual and proprioceptive senses; for example, a study showed that they couldn't differentiate whether object movement is the result of their own motor actions or an external source (Ritterband-Rosenbaum, Christensen et al. 2011). Visual acuity is generally normal, but abnormalities are found in visual fields and visual attention and include optokinetic nystagmus (e.g., involuntary eye movements) (Mercuri, Guzzetta et al. 2008). On the other hand, visual-motor coordination in the non-involved hand has not been investigated.

Children with unilateral CP are generally included in the regular educational system in Turkey. It means that they are expected to learn handwriting in the first two years of school education, like their peers. Language functions are usually normal in children with unilateral CP; they score in the average or low average range in VIQ (Goodman and Yude 1996; Sigurdardottir, Eiriksdottir et al. 2008). However, PIQ is usually 10 or more points lower than VIQ, indicating that the brain plasticity favours language skills over visuo-spatial skills (Goodman and Yude 1996; Goodman 1997). It has been known that children with delayed development and brain lesions devote more time and energy to write legibly (Berninger and Rutberg 1992), try to complete a written assignment in as few words as possible, or cannot finish it on time (Levine, Oberklaid et al. 1981; McHale and Cermak 1992). Handwriting difficulties were reported in a majority of children with unilateral CP (DuBois, Klemm et al. 2004). Considering the negative effects of handwriting problem on spelling and composition of creative and well-structured text (Christensen 2005), and on self-esteem and motivation (McKinlay 1978), children with unilateral CP might fail to make the expected academic progress. Indeed, low educational attainment has been shown for individuals with CP (Bottos, Feliciangeli et al. 2001). Therefore, investigation of handwriting problem and its relation to sensory-motor and cognitive functions are important for the inclusion of children with unilateral CP into mainstream school education.

1.4.3. Children with Childhood Apraxia of Speech

Childhood apraxia of speech is a speech motor disorder. Children with CAS have impairment in articulation and prosody that results in unclear speech. The impairment could cause communication and behavioural problems in everyday life.

According to the definition of the American Speech and Hearing Association (ASHA) (2007), 'Childhood apraxia of speech is a neurological childhood

(pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (e.g., abnormal reflexes, abnormal tone). CAS may occur as a result of known neurological impairment, in association with complex neurobehavioral disorders of known or unknown origin, or as an idiopathic neurogenic speech sound disorder. The core impairment in planning and/or programming spatiotemporal parameters of movement sequences results in errors in speech sound production and prosody'.

Based on clinical data, the prevalence estimate of CAS is 1–2 per 1,000 (Shriberg, Aram et al. 1997). Aetiology is largely unknown, but a familial link has been reported for some cases (Lewis, Freebairn et al. 2004). MRI findings have been reported to be normal for most children with CAS (Liegeois and Morgan 2012). Subtle abnormalities in brain structure or functional anomalies at the metabolic or transmitter level have been suggested as the basis of CAS (Liegeois and Morgan 2012)

1.4.3.1. *Visual-motor coordination in children with CAS*
Several studies show that speech-language disorders and motor disorders frequently are seen together (Hill 2001; Visscher, Houwen et al. 2007); for example, DCD is a very common diagnosis in children with developmental speech and language disorders and vice versa (Visser 2003; Gaines and Missiuna 2007). Diagnostic criteria of CAS have recently been identified; and it has been differentiated from other speech and language disorders as a motor originated disorder.

General motor ability in children with CAS has not been investigated systematically. There are limited number of studies reporting problems for instance, in visual-motor coordination and in the accuracy and speed of grasping and manipulating (Bradford and Dodd 1996; Newmeyer, Grether et al. 2007). Above all, 'learning to write' and 'fine hand use' have been reported as problems by parents (Teverovsky, Bickel et al. 2009). Nothing is known about visual perception or visuo-spatial skills in this population. Since phonological and language problems have been reported in children with CAS, problem in 'learning to write' is attributed to language difficulties, disregarding the reported visual motor problem.

Even though our primary aim in this population was to find out whether or not visual-motor coordination is affected, we needed to present a whole motor profile because it has not been investigated systematically before.

2.   AIM OF THE THESIS

The general aim of this thesis is to investigate the development of visual-motor coordination required for graphomotor skills in different paediatric populations in relation to sensory-motor, cognitive and behavioural parameters. The detailed aims of studies are outlined below:

- To investigate drawing skill in 5½ year old preterm and term children in relation to motor function, visual perception and cognition.
- To examine handwriting skill in children with unilateral CP compared to typically developing children in relation to sensory, perceptual, motor and cognitive functions.
- To follow-up the development of handwriting, fine motor skills and school marks of a previously studied group of children with unilateral CP and, relate the performance in handwriting skill to age and IQ.
- To profile and interrelate different motor functions and adaptive behaviour in children with CAS.

3. METHODS

3.1. STUDY OUTLINES

All the studies in this thesis are observational studies either with cross-sectional design or with longitudinal design (Table 1). Cross-sectional design was used to investigate association of different parameters with visual-motor coordination. Longitudinal design was used to better explain the associations.

Table 1 Design, sample and materials

Study	Design	Sample (n)	Assessments*
I	Cross sectional design Case-control	Preterm (n=176) Term (n=125)	Human figure drawing M-ABC VMI MVPT WPIIS-R PIQ ImageJ
II	Cross-sectional design Case-control	CP (n=26) Control (n=32)	MHA BOTMP SCSIT LOTCA MACS Pencil grip position
III	Longitudinal design Single group	CP (n=16)	MHA BOTMP WISC-R School mark
IV	Cross-sectional design Single group	CAS (n=18)	VMPAC BOT-2 ABAS

* Abbreviations of assessments are defined in Chapter 3.3.

In Study I, a cross- sectional survey was carried out in preterm cases compared with term controls. Development of human figure drawing was the main outcome and it was associated with gender, visual perception, motor and cognitive functions.

Study II has also a cross-sectional design; cases with unilateral CP and controls compared. Handwriting skill was the main outcome and it was related to sensory-perceptual-motor and cognitive functions.

Longitudinal design was used in Study III to follow up the cases in Study II on handwriting, fine motor skills and academic performance after 16 months. Development of handwriting skill was investigated in relation to cognition and age.

In Study IV, a cross-sectional design was used to profile the development of speech, non-speech oral, manual and overall body motor functions and also adaptive behaviour in children with suspected CAS. Norm-referenced tests were used in order to provide performance corrected for age

3.2. PARTICIPANTS

Table 2 Characteristics of participants in the studies

| Study | Population | Gender | | Age | | |
		Girls	Boys	Min-Max	Mean	SD
I	Preterm	92	84	5y 6mo		
	Term	60	65	5y 6mo		
II	Unilateral CP	11	15	7y 5mo-12y 4mo	9y 7mo	1y 3mo
	Control	16	16	6y 8mo-11y 6mo	8y 11mo	1y 2mo
III	Unilateral CP*	5	11	8y 9mo-13y 7mo	11y 4mo	1y 6mo
IV	CAS	5	13	4y 5mo -10y 7mo	7y 1mo	1y 11mo

* Individuals who participated in to the Study III were also included in Study II.

3.2.1. Participants and data collection in Study I

The database of a population-based longitudinal project, the SNP, was used. SNP consists of all preterm children (gestational age of <37 weeks with a very low birth weight $\leq$ 1500g, with a need of mechanical ventilation), who were born between 1988 and 1993 in Karolinska or in St. Göran Hospital. The data used in this study was collected between 1994 and 1998 when children were 5 ½ years old (n=182). Age-matched healthy children born at term at the same hospitals were recruited as a control group (n=125). The number of controls was calculated to obtain a maximal power for comparisons with the preterm group. All children who performed drawing task were included (n=301). Characteristics of participants are presented in Table 2. According to the purpose of this study cognitive and motor tests were selected from the SNP database and applied to the children's drawings of human figures. After assessing 301 human figure drawings according to developmental stages, 40 human figure drawings were selected by stratified sampling among tadpole, transitional, and conventional stages (for descriptions of stages see Table 4). Then selected drawings were analysed for underlying visual-motor parameters in scientific image analysis program, ImageJ (2012).

3.2.2. Participants and data collection in Studies II and III

Data collection in Studies II and III was conducted in Ankara, Turkey. In total, 26 children who had been diagnosed with left unilateral CP (right-hand dominant) were included in Study II (Table 2). According to the Manual Ability Classification System (MACS), nine children were on Level 1, twelve were on Level 2, and five were on Level 3. Children were tested at Paediatric Rehabilitation Research Unit of Hacettepe University. Children were attending regular or special education classes at mainstream schools. The control group of 32 typically developing right-handed children was recruited from, and tested at, state primary schools. In Study III, a follow-up was performed with 16 children with unilateral CP, who were reassessed 16 months later (Table 2). One child did not want to participate and eight children could not be contacted because of changing contact information. During the follow-up period, none of them received any treatment regarding handwriting difficulties but had continued their school education.

3.2.3. Participants in Study IV

A group of 18 children with suspected CAS was selected from the Speech and Language Pathology Department of Karolinska University Hospital, Huddinge (Table 2). Inclusion criterion was Swedish as the first language. Exclusion criteria were the presence of coexisting diagnoses, other neurological diseases, receptive language impairment, diagnosed or suspected cognitive impairment, and oral or manual structural anomalies. Children received or had been receiving speech therapy during the study, but none of them had received physiotherapy or occupational therapy.

3.3. ASSESSMENTS

Overall clinical tests, assessments of drawing and handwriting skills, an image-processing software, school marks and a classification of pencil grip positions were used in this thesis (Table 1). Clinical tests used in studies can be categorized into four major domains; 1) motor functions, 2) cognitive functions, 3) sensory perception, and 4) adaptive behaviour (Table 3). For assessing the same domain, different tests were used according to availability. Description and usage of the tests, such as whole test or selected subtests, are explained briefly below.

Table 3 The overview of the tests in alphabetical order. The major performance domain(s) assessed by each test is presented.

		ABAS-2	BOTMP	BOT-2	LOTCA	M-ABC	MACS	MVPT	SCSIT	VMI	VMPAC	WISC-R	WPPSI-R
Motor	Gross		X	X		X							
	Manual		X	X		X				X			
	Speech										X		
Cognition					X							X	X
Sensory Perception	Visual							X	X				
	Tactile								X				
	Kinaesthesia								X				
Adaptive Behaviour		X					X						

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3.3.1. Assessments of sensory, motor, and cognitive functions and adaptive behaviour

Tests are listed in alphabetical order.

3.3.1.1. Adaptive Behavior Assessment System, Second Edition (ABAS-2)
The Swedish version of ABAS-2 was used to assess conceptual, social and practical skills that children learn in order to function in everyday life (Harrison PL and Oakland T 2008). It provides Swedish normative data and has well-established psychometric properties. It is based on parents' reports. Standard score was used.

3.3.1.2. Bruininks-Oseretsky Test of Motor Proficiency (BOTMP)
Selected items from BOTMP test was used to assess manual motor skills (Bruininks 1978). These items were: bilateral coordination, coordination of visual tracking, visuomotor response speed, copying a triangle to coordinate precise visual-motor movements, manual dexterity and drawing vertical lines. Raw scores were used.

3.3.1.3. Bruininks & Oseretsky Test of Motor Proficiency, Second Edition (BOT-2)
The BOT-2 short form was used to screen for overall body motor proficiency including both gross and fine motor functions. Furthermore, all manual motor subtests from the full version of BOT-2 were used to test for hand motor functions in detail (Bruininks and Bruininks 2005). The manual subtests are: fine motor precision, fine motor integration, manual dexterity, upper-limb coordination. Standard scores were used for both BOT-2 short form and manual motor subtests.

3.3.1.4. Developmental Test of Visual Motor Integration (VMI)
VMI was used to test visual motor skills (Beery and Buktenica 1989). It consists of abstract figures in a test booklet, and the graphic responses are scored using the criteria listed in the manual. Each shape is awarded either a 1 for passing or a 0 for failing. Raw score was used.

3.3.1.5. The Loewenstein Occupational Therapy Cognitive Assessment (LOTCA)
LOTCA is a series of tests that evaluates cognitive abilities in four areas: orientation, visual and spatial perception, visual motor organization and thinking operations (Itzkovich, Elazar et al. 1993). Selected subtests were used to assess visual and spatial perception (subtests: overlapping figures, object constancy and spatial perception), motor planning ability (praxis subtest), visual-motor organization (subtests: copying geometric forms, reproduction of a 2-dimensional, drawing a clock) and thinking operations. Raw scores were used.

3.3.1.6. Manual Ability Classification System (MACS)
MACS was used to classify manual ability of children with unilateral CP in daily life (Eliasson, Krumlinde-Sundholm et al. 2006). It has five different levels and children with unilateral CP typically appear on level I or II and some in level III (Arner, Eliasson et al. 2008).

3.3.1.7. Motor-Free Visual Perception Test (MVPT)

The whole battery of MVPT was used to assess visual perceptual ability with no motor involvement needed to make a response (Colarusso and Hammill 1996). Visual perception is measured in five categories: spatial relationship, visual closure, visual discrimination, visual memory and figure-ground. Total raw score was used.

3.3.1.8. Movement ABC (M-ABC)

The whole battery was used to identify and assess problems in children's motor development (Henderson and Sugden 1996). There are three main areas: hand motor skills, ball skills, and static and dynamic balance. Total raw score ranging between 0 (very good) and 40 (extremely impaired) was used.

3.3.1.9. The Wechsler Intelligence Scale for Children- Revised (WISC-R)

The WISC-R was used to measure cognitive ability in Turkish children since it was the only intelligence test with Turkish standardization (Savasır and Sahin 1995). The scale has acceptable reliability (0.98, 0.98 and 0.96, respectively for Full Scale, Verbal and Performance) and validity. Full-scale intelligence quotient (FSIQ), standard score was used.

3.3.1.10. The Wechsler Preschool and Primary Scale of Intelligence- Revised (WPPSI-R)

Performance intelligence quotient (PIQ) score of WPPSI-R, standardized Swedish version with Swedish norms, was used to assess the children's intellectual functioning in performance cognitive domains (Wechsler 1999). Standard score was used.

3.3.1.11. The Southern California Sensory Integration Tests (Ayres SCSIT)

The design copying, graphesthaesia, and kinaesthesia subtests of the Ayres SCSIT were used (Ayres 1972). Design copying was another visual-motor task that requires drawing complex figures by connecting the dots. Graphesthaesia is a test for tactile perception and kinaesthesia was used for the perception of hand movement. Raw scores were used.

3.3.1.12. Verbal Motor Production Assessment for Children (VMPAC)

VMPAC assesses non-speech and speech oromotor control in children aged 3-12 (Hayden A and Square P 1999). It consists of linguistically meaningful and non-meaningful vocal production tasks with good psychometric properties for English-speaking children. The selected subtests (focal oromotor control, sequencing maintenance control, connected speech and language control) of VMPAC were used in Study IV. Linguistically meaningful items of VMPAC were adapted to Swedish. Validity and reliability of adapted items gave satisfactory results. Typically developing Swedish children performed similar to the norms in VMPAC for English-speaking children. The final scores are based on a percentage level (0-100). Z-scores were converted and used in the study.

3.3.2. Assessments of drawing and handwriting

Measurements of drawing and handwriting skills were main outcomes in the first three studies. Assessments used for drawing and handwriting were described in detail in the manuscripts. Consequently, only an outline is presented here.

3.3.2.1. *Drawing of a human figure*

Children's human figure drawings were assessed in three different ways; (1) according to developmental stage (Table 4), (2) according to drawing order of body parts (top-down, centre-periphery, bottom-up), (3) measurement of visual-motor parameters (size, symmetry, proportion, circularity) by using a computer program for image analysis, ImageJ (2012).

3.3.2.2. *Minnesota Handwriting Assessment (MHA)*

MHA measures the (1) speed and (2) quality categories of handwriting (Figure 1) (Reisman 1999). The Turkish version (Bumin and Tükel 2006) was used to assess manuscript writing of children. For speed, the number of letters that are copied in 1 min was taken. The quality categories are legibility, form, size, alignment and spacing (range: 0-34 for each). There is no composite score for quality in MHA. The total sum for handwriting quality was calculated by conducting necessary statistical analyses, such as the uni-dimensionality of quality categories (range: 0-170).
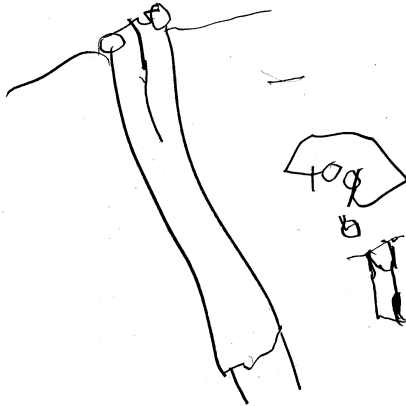
3.3.3. School marks
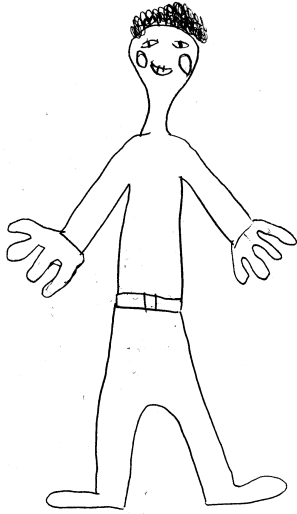
Children's school marks were used in order to assess academic achievement (Table 1). School marks show the general average of academic skills that range between 0-5, where an average of 2.5 is necessary to be promoted to the next grade.

3.3.4. Pencil grip positions

Pencil grip posture was recorded while the children were drawing and writing, and categorised according to a classification system (Schneck and Henderson 1990) (Table 1). All pencil grip positions were then divided into mature and immature grips.

Table 4 Scoring criteria for developmental stage of human figure drawings. Figure examples are from SNP database material in Study I.

STAGE	CRITERIA
Scribble 	• Nothing recognizable • Continuous circular lines • Child does not describe what is drawn
Pre-tadpole 	• Drawing but cannot identify the picture • Separation of different scribbles • Face only • Face and feet are made up of one line
Tadpole 	• Head, arms and/or legs present • Arms/legs protrude from the head • No head/body closure but face, arms, and legs present • Sun-like with arms and legs

STAGE	CRITERIA
Transitional	• Arms/legs are not protruding from the head • A line for body • Arms are connected to legs or body • No separation of head and body • Stick-man picture, sticks are used for all body parts except head • Body outline but no face • Only head and body, no arms/legs • No neck • Body outline is distorted, no order in space
Conventional-I	• Body outline: 4 parts; head, body, 2 arms, 2 separate legs • At least head and body have to be 2 dimensional • Segmented body parts • Arms are protruding from the body not head
Conventional-II	• Body parts are integrated, continuation of each other • 2-dimensional arms, legs, neck, head and body • Fingers and toes can be drawn with sticks • Has to be a face • Drawings of profile view

A

B

Figure 1 Minnesota Handwriting Assessment, Turkish version. A) Example of high performance on MHA: the letter in a circle is for calculation of handwriting speed. B) Example of low performance on MHA: shape, size, alignment and spacing of letters are inconsistent and some letters are illegible. For instance instead of writing 'z' in 'boğaz', the child writes 'w', a letter that does not exist in Turkish alphabet.

3.4. STATISTICAL ANALYSIS

Parametric statistics were used when assumptions, such as normal distribution, homogeneity of variance, and independent errors, were fulfilled in order to produce more accurate and precise estimates (Kirkwood and Sterne 2003). Otherwise, non-parametric statistics were used. Statistical tests were chosen according to design and sample size in the studies (Table 5). In all studies significance level was set to $p<0.05$. In Studies II and IV, multiple correlation tests were run on one set of data. However, due to small sample size in these studies alpha level was not corrected for the risk of Type 1 error to avoid risk of Type 2 error. Statistical programs SPSS and SPSS-AMOS were used to conduct all data analyses.

Table 5 Statistical tests used in studies.

Statistical tests	Study I	Study II	Study III	Study IV
Parametric Tests				
Pearson product moment correlation	X			X
Chi-square test	X			
Multiple linear regression analysis			X	
Stepwise regression analysis	X			
Path analysis	X			
Factor analysis (PCA)			X	
Non-parametric tests				
Mann – Whitney U Test		X	X	
Fisher's exact test	X			
Spearman correlation		X		
Wilcoxon signed-rank test			X	X
Friedman test				X

4. SHORT SUMMARY OF RESULTS AND CONCLUSION

4.1. STUDY I

Assessment of 301 human figure drawings revealed six developmental stages at 5½ years of age; scribbles, pre-tadpoles, tadpoles, transitional forms and two types of conventional figures. The most common stages were transitional and conventional stages. Computerized measurement of visual-motor parameters was carried out in 40 drawings, which are equally distributed among tadpole, transitional, conventional stages I, and II. Then, stepwise regression analysis was done to explain the developmental stage from computed visual-motor parameters, MVPT and VMI. Visual-motor control in proportionate and circular drawing movements and MVPT explained 60% of the variance in drawing development. Symmetry and VMI test, although correlating with developmental stage, did not explain any unique variance in drawing development. It means that development of human figure drawing depends on visual perception and visual-motor control in proportionate and circular movements.

When comparing preterm and term groups, especially preterm boys showed a delay in drawing development. They were significantly more represented in stages that are commonly seen before 5½ years of age, i.e. scribbles, pre-tadpoles and tadpoles (Figure 2). Preterm boys also showed immature pencil grip positions more often than other groups. Our findings in preterm boys indicate developmental delay in drawing skill probably because of underlying problems in visual perception and visual-motor coordination.
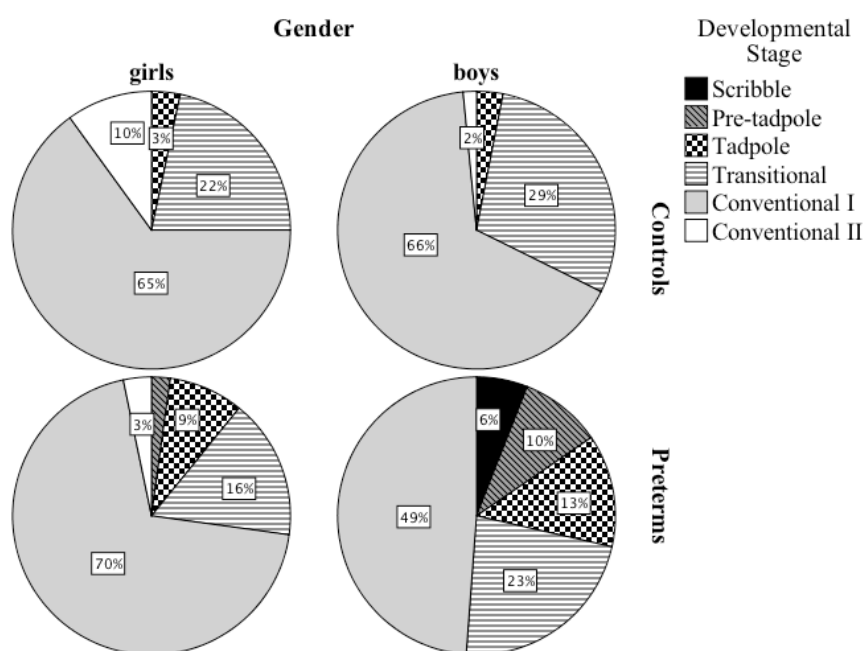
Figure 2 Developmental stages of human figure drawings in gender and gestational age groups.

Path analysis was carried out between gender, gestational age group, developmental stage and PIQ, and M-ABC. It showed that both gender and gestational age group had direct effects on developmental stage and explained 11% of the variance. This finding also confirmed that preterm boys had developmental delay in drawing skill. Developmental stage of drawing explained 39% of the variance in PIQ, whereas gestational age group and gender were not significant (Figure 3-A). It means that gender and gestational age group showed indirect effect on PIQ due to their effects on developmental stage. In M-ABC, developmental stage and gestational age group together explained 29% of variance (Figure 3-B). Similarly, gender showed an indirect effect on M-ABC. Drawing order of body parts was also assessed but no relationship was found between drawing order and gender, gestational age group, PIQ or M-ABC. These results show that developmental stage of human figure drawing can explain substantial proportion of PIQ and M-ABC since it is closely related to visual perception and visual-motor control.

A
B
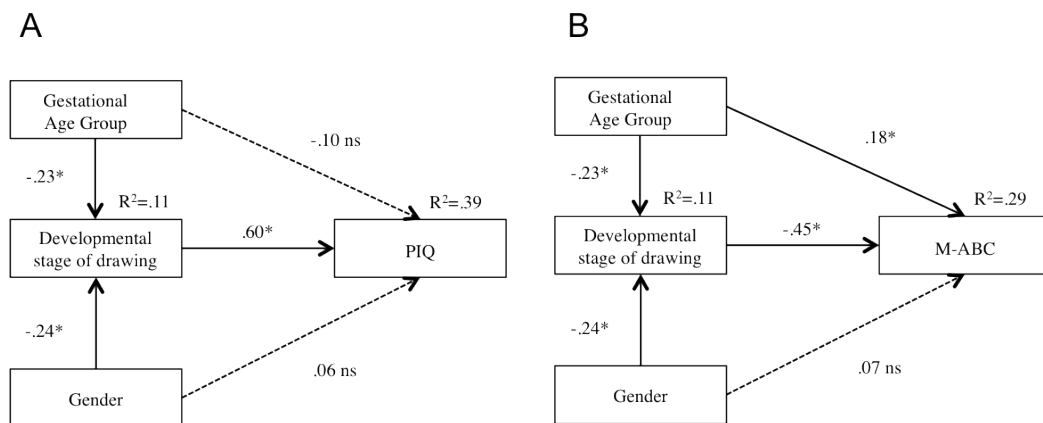
Figure 3 Path diagrams and results of the path analysis providing predictive value of drawing developmental stage. (A) Path analysis to explain PIQ from developmental stage of drawing, gestational age age group and gender. (B) Path analysis to explain M-ABC from developmental stage of drawing, gestational age age group and gender. Arrow direction shows the outcome variable. R^2 value for each outcome variable is presented above the box. Standardized Beta (β) value for each explanatory variable is presented on arrows. Solid line represents a significant thereby direct effect; dotted line represents non-significant effect.

4.2. STUDY II

The quality of handwriting skill in children with unilateral CP, using the non-involved hand, was lower compared to that of their typically developing peers, whereas speed of handwriting was the same in both groups (Figure 4). Children with unilateral CP performed lower levels in sensory and motor functions even using the non-involved right hand. Cognitive functions in children with unilateral CP were also significantly lower than the control group. The control group reached the ceiling effect of the handwriting test in all quality parameters. In both groups quality of handwriting was correlated with sensory, perceptual, fine motor and cognitive tests. Whereas handwriting speed was correlated with almost only fine

motor functions. Correlations were usually higher in children with unilateral CP than in controls. Results indicated that children with unilateral CP had decreased quality of handwriting skill. In addition, decreased quality of handwriting skill was related to problems in sensory, fine motor and cognitive functions.
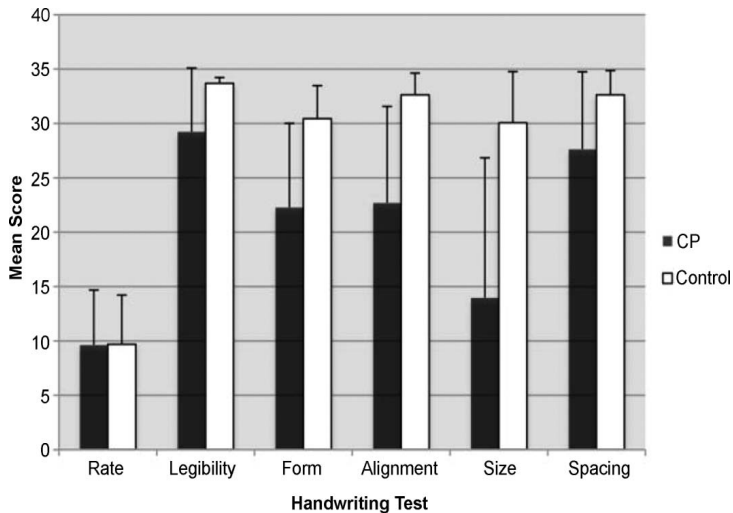
Figure 3 Comparison of speed and quality parameters of handwriting in children with unilateral CP and control group of typically developing children. All quality parameters (legibility, form, alignment, size, and spacing) were significantly lower in CP group while control group reached the ceiling effect. Handwriting rate was similar between the groups.

4.3. STUDY III

Children with unilateral CP in Study II were reassessed after 16 months at handwriting, fine motor skills and school performance. They showed significant improvement in quality composite and speed of handwriting, also in fine motor skills (especially visual motor coordination items). At individual level, difference in handwriting quality varied substantially in relation to IQ, medical condition or school leave (Figure 5). Handwriting speed, on the other hand, was not related to any factors.
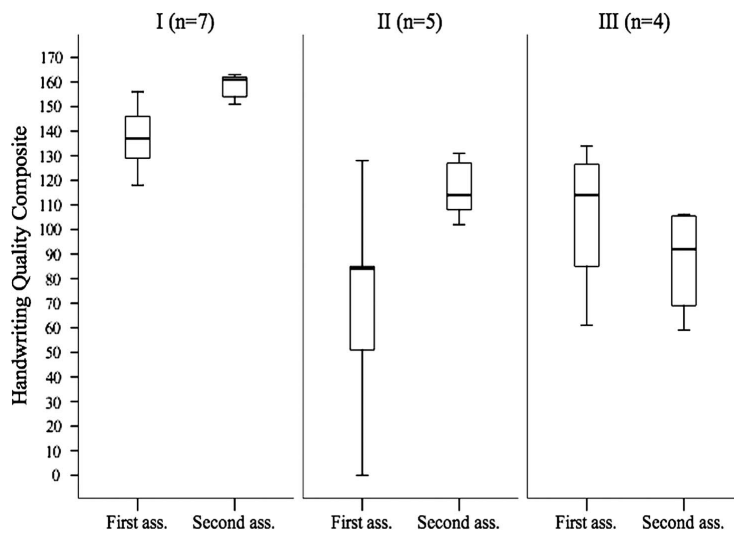
Figure 4 Change in handwriting quality composite at 16-month follow-up. Handwriting quality composite scores in the first and in the second assessments for three different groups. Box-plots show median, interquartile range and outliers. I= Children with FSIQ > 70, II= Children with FSIQ < 70, III= Children either with reported epilepsy seizure or with school leave.

Age and IQ predicted development in handwriting quality (R^2= .76, Figure 6). The biggest improvement was seen in younger children and children with lower IQ scores. Children with higher IQ scores showed smaller improvement since they already at the first assessment had achieved a better quality of handwriting skill. Findings indicated that low IQ level resulted in a slower development of handwriting quality. Children kept up their school marks and all were promoted to the next grade.
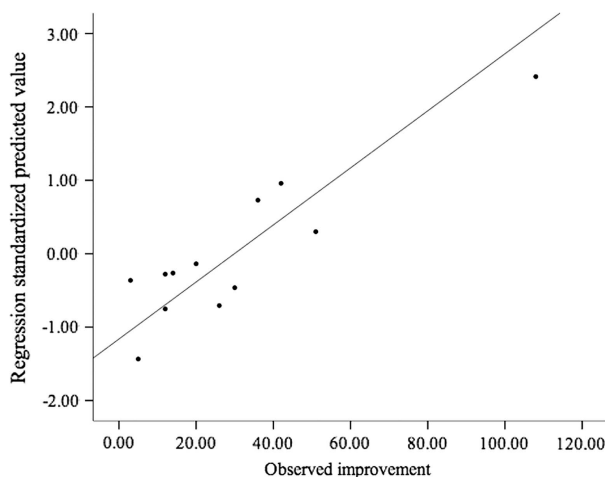
Figure 5 Accuracy of the regression model is shown. Scatter plot shows the correlation between observed improvement in handwriting quality and predicted values that are calculated via regression equation (Pearson r=0.89, p<0.05). Regression equation [Improvement =298.83 - (1.48 x Age-month) - (0.88 x FSIQ)].

4.4. STUDY IV

Median of speech and non-speech oral motor functions was very low (within -2 SD) in children with suspected CAS. Manual and overall body motor functions and adaptive behaviour were affected to a lesser degree (median within -1 SD for both).

Most of the correlations between speech, non-speech oral, manual and overall body motor functions were not significant. Only significant correlations were found between fine motor integration subtest (e.g. drawing abstract figures) and subtests focal oromotor control (r = 0.54) and connected speech and language control (r = 0.51). These relationships were considered to be meaningful since similar correlation had been shown earlier by another study. Between motor functions and adaptive behaviour domains though there was no meaningful relationship.

At individual level, co-occurrence of low performance (below -1 SD) at different motor functions (figure 7-A) and adaptive behaviour (figure 7-B) was very common despite weak correlations. However, there was no trend for the type of motor or behavioural difficulty. Children showed varied types and also numbers of difficulties.

Our results showed that children with CAS were low performers in manual and overall body motor functions suggesting a global motor deficit. Individual performance showed a very heterogeneous profile and highlighted the children with multiple problems.
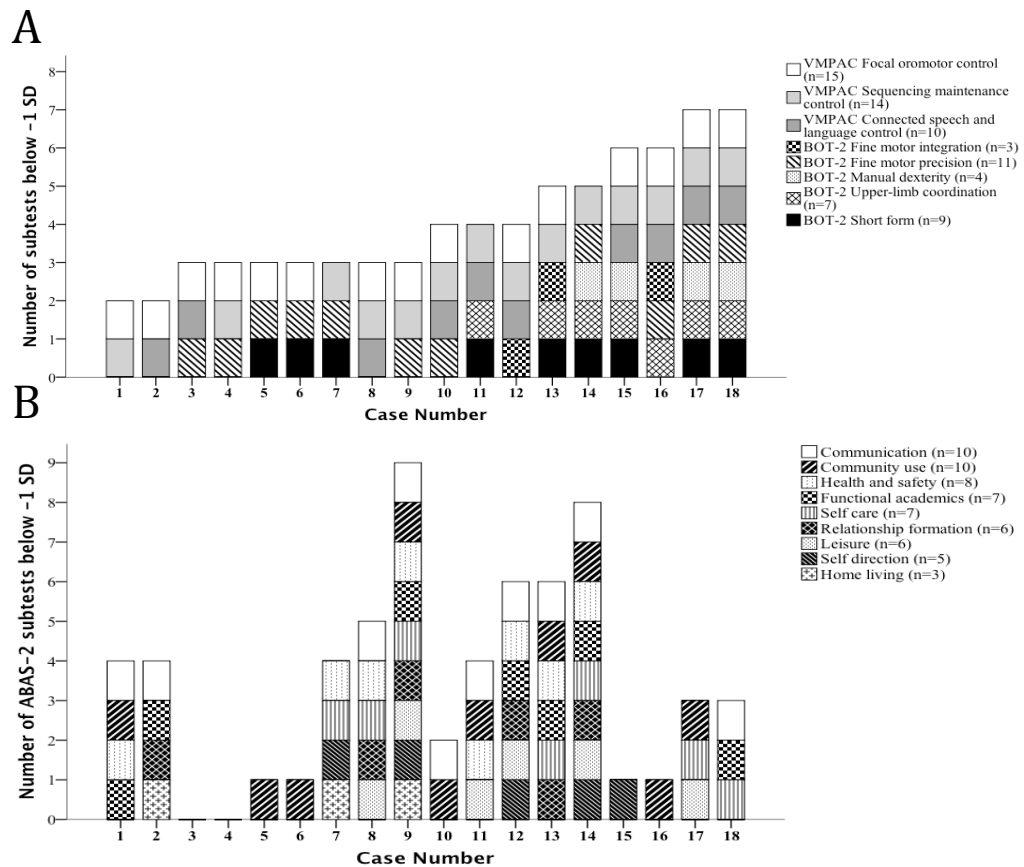
Figure 6 Individual performance is shown on different tests. Order of the participants is according to the number of motor problems (VMPAC+BOT-2) and is the same in all figures. (A) Number of subtests that were performed below -1 SD in BOT-2 short-form and manual motor subtests and VMPAC subtests for each child (i.e. figure shows co-occurrence of manual and speech/non-speech motor problems for the individual children). (B) Number of ABAS-2 subtests that were below -1 SD for each participant, figure shows the variation of behavioural problems. Numbers in brackets show number of children below -1 SD in each subtest in both figures.

5.   DISCUSSION

Paediatric populations included in this thesis have different characteristics, severity and type of motor dysfunctions. One important research question was to further explore if development of visual-motor coordination required for graphomotor skills follows the average age-expected trajectory or not. The studies were carried out differently because of the differences between the paediatric populations. For example, the existing knowledge about the motor dysfunction in CAS is very limited compared to the existing knowledge about the motor dysfunction in CP and preterm children. Discussion below is therefore outlined separately for each population.

5.1. CHILDREN BORN PRETERM

In Western countries, typically developing children show distinct stages of human figure drawings, during development. These stages are empirically well established and used in Study I as the major measure of children's drawing development. In the whole group of 301 drawings (preterm and term), the earlier established types of drawings; scribbles, pre-tadpoles, tadpoles, transitional and conventional forms were observed. This variation from very immature to advanced drawings in this group of 5½ year old children enabled us to investigate underlying perceptual-motor skills in drawing development.

Goodenough and Harris (Harris 1963) showed that symmetry and proportion of human body parts are important parameters in drawing development. This was also found in our study, proportion and circularity together with visual perception explained 60% of the variance in development of human figure drawing. Symmetry also improved when children transferred to the next stage of drawing but it was not a significant predictor for drawing development. Size of the head in relation to body decreased and reached a realistic proportion when drawing conventional forms of drawings. This is consistent with the literature describing that drawings at early stages reflect what children know, whereas, in later stages, they reflect what they see (Cox 1993). Piaget described this as a transition from intellectual realism to visual realism (Piaget and Inhelder 1948; Cox 1993). Circles got more accurate (smooth lines without gaps or overlaps) in later stages of human figure drawings showing the maturation of motor control. Results confirmed that using simple classification of developmental stages of human figure drawing is a good reflection of developing visual perception and visual motor coordination.

Boys have in general a slower development of drawing skill than girls independent of whether they are born preterm or at term (Brown 1990; Coulbeau, Royer et al. 2008). The development of visual perception and visual-motor coordination has been shown to be affected particularly in preterm boys (Geldof,

Van Wassenaer et al. 2012). We found that drawing skill is affected in preterm boys. Preterm boys showed a risk for poor visual-motor planning (relative size of body parts), poor visual-motor control (circle drawing) along with poor visual perception. They were also found to have immature pencil grip, more so than others. Immature pencil grip is an indication for problem in fine motor and somatosensory functions, which can lead to many problems in acquisition of fine motor skills including handwriting. Indeed, preterm children have been shown to have problems in handwriting legibility and speed. Even so, it has to be recognized that the majority of the preterm boys showed age expected drawings and pencil grip positions.

Visual perception especially for line orientation has been found to be affected in preterm children (Geldof, Van Wassenaer et al. 2012). This situation can influence especially learning handwriting, math, and reading skills, where the perception of line orientation is essential to discriminate different letters, numbers and symbols. For example, letters 'b' and 'd' have similar graphic patterns but line orientation is different, which can probably create a discrimination problem in preterm children. Since graphomotor ability reflects great deal of visual perception it can explain why copying geometric/abstract patterns have been associated with learning difficulties (Weil and Amundson 1994; Daly, Kelley et al. 2003; Barnhardt, Borsting et al. 2005). On the other hand, this link between garphomotor ability and learning difficulties can also be explained by problems in fine motor and executive functions (Bohm et al. 2010). Graphomotor ability (VMI test) was associated with learning difficulties even after controlling for intelligence (Taylor Kulp 1999). Apparently as it was found in our study, human figure drawing provides considerable amount of information about cognitive (Harris 1963; Abell, Wood et al. 2001; Schepers, Dekovic et al. 2012) and motor development (Schepers, Dekovic et al. 2012).

Drawing of a human figure, in spite of its simplicity as a task, bares considerable information about motor and cognitive development in preschool age. Even though developmental stages of human figure drawing have been well established empirically, underlying visual perceptual and visual motor parameters have not been investigated systematically. These underlying parameters give us a better grasp about how drawing skill is related to broad indices like PIQ and M-ABC indicating that our results provide valuable information about children's motor and cognitive performance.

5.2. CHILDREN WITH UNILATERAL CEREBRAL PALSY

Children with unilateral CP showed significantly inferior performance in visual motor coordination along with sensory, perceptual, motor and cognitive functions compared with those of their typically developing peers. Problem in visual motor coordination was consistent at several different subtests involving drawing and visuo-constructional tasks (e.g. subtest reproduction of two-dimensional model), and that these subtests were correlated with handwriting quality parameters.

Consequently, handwriting quality was also found to be lower in CP group at all measured quality categories (legibility, form, alignment, size and spacing) than in control group of typically developing children. Most of the children in the control group reached the ceiling of handwriting quality parameters. The link between the development of visual-motor coordination and learning handwriting skill is clearly established in typically developing children (Graham and Weintraub 1996).

In Study II we found that children with unilateral CP have poor kinaesthesia on both sides of the body, not just on the affected side, and visual discrimination of graphical representations (i.e. subtest overlapping figures), and that there was a correlation between these sensory modalities and quality parameters of handwriting in CP group. The poor integration of kinaesthetic and visual information is in agreement with one earlier study in children with CP (Ritterband-Rosenbaum, Christensen et al. 2011). It can also be explained such that the motor system can have difficulties to integrate information from muscles/joints about movement and the visual system processing graphical pattern that is to be copied. This integrative processing was found not only for handwriting but also other visual motor tasks, such as a peg task or motor response speed to a visual stimulus, in the non-involved, dominant hand in children with unilateral CP. We also found reduction in tactile sense in both hands. This information of decreased function even in the non-involved hand is of clinical importance. The appearance of subtle problems has been earlier described for example for fingertip force control problem in non-involved hand (Gordon, Charles et al. 1999).

Speed of handwriting is expected to increase by age in typically developing children (Graham and Weintraub 1996) but studies investigated the relationship between speed and quality of handwriting have shown rather inconclusive results (Ziviani 1984; Graham and Weintraub 1996; Graham, Berninger et al. 1998; Karlsdottir and Stefansson 2002). For children with unilateral CP, teachers and parents have reported difficulty in maintaining neatness at speed or over long periods of time (DuBois, Klemm et al. 2004). Our results indicate a similar tendency in the relationship of quality and speed. In Study II, children with CP produced letters at the same speed as their peers whereas the quality of handwriting was significantly lower in this group. This probably indicates a cost of reduced quality when adapting the speed of writing according to the needs of a classroom setting. Although, both speed and quality of handwriting increased in 16-months follow-up, the children with unilateral CP may not keep up the speed or quality of their handwriting while writing for longer durations, which could result a problem in academic achievement.

The improved ability when examining the children with unilateral CP again after 16 months (Study III) is interesting. They have yielded improvement of both

quality and the speed of handwriting. However this was later than expected development, the youngest child was about to finish the second grade at school, an age when typically developing children have reached the ceiling effect (i.e in Minnesota Handwriting Assessment) and the others were even older. The results indicated a prolonged learning phase. Similar prolonged learning period has also been reported for self care ability in daily life even in children with quite good manual ability (Ohrvall, Eliasson et al. 2010).

Measurement of handwriting quality was based on copying one sentence accurately in terms of visual parameters. Relationship between handwriting quality and cognitive functions were found in selected items of a cognitive test in Study II. In the follow-up study, this relationship was further investigated with a better cognitive index with normative data of Turkish children. Children with FSIQ > 70 had better handwriting quality than children with FSIQ < 70 in both previous and present assessments. Both age and FSIQ predicted the rate of development in handwriting quality in children between 8-13 years old. Regression models showed that children in younger ages and children with lower IQ made greater improvement based on those children, who have been healthy during the examined period.

Children with unilateral CP have shown to have a stable course of development in intellectual and academic indices (Ballantyne, Spilkin et al. 2008). In agreement with this all participants in Study III passed to the next grade at school. However, presence of seizures is known to limit cognitive development (Vargha-Khadem, Isaacs et al. 1992; Cioni, Sales et al. 1999; Kolk and Talvik 2000; Singhi, Jagirdar et al. 2003; Sigurdardottir, Eiriksdottir et al. 2008). The four children in this study who showed regression in handwriting quality either had school leave due to orthopaedic surgery or suffered severe epileptic seizures reported by their parents. Obviously, the presence of seizures and lack of practice during school leave interfered with the development of handwriting skill in this group of children. In those studies (II and III) we had only included children with left-sided hemiplegia. The reason was to minimize the influence of brain lesions in the hemisphere typically controlling the hand used for handwriting.

It seems that children with unilateral CP have a prolonged learning phase in tasks that require visual motor integration. The rate of learning was dependent on both chronological age and IQ.

5.3. CHILDREN WITH CHILDHOOD APRAXIA SPEECH

Children with suspected CAS in Study IV showed clear impairment in speech and non-speech oral motor functions according to the VMPAC norms. The results support the clinical diagnoses identifying clear impairment in sequencing speech movements, similar to earlier studies of CAS (Dewey, Roy et al. 1988; Bradford and Dodd 1994; Williams and Stackhouse 2000; Highman, Hennessey et al.

2008). It is also in agreement with diagnostic criteria clarified by ASHA (2007). Manual and overall motor functions and adaptive behaviour were also affected but to a lesser degree. Correlations were only found between drawing geometrical shapes and production of speech and non-speech oral movements. Although correlation between oral/verbal motor ability and other motor and adaptive abilities were weak in general, there was a high incidence of co-occurring motor and also behavioural problems on an individual level.

The co-occurrence of manual and oral/verbal motor problems has been recognized in clinic and in research for a long time. The research question has been whether a common motor deficit is underlying both the manual and the oral-motor problems. Co-morbid motor coordination difficulties were earlier reported to occur in 40-90% of children with any type of speech/language disorders (Hill 2001). Common underpinnings between these two types of developmental difficulties have been suggested as rule generalization (Hill 1998; Hill 2001), praxis (Hodge 1998), cerebellar deficits (Estil et al. 2003), inter-hemispheric deficits (Estil et al. 2003) or a more diffuse atypical brain development (Kaplan 1998).

Co-occurrence of speech and body motor problems is also found in children with DCD (Rodger, Watter et al. 2007). However, within all motor subtests, only fine-motor integration was correlated with speech/non speech motor subtests, i.e. between visual motor integration in drawing and the single production of non-speech and speech movements. Earlier studies have found a similar relationship in children 2 - 6 years of age with suspected CAS (Newmeyer, Grether et al. 2007), supporting our result.

When using ICF-CY, parents of children with CAS have reported problems in 'learning to write' and 'fine hand use' (Teverovsky, Bickel et al. 2009). In this study, children with CAS also show below average adaptive behaviour. In fact, there was no simple relationship between adaptive behaviour and speech/non-speech oral motor function, only one moderate correlation was found between subtest of ABAS-2 and VMPAC. ABAS-2 is based on parents' reports, and parents may develop adaptive listening and alternative ways of communication explaining no reported problem in 'home living' in spite of low performance in fine motor skills. Children's adaptive behaviour might have been reported differently by other observers, such as a teacher reporting on behaviour in a school setting. Speech and language impairment in childhood is known to relate to lower educational attainment and occupational status in young adults (Johnson, Beitchman et al. 2010; Elbro, Dalby et al. 2011).

The few and in general low correlations between subtests are interesting to discuss in relation to the individual performances. In spite of low correlation the co-occurrence of speech/non-speech and manual motor problems was commonly found on an individual level. Many children have problems in several subtests, up to seven of the eight subtests when using '-1 SD' as the cut-off point. This cut off

is equal to 16th percentile and corresponds to the common cut-off score for identifying the risk for DCD (Visser 2003). When looking at the pattern of co-occurrence to adaptive behaviour, several children with a lot of speech/non-speech and manual motor problems show minor problems in adaptive behaviour. This indicates that the co-occurrence of problems is not explained by a simple relationship. However, multiplicity of motor problems might affect the child's daily living to a greater extent than a single motor problem. The relationship between motor impairment and its consequences on daily life has to be further investigated.

5.4. METHODOLOGICAL CONSIDERATIONS

In general, there can always be discussion about sample size, and Study III and IV would of course benefit from a bigger cohort. In Study III the number of children is a limitation for the generalizability of the regression model; however, from a statistical perspective we found the result acceptable. There was a moderate deviation from normality in the regression model but since linear regression is a strong parametric test and the transformed results were not different from the untransformed, we believe that the result is acceptable. In addition, one child showed a Cook's distance value (D) > 4/n-k-1 since he was the youngest individual and made the greatest improvement. The model was still significant without this outlier. However, we kept this child in the model since he represents this age in our sample.

Statistical issues can of course be raised especially from Study II and IV since many correlations were run on one set of data. Alpha level was not corrected for the risk of type 1 error since such correction would increase the risk for type 2 errors due to small sample size in these studies. Therefore, significance level was set to $p<0.05$ for correlation coefficient. Power analyses were performed for significant correlations in Study IV to provide a better interpretation of results. In addition, findings were supported from earlier findings.

A new method for analysis of human figure drawings, the ImageJ program, was used in Study I. It is widely used to analyse scientific images but is to our knowledge, used for the first time to analyse drawings. Therefore, we did not have the possibility to compare our findings with other studies. A methodological limitation can be the circularity measurement, which gives a full-score if there is a perfect circle. In drawing, a human head does not need to be represented as a perfect circle, but we believe that circularity is a valid measure for smoothness of circular lines since the score decreases in cases of angular shapes, overlaps, and gaps.

5.5. CLINICAL IMPLICATIONS AND FUTURE RESEARCH PERSPECTIVES

Children born preterm have a risk for delayed visual-motor coordination. Most children enjoy drawing and; it is one of the major activities in day care and preschool settings. Preschool teachers and others commonly use this simple categorization of developmental stages in human figure drawing task. Our results support that this simple categorization might be beneficial for early detection of perceptual-motor difficulties in preterm children at preschool age. Early detection can of course facilitate early intervention for children with somewhat mild motor and cognitive problems.

Children with unilateral CP have handwriting difficulties although they use the non-involved hand for writing task. Since the handwriting was explained to great extent by cognition, children with CP probably show limited skills in spelling and composition of creative and well-structured text (Christensen 2005). Therefore, they might fail to make the expected academic progress associated with their chronological age. Indeed, low educational attainment has been shown for individuals with CP (Bottos, Feliciangeli et al. 2001). On the other hand, cognitive deficit and learning difficulties in school age children with spastic CP are not merely a function of an early brain lesion; they are also the result of interaction between the child and his or her environment, where the child participates in learning situations and interacts with his or her peers. Many children with writing difficulties may experience a complex cycle of frustration, loss of self-esteem and motivation (McKinlay 1978) and consequently fall further behind in their academic achievement.

For drawing and handwriting it was pretty clear that children born preterm and with CP have delayed learning. This situation of course affects their inclusion to mainstream education and consequently participation to school setting. Assistive technology or simply keyboarding can have positive effects on these aspects. For the development of cognitive functions further research should be done in order to see how using technology would affect their working memory and composition writing (Torrance, Van Waes et al. 2007). Considering that language skills are better in these populations (VIQ is usually higher than PIQ) when mechanical limitations on writing are ruled out, children can show better skills in composition writing. Some studies in typically developing children suggest an increase in text production when using keyboarding but it has not been suggested before third grade in school (Graham, S. and N. Weintraub 1996). Similarly for visuo-spatial skills computer programs can decrease the effect of motor limitation. However, especially kinaesthetic perception will be lacking for understanding the form of graphical patterns and how this would affect perceptual processes is not known. It is possible that learning and memory could be affected for recognition and retrieval of graphical patterns. Since involvement of more sensory modalities strengthens and probably fastens the learning period, a draw back in learning and

memory would be expected. Even so, assistive technology can provide promising solutions therefore should be investigated more in children with neurological dysfunctions.

Children with CAS undergo a broader motor examination and follow-up for adaptive behaviour in the clinic. Clinicians should be aware of co-occurrence between motor dysfunctions in different domains. This situation requires a team examination in a child with motor disorders. Some dysfunctions require an immediate intense intervention while probably others benefit of follow-ups. Especially children with multiple problems need further investigation and support in their daily life and in school whereas those with fewer areas of difficulty would benefit from follow-ups throughout the school age years.

6.   ACKNOWLEDGEMENTS

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